Evaluation of Outcome Measures for Adolescents with Idiopathic Scoliosis Using a Mixed-Method Approach

by

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Abstract

Adolescent idiopathic scoliosis (AIS) is a common spinal deformity among the paediatric population, affecting their physical, mental, and social well-being. Approximately 10% of those affected require interventions, including surgery. Studies on surgical fusion in AIS present conflicting findings of postoperative back pain, functional limitation, and the factors influencing these outcomes. While various Outcome Measures (OMs) are used to evaluate the outcomes of AIS interventions, there is an important gap in the comprehensive evaluation of their measurement properties. The reference standard patient-reported outcome measure (PROM) for individuals with AIS is the Scoliosis Research Society (SRS) questionnaire. However, a qualitative assessment of its appropriateness for this specific population has not been conducted previously. Therefore, this thesis presents five studies to explore the current use of OMs) for individuals with AIS, and to evaluate their measurement properties. In *Chapter Two,* a retrospective longitudinal cohort study was conducted to analyse functional outcomes in individuals with AIS using the SRS-22r questionnaire. The study compared outcomes between those who achieved the smallest detectable change (SDC) in SRS-22r and those who did not, while also identifying predictors (e.g., age) of achieving these scores. Overall, most of the SRS-22r domains reach or exceed the SDC at 1 year except function and pain scores. Findings suggest a shortcoming in the SRS-22r questionnaire which prompted a subsequent systematic review in *Chapter Three*. The systematic review aimed to identify current OMs used to evaluate Physical Functioning (PF) in individuals with AIS. Review findings revealed insufficient measurement properties, particularly in terms of content validity, for the identified PROMs based on Consensus-based Standards for the selection of Health Measurement Instruments (COSMIN) guidelines. Furthermore, only one performance-based outcome measure (PBOM) their measurement properties evaluated among individuals with AIS, indicating limited use. *Chapters Four* and *Five*

aimed to evaluate the content validity of the frequently used questionnaire for this population i.e. the SRS-22r. A qualitative study design was employed in *Chapter Four* to assess the content validity of the SRS-22r through cognitive debriefings with healthcare professionals (HCPs) and individuals with AIS. The questionnaire's comprehension, comprehensiveness, and relevance to individuals with AIS revealed numerous issues, highlighting the need for a concept elicitation study and refinement of SRS-22r questions and response options. Refinements to some of the SRS-22r questions and response options were proposed by participants. Further testing with another round of cognitive debriefing interviews is still needed to further refine the SRS-22r questionnaire. *Chapter Five* reports a qualitative study which was conducted to identify the most important concepts related to Health-Related Quality of Life (HRQOL) in individuals with AIS, building upon the findings from *Chapter Four*. Findings highlighted that the AIS has a broad impact on HRQOL of individuals with AIS, encompassing physical, activity, psychological and social effects. Comparing these themes with the contents of the SRS-22r, a limited matching was found indicating that the SRS-22r is not comprehensive enough to fully assess HRQOL of individuals with AIS. Collectively, these studies provide critical evidence underpinning the rationale to support the development of a new, age- and language-appropriate PROM that accurately captures the experiences of individuals with AIS. In *Chapter Six* the perspectives of HCPs on the use of PROMs and PBOM among individuals with AIS were explored through qualitative interviews. To address the limited utilisation of these measures in practice; previously identified in the in *Chapter Three*. Many barriers (e.g., inadequate PROMs), and facilitators (e.g., quality existing measure) were identified for the use of PROMs and PBOMs among individuals with AIS. Overall, this thesis highlights the importance of using population specific and appropriate OMs to evaluate HRQOL in individuals with AIS. It emphasises the need for valid and comprehensive measures being language and age-specific questionnaires, which are more relevant and meaningful to the unique experience of individuals with AIS.

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List of Papers and Conference Abstracts

During the course of this doctoral program in the School of Sport, Exercise, and Rehabilitation Sciences, several papers directly related to the thesis have been published and/or presented at conferences. Further studies are currently under review at the time of thesis submission. Consequently, certain parts of this thesis contain text that is identical to the published work. The sections of the thesis that were derived from the published work will resemble the original work in terms of structure and content, with additional modifications as necessary to support the overall argument of the thesis. Each relevant chapter will commence with a summary of the paper and its utilisation in the thesis.

Published papers

Alamrani S, Gardner, A., Rushton, A.B., Falla, D. and Heneghan, N.R., 2023. Predictors of Relevant Changes in Pain and Function for Adolescents with Idiopathic Scoliosis following Surgery. *Spine*. **(Appendix 1)**

Alamrani S. Gardner, A., Falla, D., Russell, E., Rushton, A. B., & Heneghan, N. R. (2023). Content validity of the Scoliosis Research Society questionnaire (SRS-22r): A qualitative concept elicitation study. PloS one, 18(5), e0285538. https://doi.org/10.1371/journal.pone.0285538. **(Appendix 2)**

Alamrani, S., Gardner, A., Falla, D., Russell, E., Rushton, A.B. and Heneghan, N.R., (2021). Content validity of Scoliosis Research Society questionnaire-22 revised (SRS-22r) for adolescents with idiopathic scoliosis: protocol for a qualitative study exploring patient's and practitioner's perspectives. *BMJ open*, *11*(12), p.e053911. **(Appendix 3)**

Alamrani, S., Rushton, A.B., Gardner, A., Bini, E., Falla, D. and Heneghan, N.R., (2021). Physical functioning in adolescents with idiopathic scoliosis: a systematic review of outcome measures and their measurement properties. *Spine*, *46*(18), pp.E985-E997. (**Appendix 4**)

Alamrani, S., Rushton, A., Gardner, A., Falla, D. and Heneghan, N.R., (2020). Outcome measures evaluating physical functioning and their measurement properties in adolescent idiopathic scoliosis: a protocol for a systematic review. *BMJ open*, *10*(4), p.e034286. **(Appendix 5)**

Papers Under Review

Alamrani S., Gardner A, Falla D. Russell E, Rushton AB, Heneghan NR. Content validity of the Scoliosis Research Society-22 revised (SRS-22r): A qualitative cognitive debriefing study (Under review - PloS one Journal).

Alamrani S., Gardner A, Falla D. Russell E, Rushton AB, Heneghan NR. The use of outcome measures by healthcare professionals for young people with adolescent idiopathic scoliosis: a qualitative exploration. (Under review – BMC health service research journal).

Conference presentations

Alamrani, S., Rushton, A. B., Gardner, A., Bini, E., Falla, D., & Heneghan, N. R.(2020). Physical functioning in adolescents with Idiopathic Scoliosis: A Systematic Review of Outcome Measures and Their Measurement Properties. World Physiotherapy Congress online. 9-11th April 2021. (Platform presentation).

Alamrani, S., Rushton, A. B., Gardner, A., Bini, E., Falla, D., & Heneghan, N. R. (2020). Measurement properties of Outcome measures evaluating Physical functioning in adolescents with idiopathic scoliosis : A systematic review. 4th Saudi Spine Society annual conference. 7-9th November 2020. (Platform presentation).

Alamrani, S., Rushton, A. B., Gardner, A., Bini, E., Falla, D., & Heneghan, N. R. (2020). Measurement properties of Outcome measures evaluating Physical functioning in adolescents with idiopathic scoliosis : A systematic review. Physiotherapy UK 13-14th November 2020. Birmingham. United Kingdom. (Poster presentation).

Alamrani S., Gardner A, Falla D. Russell E, Rushton AB, Heneghan NR. The use of outcome measures by healthcare professionals for young people with adolescent idiopathic scoliosis: a qualitative exploration. World Physiotherapy Congress online. 2-4th June 2023. Dubai. United Arab of Emirates. (Poster presentation).

Alamrani S, Gardner A, Rushton AB, Falla D. Heneghan NR. Predictive factors of clinically relevant changes in pain and function following surgery for adolescents with idiopathic scoliosis. Submitted to Physiotherapy UK- 1st November 2023. Birmingham. United Kingdom. (Poster presentation- Accepted).

Alamrani S., Gardner A, Falla D. Russell E, Rushton AB, Heneghan NR. Content Validity of The Scoliosis Research Society (Srs22r): A Qualitative Concept Elicitation Study. Physiotherapy UK- 1st November 2023. Birmingham. United Kingdom. (Poster presentation- Accepted).

Alamrani S., Gardner A, Falla D. Russell E, Rushton AB, Heneghan NR. Content Validity of The Scoliosis Research Society (Srs22r): Qualitative Cognitive Debriefings Study. Physiotherapy UK- 1st November 2023. Birmingham. United Kingdom. (Poster Presentation-Accepted).

Related publications

Tucker, S., Heneghan, N.R., Gardner, A., Rushton, A., **Alamrani, S.** and Soundy, A., 2023. Factors Influencing Participation in Physical Activity, Sports, and Exercise in Children and Adolescents with Spinal Pain or Spinal Conditions: A Systematic Review and Meta-Ethnography. Behavioral Sciences, 13(6), p.486.

Tucker, S., Heneghan, N.R., **Alamrani, S.,** Rushton, A., Gardner, A. and Soundy, A., 2023. Barriers and facilitators of physical function, activity, sports and exercise in children and adolescents with spinal pain: a protocol for a systematic review and meta-ethnography. *BMJ open*, *13*(3), p.e063946

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AIS	Adolescent idiopathic scoliosis
BSR	British Spine Registry
CI	Confidence Intervals
COREQ	COnsolidaed criteria for REporting Qualitative studies
COMET	Core Outcome Measures in Effectiveness Trials
COS	Core Outcome Set
COSMIN	Consensus-based Standards for the selection of Health Measurement Instruments
EThOS	Electronic Thesis Online Service
GRADE	Grading of Recommendations, Assessment, Development and Evaluation
HCPs	Health Care professionals
HRQOL	Health Related Quality of Life
IQR	Interquartile Range
MCID	Minimal Clinical Detectable Change
MCAR	Missing Completely At Random
NHS	National Health Service
OM/s	Outcome Measure/s
OR	Odds Ratios
PBOM/s	Performance Based Outcome Measure/s
PF	Physical functioning
PIS	Participant Information Sheet
PROM/s	Patient Reported Outcome Measure/s
PPI	patient and public involvement
QoL	Quality of Life
SDC	Small Detectable Change
SD	Standard Deviation
SRS	Scoliosis Research Society
SRS-22r	Scoliosis Research Society 22 revised
STROBE	Strengthening the Reporting of Observational studies in Epidemiology
SOSORT	Scoliosis Orthopaedic and Rehabilitation Treatment
UK	United Kingdom

Chapter One

INTRODUCTION

1.1 Scoliosis

Scoliosis is a three-dimensional deformity of the spine and trunk (Negrini et al., 2012). It involves lateral curvature and rotation of spinal vertebrae leading to a twisted appearance of the spine and torso (Reamy and Slakey, 2001). This is referred to as a "structural scoliosis", distinguishing it from a "functional scoliosis"; develops from non-structural causes such as muscle imbalances or leg length discrepancies. It is temporary, or even reversable once the underlying cause is treated (Negrini et al., 2018). This thesis is focused solely to structural scoliosis and all reference here on to scoliosis is that of a structural deformity.

The Scoliosis Research Society (SRS) suggests that a diagnosis of scoliosis is confirmed when the Cobb[1] angle is >10° accompanied with vertebral rotation (Negrini et al., 2018). (*See Figure 1.1.*). The severity of the scoliosis is determined by the magnitude of the Cobb angle (Cobb, 1948). A Cobb angle up to 20° indicates a mild scoliosis, 21° to 35° moderate scoliosis, and 36°- 40° moderate to severe scoliosis, and > 41° severe scoliosis (Negrini et al., 2018). The anatomical location of the Cobb angle using radiological images in frontal plane is used to classify scoliosis into four major

[1] Dr. Cobb invested a standardise method for measuring scoliosis curvature in 1940s. It is calculated by drawing lines perpendicular to the upper and lower endplates of the two most tilted vertebrae in a scoliotic curve. The angle formed by these two lines is measured and shows the degree of spinal curvature COBB, J. 1948. Outline for the study of scoliosis. *Instr Course Lect AAOS,* 5**,** 261-275. (See Figure 1.1.)

types thoracic, lumbar, thoraco-lumbar and S- shaped; it is the oldest classification system used for both conservative and pre-operative management decisions (Newton, 1996).

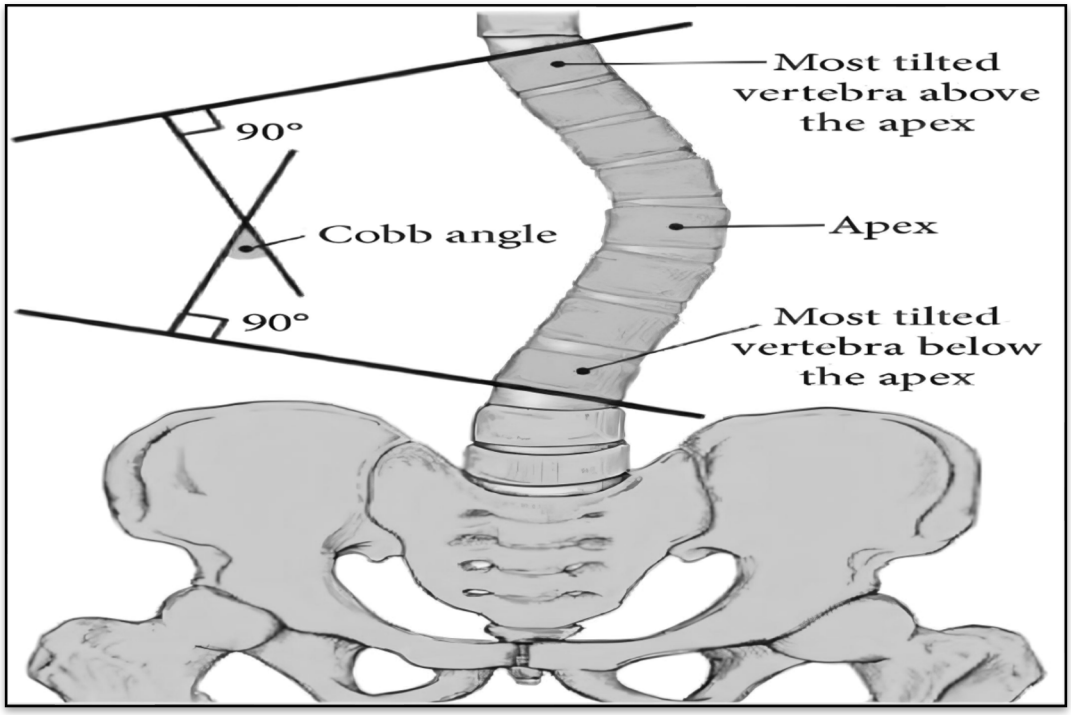


Figure 1.1: The measurement of the curve angle using Cobb method 1 . Figure from (Horng et al., 2019).

Scoliosis can be classified according to its aetiology into two main types: idiopathic and non-idiopathic, with the latter arising from a pathological cause i.e. congenital, neuromuscular, or mesenchymal, and idiopathic scoliosis being of unknown pathology (Konieczny et al., 2013). Kleinberg proposed the term idiopathic scoliosis in 1922, meaning that the aetiology is unclear and there is no identifiable reason for the resultant spine deformity (Kleinberg, 1922). Approximately, 80% of scoliosis cases are considered idiopathic, and the remaining 20% are non-idiopathic (Horne et al., 2014, Konieczny et al., 2013). Idiopathic scoliosis can manifest at distinct stages of development. If it develops between birth and 3 years of age, it is referred to as infantile idiopathic scoliosis, with juvenile idiopathic scoliosis and adolescent idiopathic scoliosis evident in those

between 4 and 10 years, and 10 and 16 years, respectively (Altaf et al., 2013). This thesis focuses on Adolescent Idiopathic Scoliosis (AIS). The term "Individual with AIS" will be used throughout this thesis to refer to those individuals who have been diagnosed with AIS.

Whilst AIS has no clear aetiology (Altaf et al., 2013) it is nonetheless a complex disease with hereditary predisposition elements (Burwell, 2003, Lowe et al., 2000). Other predisposing factors pertain to hormones (melatonin, oestrogen, and growth hormone), abnormal biomechanical pressures, neurophysiologic predisposition, connective tissue abnormalities, muscle structure, and vestibular function (Ahn et al., 2002, Kouwenhoven and Castelein, 2008). AIS usually begins during puberty, occurring between the ages of 11 to 14 years, the fastest period for spine curvature progression (Grivas et al., 2006). Whilst the incidence of having AIS is almost equal in male and female, females are reported to have 10-time greater risk of curve progression (Reamy and Slakey, 2001, Horne et al., 2014). When the Cobb angle is 10° to 20°, the ratio of affected females to males is comparable 1.3:1, increasing to 5.4:1 for Cobb angles between 20° and 30°, and 7 to 10 :1 for Cobb angles > 30° (Parent et al., 2005, Weinstein et al., 2008). The prevalence of AIS with a Cobb angle >10° in the general population varies widely from 0.13 to 12 % (Lenssinck et al., 2005, AlAssiri et al., 2022, Du et al., 2016b, Huang et al., 2011, Singh et al., 2022), whilst a ratio of 2 to 3% is the frequently reported rate in the literature (Negrini et al., 2018).

1.2 Clinical characteristics of AIS

AIS is associated with specific clinical characteristics that are often noticed by the patient, a family member, a medical practitioner, or a school nurse (Altaf et al., 2013). These characteristics are mainly aesthetic, and it may vary in its presentation depending on the degree of curve; mild scoliosis is not obvious and asymptomatic (Weinstein et al., 2008) . However, as the curve progress it becomes more visible and may associated with other symptoms such as back pain (Monticone et al., 2014). The aesthetic changes are of the most concern to individuals with AIS, which often lead to emotional and psychological distress (Motyer et al., 2021a, Aroeira et al., 2016). It includes asymmetry at the shoulder and waistline or (hips), a 'rib hump,' (i.e. an elevation of one side of the back) (Negrini et al., 2018), (*See Figure* 1.2). "Adam's forward bend test" (*See Figure* 1.2 B) is a widely used screening test used to identify the presence of rib hump during forward bending (Senkoylu et al., 2021). Unlike structural scoliosis, the rib hump/curve disappears in those with functional scoliosis (Altaf et al., 2013).

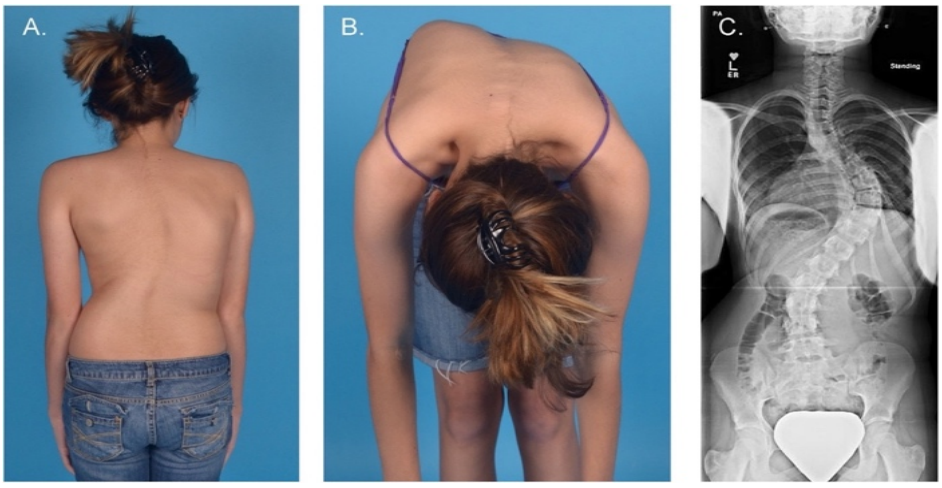
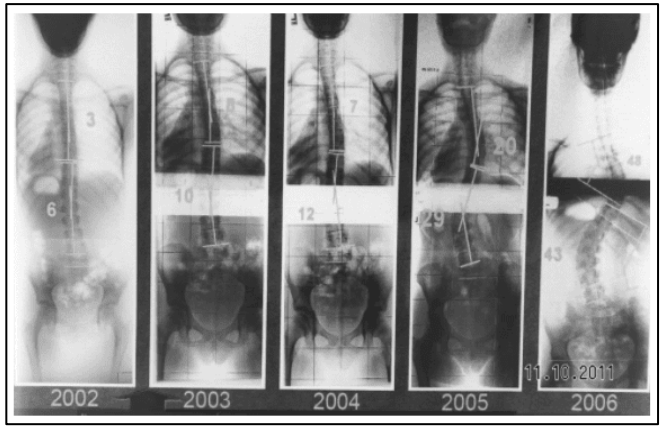


Figure 1.2: A picture of an individual with AIS, A) Standing back view, (B) forward-bending view, and (C) standing posterior anterior radiograph. Figure from (Paria and Wise, 2015).

1.3 Management of AIS

The management of AIS aims to slow or stop the progression of the curvature, and reduce the possible long-term complications of the deformity (Bettany-Saltikov et al., 2016). Curve progression is the increase in the size of the curve by > 5° at consecutive follow-up appointments of between 4 to 6 months (Altaf et al., 2013). Approximately 10% of individuals with AIS will see their spine curve progress (Horne et al., 2014). Risk factors for curve progression include being female, skeletally immature, and exhibiting a large curve at first diagnosis (Reamy and Slakey, 2001). Individuals with a Cobb angle >45º are more likely to see curve progress during their growth spurt period, whereas those with curves >50 º who are at or nearing full growth may see slow progression over time (Wong et al., 2022). Nearly 10% of individuals with AIS will require some form of conservative management and up to 0.1% will eventually require operative management (Bettany-Saltikov et al., 2016). Table 1.1 presents the indications and brief descriptions of each management approach.

Table 1.1: Management approaches for individuals with AIS			
Management approaches	**Indications**	**Descriptions**	**Examples**
Observation	▪ Cobb angle <25°	▪ To monitor curve progression ▪ It involves regular clinical assessment at specialist clinic every 2 to 3 months up to 36 to 60 months (Altaf et al., 2013). ▪ Radiographs performed during alternate clinical visits (*see Figure* 1.3) is recommended, to reduce the risk of excessive exposure to ionisation through X-ray, or use low-radiation or radiation-free methods (Luan et al., 2020).	 **Figure 1.3 :X-rays taken at regular intervals for an untreated individual with AIS.** Figure from (https://www.hudsonvalleyscoliosis.com/what-is-scoliosis/mild/)

<table>
<tr>
<td>Brace</td>
<td>

Cobb angle > 25°-30°

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To avoid the need of surgery
It involves wearing the brace e.g. Boston brace (see Figure 1.4) until skeletal maturity (i.e. recorded height growth ceased for 6 months) (Grivas et al., 2022, Kaelin, 2020).
The International Society on Scoliosis Orthopaedic and Rehabilitation Treatment (SOSORT) recommending wearing brace as follow:

Night-time Rigid Bracing: 8–12 h per day (in bed)
Soft Bracing (e.g., Spine Cor brace): 20 hour per day
Part Time Rigid Bracing: 12 to 20 hour per day (outside school and in bed)

Full Time Rigid Bracing /Cast: All the time (at school, at home and in bed)

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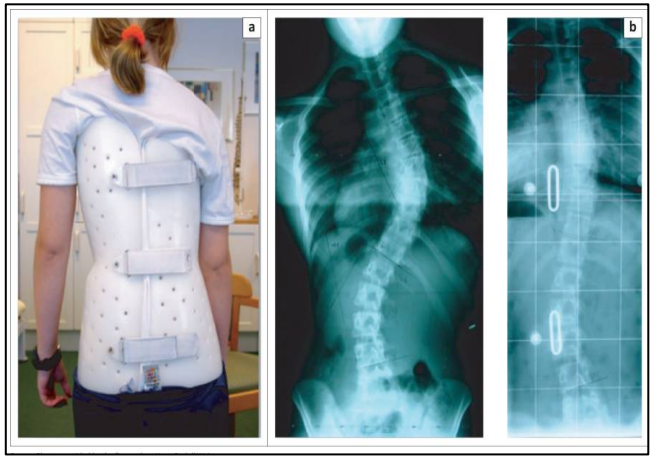

Figure 1.4: (a) skeletally immature individual wearing Boston brace, (b) In-brace correction of the spine curve.
Figure from (Weiss et al., 2021)
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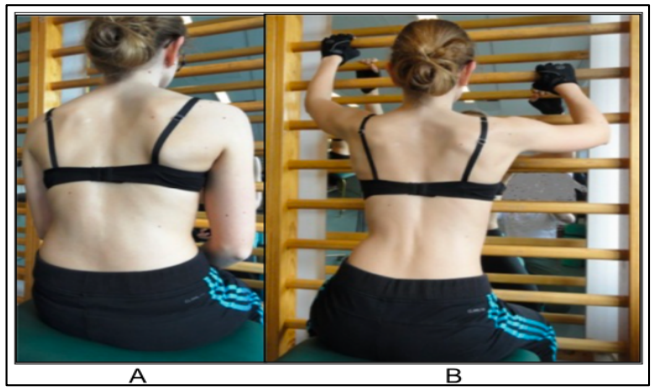
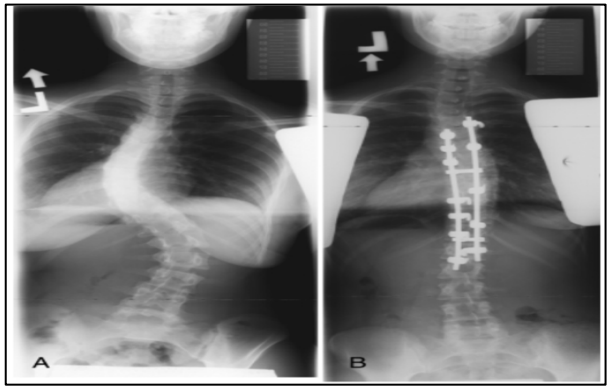
Scoliosis specific exercise	▪ Cobb angle >15°, ▪ For Individuals wearing brace	▪ Specific type of exercises to correct spine curvature e.g., Schroth method, (*See Figure* 1.5). ▪ It includes auto-correction in 3D, training in activities of daily living, stabilizing the corrected posture and patient education. ▪ The recommended sessions are from 2 to 7 exercise sessions per week depending on the exercise technique, patient cooperation, and motivation (Negrini et al., 2018).	 **Figure 1.5: (A) The Schroth exercise for individuals with AIS sits on a Swiss ball in front of a mirror and (B) performs active 3D auto self- correction using the wall bar.** Figure from (Berdishevsky et al., 2016)

Surgery	▪ Cobb angle 45° - 50° in skeletally immature individuals, ▪ For cosmetic reasons, ▪ Those with severe symptoms (e.g., back pain)	▪ Approximately 0.1 to 0.3% of individuals with AIS need surgery (Weiss et al., 2008). ▪ Posterior spinal fusion is the gold standard approach, by opening of the vertebrae at the deformity level, inserting of pedicle screws, and connecting of two metal rods to these screws. (*See Figure* 1.6.) ▪ Anterior spinal fusion approach is used for some cases to enhance rigid curve flexibility (Jada et al., 2017)	 **Figure 1.6: Individual with AIS require surgery (A) and (B) corrected spine curvature with rods and screws.** Figure From (Janicki and Alman, 2007)

1.4 Outcome measures

Measuring outcome is crucial for improving the quality of care, promoting evidence-based practice, ensuring patient-cantered care, evaluating cost-effectiveness, and fostering research and innovation, which ultimately contributes to better health outcomes for individuals (Porter, 2010, Kuhn, 2016). The Core Outcome Set (COS) is an important approach that has been developed to standardise the set of outcomes that should be measured and reported in clinical research studies for a specific health condition (Dodd et al., 2018). The aim of this approach is to promote consistency, comparability, and transparency in reporting outcomes across studies, facilitating evidence synthesis (Prinsen et al., 2016). The previous documentation of a COS for adolescent and young adults with spinal deformity has limitations in its applicability specifically to individuals with AIS as it (de Kleuver et al., 2017) as it encompassed all patients undergoing surgery for spinal deformity, leading to heterogeneity that restricts its relevance to individuals with AIS. Additionally, the targeted AIS population were not involved in the COS development, as recommended by COS developers (James et al., 2018). Furthermore, neither the targeted group nor professionals involved in the Delphi study were English speakers, raising concerns about the applicability of the findings to English-speaking populations.

When developing a COS, the International Classification of Functioning, Disability, and Health (ICF) can play a valuable role. It is a framework developed by the World Health Organization (WHO) and provides a universal comprehensive understanding of health and functioning (WHO, 2001). The ICF emphasises the biopsychosocial perspective, considering not only an individual's health condition but also the impact on their functioning in various domains of life (WHO, 2001). The COS using the ICF has been used widely among different population such as people with non-specific low back pain (Chiarotto et al., 2015), individuals with cerebral palsy (Almoajil et al., 2022)

and young people with spinal deformity (de Kleuver et al., 2017).The ICF for Children and Youth (ICF-CY) is an adaption to ICF specifically tailored to children and youth (up to the age of 18) (WHO, 2007). It considers the unique developmental characteristics and needs of children, including their emerging capacities and the influence of their family and educational environments (WHO, 2007). The ICF-CY contains four main components: body function and structure, activities and participation, personal and environmental factors (WHO, 2007).

1.4.1 Body function and structure

This component includes outcomes related to the physical aspect of AIS, such as spinal curvature, range of motion, pain levels, and muscle strength (WHO, 2007). AIS can impact various bodily functions alongside the aesthetic changes characterised by spinal curvature and vertebral rotation (Reamy and Slakey, 2001). Muscle imbalances and weakness in the back and core muscles, can impact posture, stability, and general physical strength (Danielsson et al., 2006, Karin and Karin, 2019, Alves et al., 2015). Reduced lung capacity and respiratory function can also lead to decreased aerobic capacity, endurance, and increased fatigue during physical activities (Carrasco and Ruiz, 2016, Seokwon et al., 2015).

Although the existing literature presents an ambiguous standpoint around pain in AIS. Pain and discomfort, particularly in the back are the common reported symptoms in this AIS population associated with curve progression (Danielsson et al., 2006, Bastrom et al., 2013, Makino et al., 2015, Seki et al., 2018, Horne et al., 2014, Wong et al., 2019, Lonstein, 1994). However, it is difficult to determine whether the pain is truly caused by the AIS or if it merely corresponds to the predicted incidence of back pain in the adolescent population (Kordi and Rostami, 2011, Illeez et al., 2020). The annual incidence of back pain among adolescents is 15% with a point prevalence of 12% and life

time prevalence of 40% (Leite et al., 2022). The identified risk factors were sedentary lifestyle, prolonged TV watching and computer or mobile use (Baradaran Mahdavi et al., 2021).Thus, predicting and effectively treating this pain remains difficult (An et al., 2023). Bracing has been evaluated in some studies, with conflicting results on its effects on neck and back pain and functional abilities (Misterska et al., 2017). The impact of surgery is also uncertain as some individuals only have minimal discomfort, others experience severe back pain or encounter new onset of pain with surgery (Danielsson and Nachemson, 2003, Weiss and Goodall, 2008, Bastrom et al., 2013, Rullander et al., 2013, Wong et al., 2007). Overall, the existing research emphasises the complex nature of pain in AIS and the necessity of doing more research to better understand its underlying causes.

The existing literature highlights the psychological challenges experienced by individuals with AIS. Those diagnosed with AIS or undergoing management have reported feelings of anxiety, frustration, depression, stress, denial, fear, anger, and shame (Essex et al., 2022, Motyer et al., 2021a, Toye et al., 2016, Sapountzi-Krepia et al., 2006, Baird and Gardner, 2021). Some individuals expressed shock and worry upon seeing their spinal radiographs or undergoing surgery (Honeyman and Davison, 2016, Toye et al., 2016, Rullander et al., 2013). Additionally, anger, anxiety, and social problems were also reported and associated with post-surgery pain (Rullander et al., 2016). Studies have consistently shown that individuals with AIS experience lower self-image scores and dissatisfaction with their physical appearance compared to healthy individuals (Yagci et al., 2020, Du et al., 2016a, Asher et al., 2004, Watanabe et al., 2005, Merenda et al., 2011). Severe curves tend to have a greater impact on body image dissatisfaction (Cantele et al., 2021). This can lead to feelings of embarrassment and inferiority, influencing clothing choices to conceal their back by others (Du et al., 2016a, Carrasco and Ruiz, 2014, Carrasco and Ruiz, 2016). Thus, the assessment of outcomes for individuals with AIS should include a broad evaluation of psychological symptoms, such as anxiety, depression, stress and body image dissatisfaction.

1.4.2 Activities and Participation

This component focuses on outcomes related to the individual's ability to engage in daily activities, such as self-care, mobility, school performance, recreational activities, and social participation (WHO, 2007). Individuals with AIS may experience limitations in their functional ability including difficulties in movement, maintaining body position, and participating in activities requiring strength and endurance (Du et al., 2016a). Research has shown reductions in spinal mobility, lumbar movement, and muscle endurance among individuals with AIS who have undergone operative or conservative management compared to healthy controls (LaMontagne et al., 2004, Rullander et al., 2013, Rullander et al., 2017, Danielsson et al., 2006, Wilk et al., 2006, Aghdasi et al., 2020). Pain and fear of injury lead to the avoidance of physical activities and reduced participation and enjoyment of sports or recreational activities (Kakar et al., 2017, Motyer et al., 2021a, Motyer et al., 2022, Du et al., 2016a). Furthermore, challenges associated with follow-up appointments and the long recovery period from surgery impact school activities and participation (Rullander et al., 2013, Honeyman and Davison, 2016, Swierkosz et al., 2015, Bull and Grogan, 2010, Motyer et al., 2021b). The findings emphasise how AIS impacts daily activities and participation for individuals with AIS. The literature primarily focuses on assessing function using radiographs (Bagó et al., 2013). Although it is important to incorporate self-report measures or interviews, to evaluate pain intensity and fear of injury (White et al., 1999). In addition, for a comprehensive assessment of functional ability it should include physical measures to assess spinal mobility, lumbar movement, and muscle endurance using performance-based measures (Karin and Karin, 2019).

1.4.3 Environmental and personal factors

Environmental factors within the ICF-CY framework encompass social and attitudinal aspects that can either support or impede an individual with AIS's functioning and participation (WHO, 2007). Positive social support and assistance that individuals receive from their family, friends, peers, and HCPs significantly impact their well-being (Motyer et al., 2022). Providing emotional support has been identified as highly beneficial for individuals with AIS, such as brace compliance monitoring and counselling, which improves brace compliance and mental outcomes (van Niekerk et al., 2023). Some individuals with AIS choose not to disclose their health condition to their peers, which may restrict their engagement in social activities (Motyer et al., 2021a). Therefore, providing training and education on the way of communication was found to be beneficial for this group of individuals (Truumees et al., 2021).

Personal factors acknowledge aspects like coping strategies, motivation, and self-esteem that can shape the experiences of individuals with AIS (WHO, 2007). Individuals with AIS may experience lower self-esteem due to the physical changes associated with AIS. However, surgery has been shown to improve satisfaction and potentially contribute to an increase in self-esteem compared to conservative management (Cortes Paredes et al., 2011, Perry et al., 2018). Psychological interventions, such as coping instruction, have been found to be effective in reducing pain and improving psychological outcomes postoperatively (LaMontagne et al., 2003). Therefore, a comprehensive assessment approach should consider both environmental and personal factors to gain a holistic understanding of the impact of AIS on individuals' functioning and well-being.

1.5 Patient Reported Outcome Measures

Traditionally, the evaluation of the intervention effectiveness for individuals with AIS has primarily been focused on ,radiographs, to assess the degree of spinal curvature correction (Bagó et al., 2013). A recent scoping review evaluated the use of OMs among those with AIS regardless of management type, revealed that amongst 158 studies, > 61.38% of papers relied on radiographic assessments (Joarder et al., 2023). However, healthcare currently places more of an emphasis on patient-centred care, which prioritise the individual needs, preferences, values, and goals (Jayadevappa and Chhatre, 2011). Adopting this approach brings many benefits to the patient, such as enhancing patient-provider communication, improving patient outcomes, and increasing overall satisfaction (Epstein and Street, 2011). Patient reported outcomes measure (PROMs) have the potential to play a key role in this by bringing the patient's voice to the patient-centred care (Jayadevappa and Chhatre, 2011, Richards et al., 2015). PROMs or self-reported measures are the tools that are used to measure an individual's self-report about their functioning with regards to their health or treatment (Snyder et al., 2007). General PROMs, evaluate health related outcomes across different populations and conditions e.g. SF-36 (Brazier et al., 1992). Condition specific PROMs on the other hand are designed to evaluate health-related outcomes for a specific condition or treatment e.g. Neck Disability Index for people with neck pain (Beighley et al., 2022).

The SRS is the reference standard PROM for Health-Related Quality Of Life (HRQOL) assessment for individuals with AIS (Haher et al., 1999). It encompasses pain, function, self-image, mental-health and satisfaction with the treatment domains (Asher et al., 2006). The initial version of the SRS consisted of 24 items, developed by incorporating questions from previous questionnaires (Haher et al., 1999), which was later modified to 22 items (SRS-22) (Asher et al., 2000). The function domain of the SRS-22 was subsequently revised resulting in the SRS-22r (Asher et al., 2006). Over

time various variants of the SRS have been produced including the SRS-7 and the SRS-23, and the SRS-30 and most recently the SRS-16 (Baldus et al., 2011, Chen et al., 2013, Mannion et al., 2022). The existence of multiple variants of the SRS questionnaire raises concerns about the consistency and comparability of HRQOL assessment in individuals with AIS. While modifications aim to improve the questionnaire, the proliferation of versions creates confusion and challenges for researchers and clinicians in selecting the most appropriate version to use. To address this issue, a critical evaluation of the psychometric properties of different SRS versions is necessary to determine the most appropriate and reliable options for this specific population (Mokkink et al., 2018b). Achieving standardisation and consensus on a single, widely accepted SRS questionnaire version would greatly enhance the comparability and reliability of HRQOL measurements in individuals with AIS (Prinsen et al., 2016).

1.5.1 Development of a PROM

The process of development of a PROM consists of several steps, which is an iterative rather than linear process requiring re-evaluations and adaptations of the PROM, before it can be used in its final version (Patrick et al., 2011a). *See Figure* 1.7 which illustrates this process. Defining the construct that the PROM aims to assess is the initial step in developing a PROM (De Vet et al., 2011). This involves identifying the relevant aspects of the individual's experience that developers are interested in measuring, such as symptoms or functioning (Patrick et al., 2011a). Also, the relevant characteristics of the targeted population such as age, gender and the severity of the disease should be identified (De Vet et al., 2011). Furthermore, it is important to decide the context of use of the PROM whether it is designed for diagnosis, evaluative or prognostic purposes (De Vet et al., 2011, Patrick et al., 2011a).

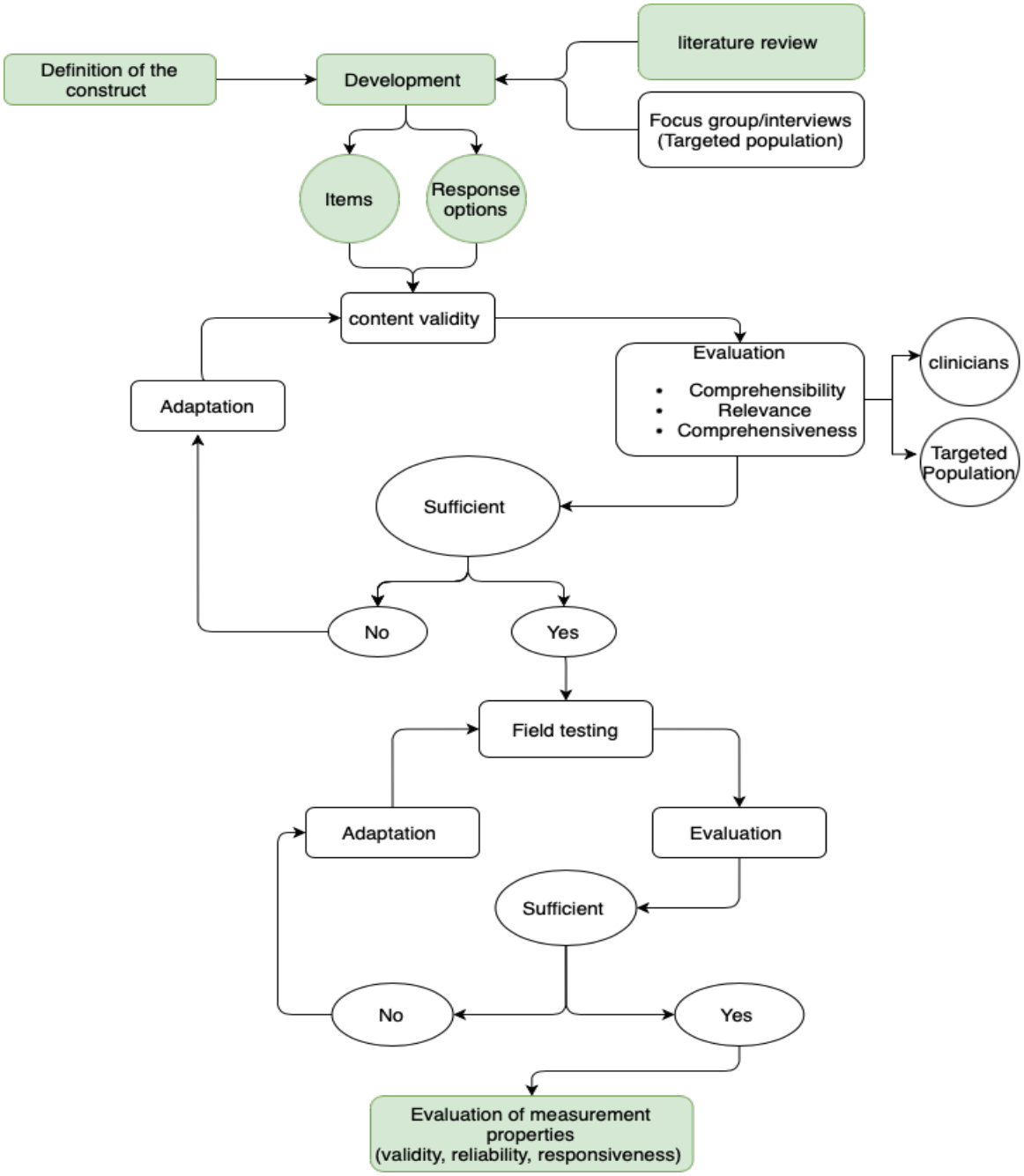


Figure 1.7: Overview of the steps in the development and evaluation of a PROM.

Figure adapted from (De Vet et al., 2011). Green coloured shapes are the steps that has been completed for the SRS.

The second step includes generating the first draft of a PROM as a pool of potential items or questions to assess the identified concepts/domains (Mohtadi, 2016). These items are often derived from different sources such as concept elicitation interviews with targeted population, existing PROMs, or literature review (De Vet et al., 2011). It is crucial to consider the contribution of the targeted population in this step, as they are the key experts in their own health condition (Lasch et al., 2010, Brod et al., 2009, Mohtadi, 2016). However, the current focus of PROM development has given limited attention to this approach, as tends to prioritise quantitative methods (Brédart et al., 2014). A well-designed PROM should have questions that are easily understood by the target population regardless of their educational level, use simple language, and avoid complex or ambiguous terms. Also, it should have a clear instructions and specified time frames (Streiner et al., 2015, De Vet et al., 2011). Designing a questionnaire specifically for adolescents aged 10-18 years old requires consideration of their unique characteristics and needs (Brédart et al., 2014). Adolescents have cognitive abilities and developmental milestones that differ from those of adults (Matza et al., 2013). To ensure accurate understanding and response to the questionnaire, it is important to design it using language that is suitable for their age and level of comprehension (Matza et al., 2004).

Pilot testing to evaluate the content validity of a PROM is the next step in the PROM development process (De Vet et al., 2011). In this step, the target population are asked about comprehensibility, relevance, and comprehensiveness of questionnaire items as well as about acceptability, and feasibility of the questionnaire (Terwee et al., 2018c). This evaluation process is known as cognitive debriefing in which participants provided feedback on the questionnaire through techniques such as think aloud and verbal probing (Patrick et al., 2011b). It is considered a critical step in PROM development usually carried out prior to quantitative psychometric testing (Patrick et al., 2011b). It provides evidence of the content validity of the PROM, by eliciting potential errors, and the pattern of interpretation amongst a number of related and relevant respondents. If the

interviews reveal any misunderstanding, then modification of the questionnaire is needed (Rothman et al., 2009). If no problems are detected then the PROM can be used in field testing and other measurement properties can be assessed (De Vet et al., 2011). Methods like factor analysis and item response theory are used in field testing to assess the PROM dimensionality in a larger sample of targeted population, unlike the pilot testing which involves qualitative analysis with fewer participants (De Vet et al., 2011).

The SRS was originally developed for surgically manged individuals by incorporating questions from previous questionnaires designed for adult populations (Haher et al., 1999). For instance, the mental health domain of the SRS includes items derived from the SF-36 survey (Asher et al., 2000), which was designed for an adult population, and serve as a generic rather than condition-specific measure. Further, the mental health domain of SRS aims to evaluate aspects like self-confidence and self-esteem but it fails to capturing the complex emotions of social isolation, the need for reassurance, and the fear because of lack of comprehensive understanding about their condition (Joarder et al., 2023).

1.5.2 Content validity of a PROM

Content validity refers to "the degree to which elements of an assessment instrument are relevant to, and representative of the targeted construct for a particular assessment purpose" (Haynes et al., 1995). It ensures that content of the PROM is consistent with the perspective, words, and experiences of the target population (Brod et al., 2009, Rothman et al., 2009). The initiatives of the Core Outcome Measures in Effectiveness Trials (COMET) consortium emphasise importance of high-quality evidence of good content validity when selecting a PROM in a COS (Prinsen et al., 2016). The COSMIN group also suggested that the recommendation of the most suitable PROM to

be selected for a particular purpose is based on the high quality evidence of sufficient content validity (Mokkink et al., 2018b). They also provide guidelines on the methods of evaluating the content validity (Terwee et al., 2018b). Content validity may affect other measurement properties of PROM such as internal consistency, structural validity, and responsiveness (De Vet et al., 2011). A PROM with adequate content validity ensures that it capture the experiences and perspective of the population of interest, while a PROM that lacks content validity might compromise the validity of study findings (Vogt et al., 2004). Qualitative research methods, including concept elicitation and cognitive debriefing methods may usefully assess content validity by interviewing the target population to understand their perspectives regarding the impact of the disease on their own health condition as well as their feedback on the questionnaire items (Holman, 1993, Toye et al., 2016, Brédart et al., 2014).

1.5.3 Content validity of the Scoliosis Research Society questionnaire

The content validity of the SRS is compromised by two key issues: the absence of qualitative assessment involving the target population during the development of the SRS. Furthermore the lack of cognitive debriefing in the pilot testing assessment (Haher et al., 1999). According to the COSMIN guidance, these shortcomings indicate insufficient development of the PROM and compromise its content validity (Terwee et al., 2018c).

Figure 1.7 illustrates the development process of a PROM, the highlighted shapes represent the completed steps for the SRS, which include defining the construct, developing the items and response options through literature review, and assessing the psychometric properties. However, the qualitative assessment steps, concept elicitation and cognitive debriefings were not completed for this

questionnaire. These qualitative assessments are crucial for ensuring content validity and understanding the perspectives and experiences of the target population (Willis, 2017, Willis and Artino, 2013).

1.6 Performance-based outcome measures

Radiographic measurement is the gold standard and traditional measure used to evaluate spine deformity. However, these measurements may not be consistent with clinical outcomes or predict the progression of deformity. Some individuals with severe curves may have minimal functional limitations, while others with less severe curves experience significant symptoms (Kieser et al., 2022, D'Andrea et al., 2000). These variations highlight the need for an additional measure that captures the holistic impact of AIS on an individual's life. While PROMs offer benefits in understanding the perspective of an individual about the impact of the AIS on their own functional ability. However, it is subjective and affected by the individual's emotional and cognitive status (Reiman and Manske, 2011). Factors like pain and avoidance of certain activities as well as psychological stress are often reported in this population (Leszczewska et al., 2012, Rullander et al., 2016, Bastrom et al., 2013, Théroux et al., 2017). These factors can influence self-report measures potentially leading to an incomplete understanding of an individual's functional ability (Bastrom et al., 2013, Théroux et al., 2017).

Given the limitations associated with using radiographic measurements in assessing individuals with AIS, alternative methods such as Performance-Based Outcome Measures (PBOMs) can provide additional insights into these assessments. PBOMs involve assessment of an individual's functional ability during performance of a specific task or activity, measured by time, or distance. These measures rely on the clinician or observer evaluation (Bean et al., 2011). PBOMs are measures

that provide quantifiable data, which reduce the potential for bias and provides a standard approach to assess function (Harvey et al., 2007)

The use of PBOMs offer a different perspective by focusing on evaluating individual's functional ability and performance by directly evaluating specific functional domains e.g. strength and endurance (Studenski et al., 2003). This enables targeted assessment of functional abilities that is relevant to the individual's goals or condition. This will help clinicians to demonstrate changes in functional ability, design tailored interventions and track improvement in those specific domains (Munguía-Izquierdo et al., 2021). In combining PBOMs with PROMs researchers and clinicians can gain a more comprehensive understanding of an individual's condition, treatment outcomes, and overall well-being (De Vet et al., 2011, Danielsson et al., 2006, Karin and Karin, 2019, Rigo, 2011). However, limited information currently exists about the available PBOMs for individuals with AIS and their measurement properties.

1.7 Body structure and function measures

Body structure and function measures directly assess a specific aspect of the individual's body structure and function (Reiman and Manske, 2011). Such as "the physiological function of body systems e.g. strength and / or the anatomical parts of body i.e. range of motion (Taylor et al., 2016a). These measures are performed by clinicians and can provide quantitative data on the physical or biological aspects of the health condition (De Vet et al., 2011). Imaging such as radiographs are the frequently used measure for individuals with AIS (Joarder et al., 2023). Using of the radiographs as the primary measure increase exposure of the individual to ionizing radiation, which carries risks especially for young individuals who may require long-term monitoring (Luan et al., 2020, Rose et al., 2023). Furthermore, radiographic assessment focuses primarily on the structural aspect of the

spinal deformity i.e. curve angle, which is important for diagnostic and monitoring purposes (Reiman and Manske, 2011). However, it does not provide a comprehensive assessment of functional limitations, pain, or QOL experienced by individuals with AIS. Moreover, radiograph lack patient-centred perspective because it is clinician-focused unlike PROMs or PBOMs (Taylor et al., 2016a). Different physical functional tests such as those used by physiotherapists, are available to evaluate muscle strength or range of motion. The literature is increasingly recognising the potential benefits of assessing and evaluating these measures (Panagiotis et al., 2019, Master et al., 2020). If their measurement properties are established for AIS, they could serve as additional options for assessing spine health and allow for a more comprehensive evaluation of patients.

1.8 Measurement properties of outcome measures

Measurement is an essential part of evidence-based paractice since it enable diagnosis, prognosis, and evalaution of interventions (Antunes et al., 2018). Therefore, it is crucial to follow rigorous scientific guidelines while evaluating measurement properties of an OM to ensure its appropriate use and accurate interpretation of the results (Prinsen et al., 2018). The COSMIN group has made substantial contributions in this regard (Mokkink et al., 2010, Prinsen et al., 2016). They created a taxonomy of measurement properties to help guide the selection of OMs, with an emphasis on three domains accompanied with additional subgrouping; reliability i.e. “the degree to which the measurement is free from measurement error”, validity i.e. “the degree to which a PROM measures the construct(s) it purports to measure” , and responsiveness i.e. “the ability of a PROM to detect change over time in the construct to be measured”, (Mokkink et al., 2010). They have also included interpretability which is the degree that qualitative meaning can be assigned to the quantitative scores or change in the scores (Mokkink et al., 2010). These measurement properties along with their definitions are presented in Table 1.2. The COSMIN group also provided

guidelines for conducting systematic review of measurement properties (Mokkink et al., 2018b). Furthermore, the COSMIN group has created a checklist to evaluate the methodological quality of single studies on the measurement properties of PROMs (Mokkink et al., 2018a). This checklist has been tailored specifically for systematic reviews of PROMs, offering a standardised and rigorous way to assessing measurement quality (Mokkink et al., 2018a). It should be noted that the COSMIN group promotes the modification of these tools for use with OMs other than PROMs such as PBOMs and body structure and function measure (Mokkink et al., 2018b). Utilizing a validated and reliable measure and adhering to rigorous measurement guidelines in AIS (Mokkink et al., 2010, Terwee et al., 2018c, Prinsen et al., 2018), will lead to improved diagnostic precision, enhanced monitoring of curve progression, better evaluation of treatment outcomes, and the accumulation of evidence-based knowledge (David, 2017, Mistovich et al., 2023). This ultimately contributes to improved care and outcomes for individuals with AIS.

Table 1.2: COSMIN definitions of domains and measurement properties. Adapted from (Mokkink et al., 2010)

Terminology		**Definition**
Domain	**Measurement property**	
Reliability		Degree to which the measurement is free from measurement error
	Internal consistency	The degree of the interrelatedness among the items
	Reliability	The proportion of the total variance in the measurements which is due to 'true'† differences between patients
	Measurement error	The systematic and random error of a patient's score that is not attributed to true changes in the construct to be measured
Validity		The degree to which a PROM measures the constructs measure what is supposed to measure
	Content validity	The degree to which the content of a PROM is an adequate reflection of the construct to be measured
	Face validity	The degree to which (the items of) a PROM indeed looks as though they are an adequate reflection of the construct to be measured
	Construct validity	The degree to which the scores of a PROM are consistent with hypotheses
	Structural validity	The degree to which the scores of a PROM are an adequate reflection of the dimensionality of the construct to be measured
	Cross- cultural validity	The degree to which the performance of the items on a translated or culturally adapted PROM are an adequate reflection of the performance of the items of the original version of the PROM
	Criterion validity	The degree to which the scores of a PROM are an adequate reflection of a 'gold standard'
Responsiveness		The ability of a PROM to detect change over time in the construct to be measured
Interpretability		The degree that qualitative meaning can be assigned to the quantitative scores or change in the score

1.9 Research and knowledge gaps

This introductory chapter highlights the limited existing knowledge regarding the assessment of outcomes for individuals with AIS. Several research and knowledge gaps were identified that led to research studies that has been undertaken in this thesis.

1.9.1 Assessment of functional outcomes

The rate of surgical interventions for scoliosis has risen globally (von Heideken et al., 2018). In the United Kingdom, between 2005 and 2018, the number of scoliosis surgeries per 100,000 individuals rose from 4.4 to 9.8 (Tsirikos et al., 2020). The existing literature and our understanding of the impact of surgery on pain and physical functioning (PF) for individuals with AIS are inconsistent. While some studies suggest that surgery can result in minor discomfort, other data indicates that adolescents may experience significant back pain after surgical fusion (Raf et al., 2022, Bastrom et al., 2013, Ramirez et al., 1997, AlAssiri et al., 2022, Seki et al., 2018, Wong et al., 2007, Bailey et al., 2021). Furthermore, the factors contributing to pain are poorly understood, and anticipating and successfully treating this pain remains challenging (An et al., 2023). While there is some evidence suggesting that surgery can lead to functional limitations, including reduced spinal mobility and muscle endurance (Nokariya et al., 2022, Karin and Karin, 2019, He and Wong, 2018), there is a lack of a thorough understanding of the variables influencing functional limitations following surgery. Therefore, comprehending how surgery has impacted an individual with AIS's functional outcomes, as well as whether observed improvements persist or decline over time, is crucial for providing comprehensive insights into surgery outcomes.

1.9.2 Content validity

The SRS is the reference standard PROM for individuals with AIS. It was developed in 1999, to evaluate HRQOL following surgical approaches in individuals with AIS. It includes four domains: function, pain, body-image, and mental health (Haher et al., 1999). The SRS has been revised and multiple versions are now available that have been adapted and translated into more than 10 different languages (Alanay et al., 2005, Beausejour et al., 2009). The popularity and the availability of various SRS questionnaire variants suggest that it is considered an appropriate tool for assessing outcomes in individuals with AIS. However, a comprehensive evaluation of its appropriateness specifically for individuals with AIS has been disregarded. Current research focused on measurement properties tends to emphasise quantitative assessments (Asher et al., 2003c, Parent et al., 2009, Berliner et al., 2013, Asher et al., 2000, Wong et al., 2017, Lee et al., 2017, Cheung et al., 2007, Asher et al., 2003d, Alanay et al., 2005, Wu et al., 2014). This approach often neglects examination of the questionnaire's content validity, concerning its relevance, comprehensiveness, and comprehensibility (De Vet et al., 2011).

Content validity is an important aspect of questionnaire development, ensuring that the items and domains of the questionnaire accurately capture the relevant aspects of the condition being assessed (De Vet et al., 2011, Magasi et al., 2012). In the case of individuals with AIS, it is crucial to have PROMs that closely relevant to their age, experiences and activities, considering their specific challenges and concerns they may face (Matza et al., 2013, Matza et al., 2004). There is a growing emphasis on utilizing qualitative methods to gain deep understanding of adolescents' perspectives through qualitative research approaches (Rullander et al., 2013, Motyer et al., 2021a, Motyer et al., 2021b, McNeill et al., 2021). However, the appropriateness of the SRS to individuals with AIS through qualitative assessment have not yet been investigated.

1.9.3 Measurement properties

Currently, the radiograph is the gold standard body structure OM, and the SRS questionnaire is the reference standard PROM (Joarder et al., 2023, Asher et al., 2003c). However, concerns have been raised regarding its applicability to this population and usefulness in non-operative management approaches (Asher et al., 2000). The existence of multiple versions of SRS and the lack of unified all-encompassing PROM prevents data pooling and synthesis. This leads to avoidable inefficiencies in research production and reporting, along with difficulties in effectively using research findings (Chalmers and Glasziou, 2009). To address this issue, a critical evaluation of the psychometric properties of the different OMs available for individuals for AIS is necessary to determine the most appropriate options while ensuring sufficient measurement properties for this specific population (Mokkink et al., 2018b, Mokkink et al., 2018a, Matza et al., 2004, Patrick et al., 2011b).

1.9.4 Performance based assessment

There is a lack of understanding about prevailing practices and the holistic impact of interventions like surgery and exercise on functional performance for individuals with AIS. The majority of research conducted in AIS has primarily focused on assessing outcomes using radiograph (Joarder et al., 2023) or PROMs (Honeyman and Davison, 2016). While PROMs provide valuable insights into patients' personal experiences, they may not capture the complete picture of functional ability (Reiman and Manske, 2011). Furthermore, they are influenced by pain and psychological problems often reported by this population (Rullander et al., 2016, Bastrom et al., 2013). The PBOMs can provide quantifiable data that reflect the actual physical performance of an individual's ability (Reiman and Manske, 2011). By incorporating PBOMs alongside PROMs, healthcare professionals (HCPs) can gain a more comprehensive understanding of an individual's functional ability (Bean et

al., 2011). PBOMs offer a unique perspective on the functional ability of individuals by directly evaluating specific functional domains like strength and endurance (Reiman and Manske, 2011). They provide a more targeted assessment of functional abilities that are relevant to the individual's goals or condition (Taylor et al., 2016a, Beaudart et al., 2019). This will allow clinicians to accurately assess functional abilities, design personalized interventions, and track progress in those specific domains (Munguía-Izquierdo et al., 2021). However, there is limited research exploring the use of PBOMs specifically in individuals with AIS.

1.10 Thesis aim and objectives

The aim of this thesis was to explore the current use of OMs for individuals with AIS and evaluate their measurement properties. This was achieved through the following specific objectives:

1. To evaluate clinically relevant changes in surgical outcomes for individuals with AIS, comparing those who achieved smallest detectable change (SDC) in pain and function at 1-year post-surgery and those who did not, and to evaluate the influencing factors. (*Chapter Two*)

2. To identify OMs used to assess physical functioning in individuals with AIS, and to summarise their measurement properties. (*Chapter Three*)

3. To evaluate the contents of the SRS-22r questionnaire regarding its relevance, comprehensiveness, and comprehensibility from individuals with AIS and HCPs perspectives. (*Chapter Four*)

4. To determine whether the contents of the SRS-22r questionnaire are relevant and important to individuals with AIS, by exploring the impact of scoliosis and its associated treatment on their HRQOL. (*Chapter Five*)
5. To explore the current practice of the HCPs when assessing outcomes for individuals with AIS, and to understand the perceived barriers and facilitators of HCPs to use PROMs as well as PBOMs. (*Chapter Six*)

See Figure 1.8 which presents a visual representation of the objectives of this thesis.

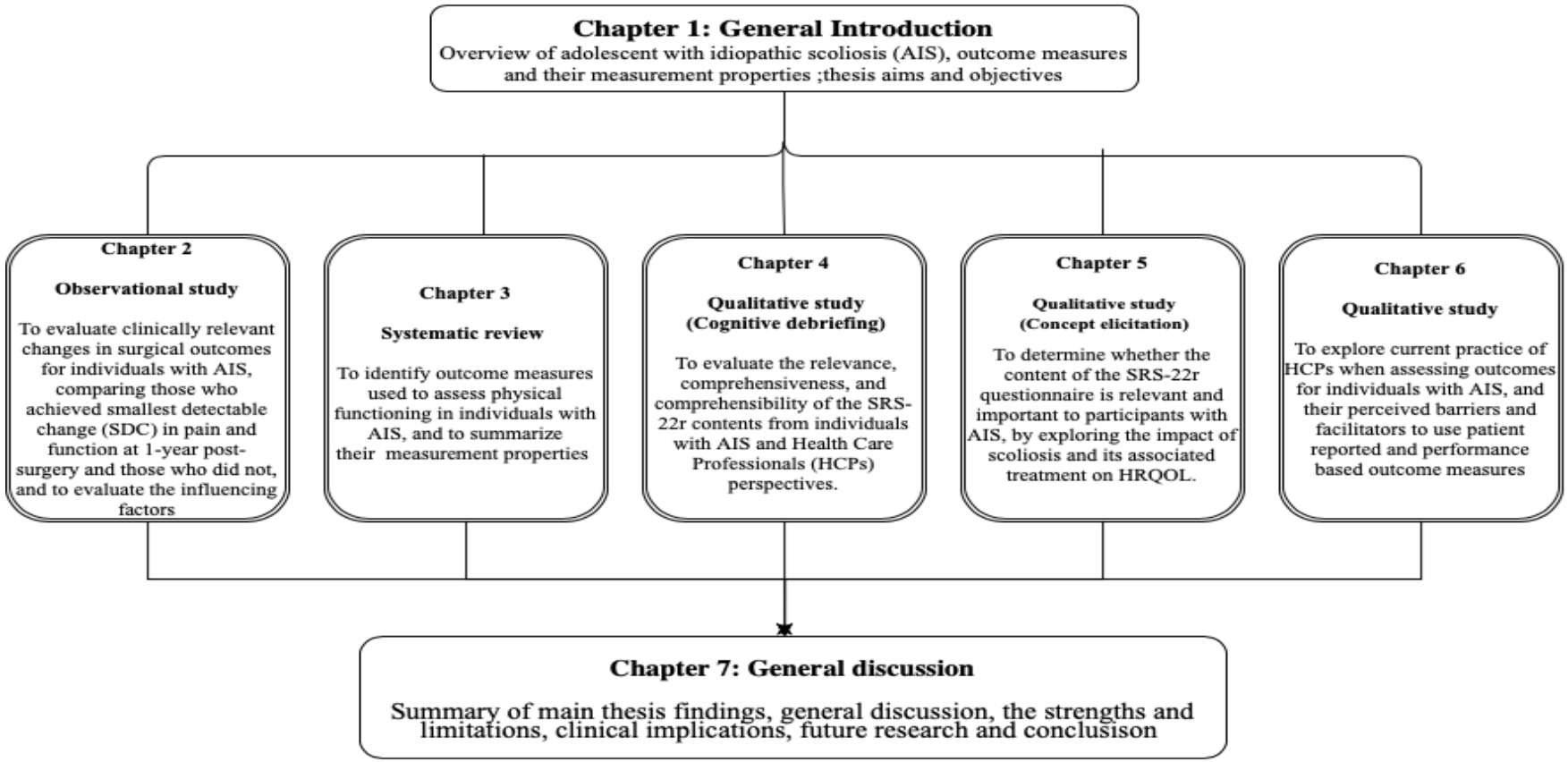


Figure 1.8: Visual representation of the thesis, including the objectives of each chapter

1.11 Thesis Chapters Overview

Chapter One provides an overall summary on the current research of HRQOL outcomes particularly pain and function for individuals with AIS as well as the method of evaluations. *Chapter Two* reports the results of cohort study that evaluated the clinically relevant changes in surgical outcomes for individuals with AIS, comparing those who achieved SDC in pain and function at 1-year post-surgery and those who did not, and discussed the influencing factors. In light of the high ceiling effects observed in the SRS-22r, *Chapter Two* highlights the need to identify appropriate OMs that can confidently assess function in individuals with AIS. This sets the stage for further exploration of OMs and their measurement properties in *Chapter Three* of the thesis. The systematic review in *Chapter Three* evaluates physical functioning measures for individuals with AIS, and it summarises their measurement properties. Review findings revealed insufficient measurement properties of OMs, particularly in terms of content validity of PROMs. These results led to the subsequent studies in *Chapter Four, Chapter Five and Chapter Six.*

Chapter Four presents the results of a qualitative study with HCPs and individuals with AIS who critically evaluated the content of the SRS-22r questionnaire with regards of its relevance, comprehensiveness, and comprehensibility. *Chapter Five* presents the results of a qualitative study with individuals with AIS who determined the relevant and key areas from their perspective about the impact of scoliosis and its associated treatment on their HRQOL and compares it with the domains and items of the SRS-22r questionnaire. *Chapter Six* presents the results of a qualitative study with HCPs that aimed to understand the current practice of assessment of using OMs for individuals with AIS and presents perceived barriers and facilitators of HCPs to use PROMs as well as PBOMs for this population.

Chapter Seven discusses the results of the studies in *Chapters Two, Three, Four, Five, and Six.* This chapter summarises the key research findings of the thesis and includes a general discussion of the research, as well as reflections on its strengths and limitations. In addition, it considers the clinical implications of this research and makes recommendations for future research.

The results from the studies in this thesis are intended to help researchers and HCPs on understanding of the impact of surgery on functional outcomes for individuals with AIS, and the best available OMs for evaluating HRQOL including PF for this population, enhance the content validity of the frequently used PROM for this population as well as make recommendations to enhance use of PBOM for AIS.

Chapter Two

PREDICTIVE FACTORS OF CLINICALLY RELEVANT CHANGES IN PAIN AND FUNCTION FOLLOWING SURGERY FOR ADOLESCENTS WITH IDIOPATHIC SCOLIOSIS

This chapter reports in full the contents of a published manuscript by the thesis author (Alamrani, et al., 2023). It includes verbatim text from the published manuscript and some changes in the introduction section were employed for the purpose of this thesis.

Publications and Conference presentation

- **Alamrani S,** Gardner A, Rushton AB, Falla D, Heneghan NR. 2023 Predictors of Relevant Changes in Pain and Function for Adolescents with Idiopathic Scoliosis following Surgery. Spine (Phila Pa 1976). 2023 May 5. doi: 10.1097/BRS.0000000000004705. Epub ahead of print. PMID: 37146097. (**Appendix 1**)

- **Alamrani S,** Gardner A, Rushton AB, Falla D. Heneghan NR. Predictive factors of clinically relevant changes in pain and function following surgery for adolescents with idiopathic scoliosis. Physiotherapy UK- 1st November 2023. Birmingham. United Kingdom. (Poster presentation).

2.1 Abstract

Recent work reported that the Small Detectable Change (SDC) value for the SRS-22r is larger than Minimal Clinically Important Difference (MCID) value. Therefore, it has been suggested to use SDC to evaluate benefits from surgery for individuals with AIS as alternative to MCID. This *chapter* aimed firstly, to evaluate relevant changes of surgical outcomes of individuals with AIS, over short and long-term periods using SDC. Secondly, to compare between individuals with AIS, who achieved SDC in pain and function domains at 1 year following surgery, and those who did not. Finally, to evaluate influencing factors on achieving SDC at both domains.

A retrospective analysis of data from individuals with AIS, who underwent surgical correction at a tertiary spinal centre, between 2009 and 2019. The course of surgical outcomes was assessed using SRS-22r at both short term (6 weeks, 6 months) and long term (1 year ,2 years) following-surgery. The difference between 'successful' (change in scores ≥ SDC) and 'unsuccessful' (change in scores < SDC) groups was assessed by independent t-test. Univariate and logistic regression analysis were used to assess influencing factors on achieving SDC value at both domains.

All SRS-22r domains, except self-image and satisfaction, decreased at 6 weeks following-surgery for all eligible participants (n=124). At 1 year, the greatest change was in self-image increasing by 1.21point, with function increasing by 0.19 point, while pain decreasing by 0.12 point. At all SRS-22r domains, the successful group had low pre-surgery scores and were statistically different than unsuccessful group. The difference remains statistically significant at follow-up except in function and subtotal domains. Being older and have low pre-surgery scores increase the chances of achieving SDC in function at follow up. While achieving SDC at pain domain was significantly

associated with age, gender and length of hospital stay, low pain scores and high function scores pre-surgery.

Notably, the self-image domain showed the largest change compared to other SRS-22r domains. Participant with low pre-surgery scores classified as successful and they were more likely to had benefit from surgery, indicating an inherent defect within the SRS-22r. Further research is needed to identify the best available PROM to evaluate functional outcomes for individuals with AIS.

2.2 Introduction

As was detailed in *Chapter One: sections 1.41.5,* there has been an exponential increase in surgical rates worldwide (Tsirikos et al., 2020, von Heideken et al., 2018, Vigneswaran et al., 2015). Given this increase in the using of surgery in the management of AIS, and the associated costs, an assessment of the surgery benefits from a patient point of view, is timely. The primary aim of surgery is to stop curve progression and improve self-image (Bettany-Saltikov et al., 2016, Lee et al., 2016, Fernandes et al., 2019, Helenius et al., 2019). However, it has been reported that scoliosis correction surgery is associated with consequences such as pain, reduction in spinal mobility and flexibility (Rullander et al., 2013, Wong et al., 2007, Weiss and Goodall, 2008, Bastrom et al., 2013), which affect participation in activities (Aghdasi et al., 2020, Kakar et al., 2017). Therefore, it is important to evaluate not only the absolute effect of surgery on pain and function, as important HRQOL outcomes (Hughes et al., 2021), but also to understand if these changes are relevant and important to patients' perspective.

The smallest change in the outcome score that a patient perceives as important is the MCID (Hays and Woolley, 2000). For a clinical change to be detected, the MCID value should be larger than the SDC , which is the smallest change that exceeds the measurement error within the instrument (De Vet et al., 2011). The MCID is a clinical concept that determines whether the change is meaningful and important to the patient, whereas the SDC is a statistical concept that ascertains that the change is real (Spratt, 2009, De Vet et al., 2011). Therefore, it is important to consider both values as statistical significance may not be reflecting clinical significance. Any change in the score below the SDC is attributed to a measurement error (De Vet et al., 2011). Both values of MCID and SDC refer to the interpretability, which is an important aspect of an instrument's measurement properties, as reflected in its inclusion in the COSMIN taxonomy (Mokkink et al., 2010).

The SRS-22r is a frequently used questionnaire to evaluate HRQOL for individuals with AIS, consisting of five domains, including pain and function (Asher et al., 2006). It has been found that the MCID values for the pain and function domains are smaller than the calculated SDC values. Specifically, the MCID values are reported as 0.2 and 0.08, while the SDC values are 0.3 and 0.24, respectively (Kelly et al., 2019, Carreon et al., 2010). Due to this discrepancy, it has been suggested to use the SDC as a threshold for clinically relevant change instead of the MCID (Kelly et al., 2019, Kelly et al., 2018). The rationale behind this suggestion is that using the SDC ensures that any observed changes are beyond the measurement error and thus more likely to be real and meaningful (De Vet et al., 2011). Noteworthy, that the SRS-22r has been reported to exhibit a high ceiling effect in its function domain, which affects its ability to discriminate change (Asher et al., 2006, Glattes et al., 2007). To address concerns about distinguishing real change from measurement error, Spratt et al. proposed a 30% rule for measuring clinical change by accommodating PROM baseline scores (Spratt, 2009). However, it is important to recognise that Spratt's method is theoretical, and the validity of alternative rules, such as the 20% or 50% rule, remains questionable (Spratt, 2009).

Previous studies that used the MCID for the SRS-22 or SRS-30 showed an improvement in pain and function scores (Mens et al., 2022, Djurasovic et al., 2016, Bailey et al., 2021), while others revealed no change (Rushton and Grevitt, 2013). To the best of our knowledge, no previous study has specifically used the SDC values to evaluate changes following surgery. Additionally, the factors that influence the achievement of the SDC values in this questionnaire are poorly understood.

This chapter addresses the first objective of this thesis as follows:

1- To evaluate the relevant changes in surgical outcomes for AIS surgery, over the short and long-term using SDC values.

2- To compare between individuals with AIS who achieved the SDC in function and pain at 1 year follow up and those who did not.

3- To identify factors that may influence reaching the SDC value for the function and pain domains at a 1-year follow up.

2.3 Methods

This study is reported in line with the Strengthening the Reporting of Observational studies in Epidemiology (STROBE) guidelines (von Elm et al., 2014) (*see Appendix* 6: STROBE checklist).

2.4 Study design

A cohort study design using routinely collected longitudinal outcome data from individuals with AIS, who have undergone spinal fusion surgery.

2.5 Setting

The data was collected between 2009 and 2019. The data was obtained from the British spine registry (BSR) at single tertiary spine centre in the Birmingham, United Kingdom (UK). The primary purpose of this registry is to monitor and track the outcomes of patients who undergo spine procedures, including individuals with AIS. As part of the registry, patients are invited to complete the SRS-22r questionnaire at various stages during their treatment.

2.6 Participants

➢ Eligibility criteria

Adolescents (aged 10 to 18 years old), diagnosed with AIS, who provide the SRS-22r questionnaire before or after the surgery, and provided a complete set of scores that permit calculation of valid score. The domain of the SRS-22r cannot be scored if fewer than 3 questions were answered.

➢ Exclusion criteria

Patients with other spine pathologies (e.g., neuromuscular scoliosis, Juvenile idiopathic scoliosis) were excluded.

2.7 Ethics

Approval to conduct this study was granted from the institution review board at the Royal Orthopaedic Hospital, Birmingham, UK (DP/ROH20ORTH01) (*see Appendix* 6). The data was a routine care data, previously collected, anonymised with no potential of affecting the care of the individuals concerned, thus consent was not deemed necessary.

2.8 Variables

The functional outcomes of individuals with AIS following spinal fusion surgery were assessed using the SRS-22r questionnaire at various time points (6 weeks, 6 months, 1 year, and 2 years post-surgery). The primary aim was to determine successful and unsuccessful outcomes based on meeting or not meeting SDC values. The exposure consisted of individuals with AIS undergoing spinal fusion surgery. Predictors of interest for achieving SDC values in function and pain domains at 1-year follow-up included age, gender, length of hospital stay, pre-surgery scores, and other clinical and demographic characteristics.

2.9 Data Sources

The data were collected as routine standard of care and stored electronically. The collected data included demographic and clinical information as well as the SRS-22r questionnaire. The SRS-22r is a self-reported questionnaire used to evaluate the perceived HRQOL in individuals with AIS (Asher et al., 2006). The SRS-22r demonstrates reliability, concurrent validity (Glattes et al., 2007), and it has been shown to be responsive to change during the post-surgical period (Asher et al., 2003b). The SRS-22r questionnaire comprises five domains including function, pain, self-image, mental health, and satisfaction. Each domain consists of 5 items, except for satisfaction which has 2 items. The response options are based on a 5-points Likert scale where higher scores indicates better outcomes (Asher et al., 2006). Additionally, to the SRS-22r questionnaire, demographic and clinical characteristics of individuals with AIS were extracted from their medical records. This included information such as age, sex, curve severity, curve type determined according to the Lenke classification (Lenke et al., 2001), surgical procedure performed, and length of hospital stay.

To check data validity the following steps were conducted :

1- The accuracy of the data source was verified as it has been collected from a reliable and secure system within the hospital accessed only to authorized individuals. Irrelevant, and duplicates data were assessed and then removed.

2- Data were checked for completeness and that all required field are completed. The score of SRS-22r domain was considered missing and excluded from the analysis when a participant answered less than 3 questions out of the possible 5 that contributed to the total domain score.

3- The data format was checked for accuracy and consistency. Ensure for each entry that the accurate data format is entered. For example, age (numbers), size of curve (numbers), type of surgery (words) (Emanuelson and Egenvall, 2014).

2.10 Missing data

A descriptive univariate analysis test was performed to check the amount of missing data and if it is related to specific variable/variables. The analysis showed that the there was more than 25 % of missing values at study outcome variables (i.e., function, pain, self-image, mental health, subtotal, and total scores). The missingness percentages at different time points were as follows: 26% at baseline, 73% at 6 weeks, 83% at 6 Months, 51 % at 1 year and 59% at 2 years. However, clinical, and demographic characteristics were completed. To investigate the missing data mechanism, Little's MCAR test was conducted to assess the missing data mechanism (Little, 1988). The results indicated that the missingness pattern was not significantly associated with the observed variables, $\chi^2(797) = 0.861$, $p > .05$, suggesting that the missing data could be considered as missing completely at random (MCAR). Therefore, participants data with incomplete observations were dropped from the final analysis (Sainani, 2015).

2.11 Bias

The loss at follow up may introduce bias into the results, therefore a comparison between the study cohort and the non-response group was performed to identify any potential of selection bias (Howe et al., 2016b) .

2.12 Statistical methods

Demographic and clinical characteristics were descriptively summarized using mean and standard deviation (SD) or median and interquartile range (IQR), as appropriate. Categorical variables were presented as frequencies and percentages. Parametric assumptions were checked using the Shapiro-wilk test (Razali and Wah, 2011).

To check for potential selection bias, a comparison between the study cohort and the non-response group was performed (Howe et al., 2016a), with an independent-samples t-test or Mann-Whitney U test for continuous data (Kim, 2019), and a chi-squared test for ordinal data (Fisher's exact test for small sample sizes) (Shan and Gerstenberger, 2017).

The course of the clinical outcomes for AIS was graphically presented using boxplot for each domain of the SRS-22r scores, at 6 weeks, 6 months, 1 year, and 2 years. Pre-surgery scores were used as baseline scores to determine changes at 1 and 2 years. An increase in the score beyond the SDC is consider a positive change indicating improving in participants' outcomes, and a decrease below the SDC is considered a negative change indicating worsening in participants' outcomes.

Participants' data were then classified into successful (≥ SDC) and unsuccessful (< SDC) groups and compared using an independent-sample t-test. The SDC values for pain (0.3), function (0.24), self-image (0.3), mental health (0.27), and the subtotal score (0.23) have been previously determined (Kelly et al., 2019).

A univariate analysis was performed to evaluate factors associated with reaching SDC in the function and pain domains at 1 year, as it is the frequently used time point for the assessment of HRQOL for spinal fusion surgery (Adogwa et al., 2016). The strength of the correlation was interpreted as follows: .90 to 1.00 (−.90 to −1.00) as a very high positive (negative) correlation, 70 to

.90 (−.70 to −.90) as a high positive (negative) correlation, .50 to .70 (−.50 to −.70) as a moderate positive (negative) correlation, .30 to .50 (−.30 to −.50) as a low positive (negative) correlation, and .00 to .30 (.00 to −.30) as a negligible correlation (Mukaka, 2012).

Logistic regression analysis was then performed to ascertain the effects of independent variables on the pain and function at 1 year follow up (Sperandei, 2014). The dependent variables (pain and function scores at 1 year) were dichotomized into “successful” and “unsuccessful”, using the SDC value of each domain. The Final prediction models with odds ratios (OR) and 95% confidence intervals (95% CI) were calculated for each predictive factor (Sperandei, 2014). Statistical analysis was performed using SPSS version 28.0 the Mac package (Armonk, NY: IBM Corp.). Statistical significance was set at $p<0.05$.

2.13 Results

2.14 Participants

Overall, 304 patients met the inclusion criteria, however only 124 patients had a complete follow up data at 1 year and were included in the analysis. The study cohort was compared with those who had no responses with respect to their baseline characteristics. A significant difference was found only for pre-surgery function scores (p=.037), where the study cohort had a higher functional median score (4: IQR1: 2) than the non-response group (3.8: IQR :0.95) (*see Table 2.1*).

Table 2.1: Characteristics of study participants

Variables	Study cohort n=124	No-responses n=180	*P* value
Age, median (IQR)	14.98 (1.58)	15.53 (3.27)	.097
Sex, n (%) **Female** **Male**	 106 (85.4) 18 (14.5)	 156 (86.6) 24 (13.3)	.467
Major Cobb angle, median (IQR)	63˚ (22˚)	64.35˚ (28.75˚)	.769
Lenke classification, n (%) **Lenke 1** **Lenke 2** **Lenke 3** **Lenke 4** **Lenke 5** **Lenke 6**	 70 (56.5) 1 (.8) 17 (13.7) 2 (1.6) 31 (25) 3 (2.4)	 93 (51.7) 7 (3.9) 34 (18.9) NA 43 (23.9) 3 (1.7)	.200
Procedure, n (%) **Posterior approach** **Anterior approach** **Anterior and posterior approach**	 115 (92.7) 7 (5.6) 2 (1.6)	 159 (88.3) 18 (10) 3 (1.7)	.396
Length of hospital stay, Median (IQR)	7(3)	7 (3)	.889
SRS-22r pre-surgery scores, median (IQR) **Function** **Pain** **Self-Image** **Mental Health** **Satisfaction** **Sub-total** **Total**	 4 (1.2) 3.6 (1.4) 2.8 (1) 3.8 (1.4) 3.5 (1.5) 3.6 (1.2) 3.5 (1.04)	 3.8 (.95) 3.6 (1.2) 2.8 (1) 3.7 (1.4) 3.5 (1) 3.4 (.89) 3.5 (.85)	 .037 * .511 .682 .511 .218 .182 .532

n indicates sample size; IQR, Interquartile Range, SRS-22r, scoliosis research society questionnaire-22 revised, * significant at the 0.05 level (2-tailed)

2.14.1 Relevant changes at the course of clinical outcomes

A graphical representation of the distribution of the SRS-22r scores at different time points are presented in the figures 2.1A to 2.1 G. The descriptive values of SRS-22r scores for each time point are presented in *Table* 2.2.

At 6 weeks following-surgery, function, pain, and mental health scores decreased, compared to the pre-surgery (baseline) scores. However, self-image and satisfaction scores improved immediately following surgery. The SRS-22r scores significantly improved up to 2 years follow up. Pain scores were slightly decreased at 1 year but then improved at 2 years.

Overall, most of the SRS-22r domains reach or exceed the SDC at 1 year except function and pain scores. The function scores increased but did not exceed the SDC value (+0.19), while the pain scores decreased by (- 0.12). At 2 years, pain and function improved by (+ 0.53), (+0.3), respectively. The highest improvement was in self-image (+1.26) and satisfaction (+1) domains, while the least improvement was in the function domain (+0.3). (*See Table 2.2*).

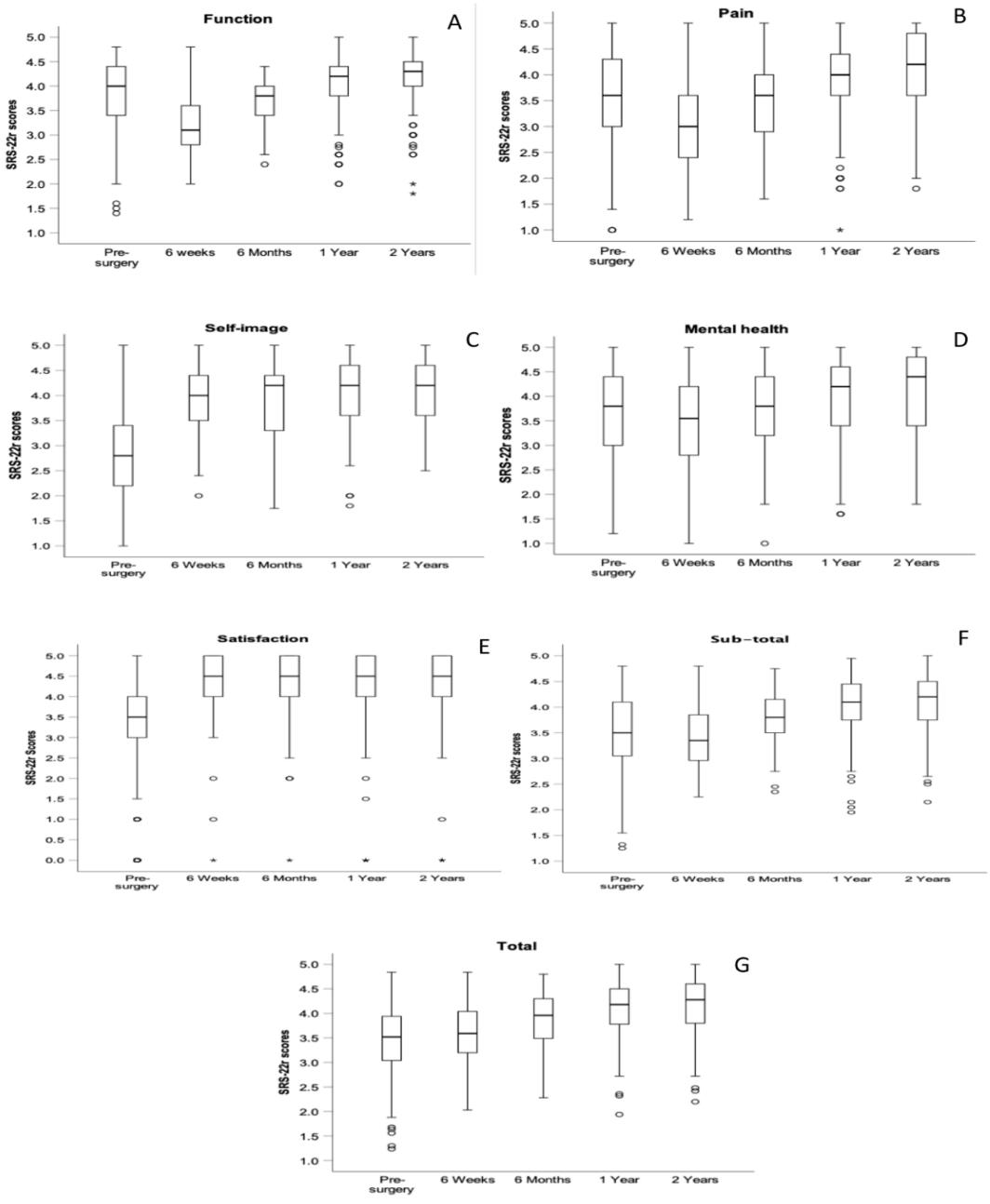


Figure 2.1: Boxplots showing distribution of the SRS-22r scores of individuals with AIS at five time points.

Table 2.2: Descriptive values of SRS-22r domain scores per time points

SRS-22r scores Mean (SD)	Baseline	6 weeks	6 months	1 year	2 years	Changes at 1 year	Changes at 2 years
Total	3.44 (.69)	3.59 (.54)	3.84 (.59)	4.07 (.54)	4.12 (.58)	0.63	0.68
Subtotal	3.49 (.73)	3.42 (.67)	3.75 (.56)	4.01 (.55)	4.08 (.56)	0.52	0.59
Function	3.83 (.70)	3.18 (.63)	3.67 (.53)	4.02 (.56)	4.13 (.58)	0.19	0.3
Pain	3.57 (.93)	3.03 (.87)	4.01(.77)	3.45 (.87)	4.1 (.76)	-0.12	0.53
Self-image	2.81(.82)	3.93 (.68)	4.04 (.70)	4.02 (.70)	4.07 (.68)	1.21	1.26
Mental health	3.6 (.91)	3.49 (.93)	3.80 (.83)	4 (.80)	4.07 (.84)	0.4	0.47
Satisfaction	3.3 (1.39)	4.27 (.88)	4.2 (1.01)	4.29 (.96)	4.3 (.93)	0.99	1

SRS-22r indicates Scoliosis Research Society-22r; SD standard deviation

2.14.2 Comparison analysis

Using the SDC values for each domain, fewer participants were assigned to the successful group than the unsuccessful group. No significant differences identified between the two groups when compared with respect with their general demographic and clinical characteristics except in age (p=.027) (*See Table 2.3*).

For all SRS-22r domains, the successful group had a low pre-surgery score, which is statistically different than the unsuccessful group. The difference between groups remains statistically significant at 1-year following surgery for all but the function and subtotal domains. The successful group achieved and exceeded the SDC value for all domains. In contrast, the unsuccessful group were unable to achieve the SDC values at any of the SRS-22r domains expect in the mental health domain (+0.4). Furthermore, the difference between the two groups in achieving the SDC values was also significant in all domains (*see Table 2.4*)

Table 2.3: General comparison between successful and unsuccessful groups characteristics

Demographic & clinical characteristics	Successful ≥ SDC n=33	Un-successful < SDC n=91	*P* value
Age (mean, SD)	15.4 (1.2)	14.8 (1.2)	.027*
Sex, n (%)	27 (81.8)	79 (86.6)	.485
Major Cobb angle, median (IQR)	65.5 (19.3)	66.2 (17.2)	.850
Lenke classification, n (%) **Lenke 1** **Lenke 2** **Lenke 3** **Lenke 4** **Lenke 5** **Lenke 6**	 22 (66.6) 0 1 (3) 2 (6) 8 (24.4) 0	 48 (52.7) 1 (1.1) 16 (17.5) 0 23 (25.2) 3 (3.2)	.371
Procedure, n (%) **Posterior approach** **Anterior approach** **Anterior and posterior approach**	 32 (96.9) 1 (3) 0	 83 (91.2) 6 (6.5) 2 (2.1)	.507
Length of hospital stay, median (IQR)	7(3)	7(3)	.854

SDC indicates the Minimal Detectable Minimal Difference, n sample size; IQR, Interquartile Range,

* Significant at the 0.05 level (2-tailed)

Table 2.4: Comparison analysis between successful and unsuccessful groups of SRS-22r domain scores

SRS-22r domain	Successful ≥ SDC n (%) mean (SD)	Unsuccessful < SDC n (%) mean (SD)	*P* value	SDC
Function **Pre-surgery** **1 year** **Difference**	33 (26.6) 3.2 (0.7) 4 (0.6) 0.8	91 (73.3) 4.1 (0.5) 3.9 (0.5) 0.2	<.001** 0.7 <.001**	0.24
Pain **Pre-surgery** **1 year** **Difference**	51 (41.1) 3.1 (0.8) 4.2 (0.6) 1.1	73 (59.3) 3.9 (0.9) 3.7 (0.9) -0.2	<.001** .003** <.001**	0.3
Mental health **Pre-surgery** **1 year** **Difference**	44 (29.6) 3.19 (0.7) 4.17 (0.7) 0.9	54 (70.4) 4.1 (0.7) 3.7 (0.8) 0.4	<.001** .004** <.001**	0.27
Self-image **Pre-surgery** **1 year** **Difference**	76 (81.7) 2.7 (0.8) 4.1 (0.6) 1.4	17 (18.3) 3.4 (0.8) 3.3 (0.6) -0.1	<.001** <.001** <.001**	0.3
Subtotal **Pre-surgery** **1 year** **Difference**	59 (63.4) 3.2 (0.7) 4.0 (0.5) 0.8	39 (41.9) 3.9 (0.6) 4.1 (0.5) 0.2	<.001** 0.32 0.03	0.23

SDC indicates the Minimal Detectable Minimal Difference, n: sample size, SRS-22r: Scoliosis Research Society-22r, SD: standard deviation,

*** Significant at the 0.05 level (2-tailed)**

**** significant at the 0.01 level (2-tailed)**

2.14.3 Predictors of relevant changes in function and pain outcomes at 1 year

Univariate analysis revealed a significant but low association between function scores and age. However, a significant but negligible association was found between function scores and length of hospital stay. A significant negative moderate association was found between function scores at 1-year and function pre-surgery scores. However, a significant but low negative association was found between function scores and pain, self-image, and mental health pre-surgery scores.

Whilst for pain, there was a statistically significant low association between pain scores at 1-year with sex and age. Further, significant negative low associations were found between pain scores at 1-year and pre-surgery scores for pain, function, self-image, and mental health domains (*see Table 2.5*).

Table 2.5 : Univariate analysis of factors associated with pain and function at 1 year follow up

Variables	Function n=124		Pain n=124	
	Correlation coefficient	*p* value	Correlation coefficient	*p* value
Sex	.074	.785	.452	.033*
Age	.222	.014*	.337	<.001**
Major Cobb angle	.005	.957	-.041	.665
Length of hospital stay	-.003	.927**	.071	.440
Function (pre-surgery)	-.554	<.001**	-.215	.016*
Pain (pre-surgery)	-.431	<.001**	-.462	<.001**
Self-image (pre-surgery)	-.365	<.001**	-.275	.002**
Mental health (pre-surgery)	-.348	<.001**	-.292	<.001**

Dependent variables were dichotomized into successful and unsuccessful using SDC value of pain (0.3) and function (0 .24)

***Correlation is significant at the 0.05 level (2-tailed)**

****Correlation is significant at the 0.01 level (2-tailed)**

Logistic regression analysis was performed to ascertain the effects of these factors on the likelihood that individuals with AIS reach the SDC value at 1 year follow-up. The model explained 44.7% (Nagelkerke R2) of the variance in function scores at 1 year and correctly classified 83% of cases. Older adolescents are 1.6 times more likely to achieve SDC value when compared to younger adolescents. While the chance of reaching the SDC in function at 1 year is decreased by .105 with each increase in function pre-surgery scores (OR: .105, 95% CI:.045-.245) (*p*=.001*) (*See Table 2.6*).

Table 2.6: Multiple logistic regression analysis for factors predicted function outcome† at 1-year follow-up

Backward selection method (final model)	**b (SE)**	***p***	**95 % confidence interval for Exp (b)**		
Independent factors			**Exp (b)**	**Lower**	**Upper**
Age	.488 (.224)	.029 *	1.630	1.051	2.227
Function(pre-surgery)	-2.255 (.432)	<.001**	.105	.045	.245

† Dependent variables were dichotomized into successful and unsuccessful using SDC value for function (0 .24)
R2 = .578 (Hosmer and Lemeshow), .238 (Cox and Snell), .447 (Nagelkerke)
*** p< 0.05**
****p<.001**

The logistic regression model for pain explained 52.6 % (Nagelkerke R2) of variance in pain scores and correctly classified 82% of cases. Sex, age, length of hospital-stays and pre-surgery scores for function and pain were significantly associated with achieving SDC values.

Male individuals with AIS are 4.5 times more likely to achieve the SDC compared to females (OR: 4.5, 95% CI: 1.10 – 18.5). Similar to the function model, older age individuals are 2.7 times more likely to achieve SDC than younger ages (OR:2.75, 95% CI: 1.68 – 4.5). Individuals with AIS who spent more days at hospital are 1.13 more likely to achieve the SDC following-surgery (OR:1.13, 95% CI: 1.005 – 1.269). The chance of reaching the SDC for pain decreased by 0.116 with each increase in pain scores pre-surgery (OR:0.116, 95% CI: .047 – .289), while the chance is increased by 3.5 with every increase in the function score pre-surgery (OR:3.531, 95% CI: 1.33 – 9.3) (see Table 2.7)

Table 2.7: Multiple logistic regression analysis for factors predicted pain outcome† at 1 year follow up

Backward selection method (final model)	**b (SE)**	***p***	**95 % confidence interval for Exp (b)**		
Independent factors			**Exp (b)**	**Lower**	**Upper**
Sex	1.510 (.719)	.036 *	4.528	1.106	18.532
Age	1.012 (.251)	<.001 **	2.750	1.680	4.502
Length of hospital stay	.122 (.059)	.040 *	1.130	1.005	1.269
Function (pre-surgery)	1.262 (.498	.011*	3.532	1.332	9.366
Pain (pre-surgery)	-2.150 (.463)	<.001**	.116	.047	.289

† Dependent variables were dichotomized into successful and unsuccessful using SDC value for pain (0 .3)

R2 = .107 (Hosmer and Lemeshow), .390 (Cox and Snell), .526 (Nagelkerke)

*** P < 0.05**

****p<.001**

2.15 Discussion

This study evaluated the relevant changes in individuals with AIS outcomes following-surgery at both short term and long term. Function, pain, and mental health scores declined while self-image and satisfaction improved immediately following surgery. All the SRS-22r domains improved from 6 months up to 2 years. The highest improvement was in self-image domain at 1 year (+1.2), and the least improvement was in function (+.19). Pain was the only domain that decreased by (-.12) at 1 year, then subsequently improved by (+.53) at 2 years.

In general, fewer numbers of individuals with AIS were successfully reached or exceeded the SDC values at 1 year for most of the SRS-22r domains. The successful group had low scores at all SRS-22r domains pre-surgery compared to the unsuccessful group as well as achieved the SDC values at all domains. Both groups were statistically different following-surgery at all SRS-22r domains except at function and subtotal domains.

Increasing age, and having low function scores pre-surgery, were predictive factors for achieving the SDC in the function domain following-surgery. While, increasing age, length of hospital stays, being male, and having high scores in the function domain and low scores in the pain domain pre-surgery, increased the likelihood to reach SDC at pain following-surgery.

2.15.1 Relevant changes at the course of clinical outcomes

Spinal fusion surgery requires muscle dissection and spinal immobilization through rigid internal fixation (Madera et al., 2017). Thus, the decrease in functional scores seen immediately post-surgery could be reasonably attributable to post-surgery pain, healing, and reduced spinal mobility (Kakar et al., 2017, Fabricant et al., 2012). The SRS-22r is a widely used PROM for evaluating HRQOL in AIS as well as in adult populations (Asher et al., 2006). The functional domain of this questionnaire had a high ceiling effect that may limit its ability to detect changes (Asher et al., 2006). In this study, the function domain was the farthest domain from achieving the SDC, with increases (+0.2) only at 1 year and (+0.3) at 2 years compared to the other domains. The function was also the least sensitive domain to change in previous studies, attributed to the fact that AIS surgery is mainly cosmetic rather than functional impairment (Kelly et al., 2019). In contrast to this study, the function domain had the highest improvement (+ 1.2) at the years follow up (Mens et al., 2022), in which the pre-surgery score was lower than that in this study cohort (median 3.8) compared to (median 3.2)

(Mens et al., 2022). Although the functional domain of the SRS-22 has been refined to enhance its internal consistency (Asher et al., 2006), true changes may not be detected, and this could be related to the lack of content validity or the ceiling effect (De Vet et al., 2011). Ceiling effects can make a PROM insensitive to change, as it cannot detect improvements beyond a questionnaire width (Streiner et al., 2015). Therefore, a PROM that is sensitive to change near the ceiling effects is needed.

Pain was the only domain that worsened at 1 year compared with the other SRS-22r domains. This can be potentially explained by baseline risk factors such as anxiety and pain, which may lead to persistent pain post-surgery (Bailey et al., 2021). Surgery has also been reported as the leading cause of pain (Wong et al., 2007), as well as inadequate postoperative pain management (Seki et al., 2018, Fletcher et al., 2015). The literature suggests that some adolescents develop new-onset pain post-surgery, (Bastrom et al., 2013) or experience moderate pain 1-year post-surgery (Bailey et al., 2021). This further supports the need for a better understanding of the factors that influence pain.

Self-image showed the highest change at 1-year following surgery. These findings are consistent with those of other studies, where the perceived self-image improved immediately following surgery (Carrasco and Ruiz, 2014), indicating that AIS is principally focused on back shape and spinal deformity (Sanders et al., 2010). As a result, it is important to manage any misconceptions and unrealistic expectations about the surgery, with those who at risk of post-surgery dissatisfaction, because of their pre-surgery self-image. In this case, they may need additional support to cope with negative emotions associated with self-image.

Poor mental health scores following surgery might be explained through symptoms of depression, isolation and the impact of spine deformity on their mental health domain (Fernandes et al., 2019). A study showed that patients with AIS (n=685) had more suicidal thoughts, concerns about body image and development, and relationships compared to their peers (Payne et al., 1997).

Therefore, early intervention (pre-16 years) has shown an improvement in the psychological well-being of adolescents, supporting the recommendation that surgery should not be delayed, especially in those with poor mental health scores (Andersen et al., 2002).

Participants were satisfied with their management at all time points, with this domain having the highest score (5). With a high ceiling effect in surgically managed participants and among young patients (Parent et al., 2007), this domain likely needs some revisions for those managed with other approaches.

2.15.2 Comparison analysis

A small number of those with AIS successfully reached the SDC at 1 year at most of the SRS-22r domains. Pre-surgery scores for the successful groups, were lower and statistically different than to those in the unsuccessful groups at all domains. Bago et al. reported consistent results for older AIS (mean age 18.1 years) using the MCID of the subtotal score. The successful group had poorer pre-surgery scores and significantly improved compared to the un-successful group (Bago et al., 2012). A defect in the SRS-22 that causes the high ceiling effects (over 20%) was the attributed cause for this difference in achievement of scores (Bago et al., 2012). Thus, reassessment of this measurement instrument is needed to ensure the true change in patients score is detected. Findings implies that the unsuccessful group had minimal potential for clinically meaningful improvement. Thus, it is essential for surgeon to discuss surgery's objectives with their patients and, managing expectations which may lead to different treatment strategy.

2.15.3 Predictors of relevant changes in function and pain outcome at 1 year

In this study, poor function scores pre-surgery predicted the attainment of SDC values in the long-term. Consistent findings showed that inactivity pre-surgery predicted reaching the MCID post-surgery (Raad et al., 2018). Factors such as a large curve size >70° (Tarrant et al., 2014), and functional limitation (Sieberg et al., 2013), are associated with poor preoperative scores.

Age was a significant predictor of SDC pain and function. Previously shown older patients with AIS had poor pre-surgery scores as well as more back pain and cosmetic concerns (Sanders et al., 2010, Rodrigues et al., 2017). Those who underwent surgery at a later stage of their adolescence had more pain than younger patients with AIS (Rodrigues et al., 2017), and young age could be a protective factor (Sieberg et al., 2013). Our results support previous suggestions of revision of SRS-22r since the ceiling effect is reported more with young AIS, which limits the questionnaire discriminative ability (Parent et al., 2010).

The length of hospital stays predicted attainment of SDC at pain scores. Increased hospital stay was associated with an increase in the amount of instrumentation and operation time (Basques et al., 2015, Danielsson et al., 2006), and was reduced in those who had an enhanced recovery program after surgery (White et al., 1999). In contrast, a shorter hospital stay was associated with being active and having better preoperative function scores (Raad et al., 2018).

Male sex also predicted the SDC value for pain post-surgery. Males are known to have higher functional requirements and expectations than females, which may explain their poor pre-surgery

scores. In contrast, females had better outcomes in terms of function and self-image after surgery (White et al., 1999).

2.16 Strength and limitations

The main strength of this study is the use of SDC value to evaluate real clinical benefit of surgery for individuals with AIS, in both short and long terms. Although it is the smallest value that can detect change, majority of patients fail to reach this value. Individuals with AIS, who had a high pre-surgery score would not be benefit from surgery based on change in their scores. Highlighting an inherit problem in the SRS-22r (ceiling effects) that require further investigations.

The SRS-22r has no actual cut-off point to accurately classify those with AIS into successful and unsuccessful groups. In this study, the SDC has been used as cut off point to classify patients based on the achievement of clinical change (Fernandes et al., 2019, Kelly et al., 2019, Carreon et al., 2010). The SDC is more likely to be consistent in individuals with AIS who have had surgery, as there is no evidence that it will differ between surgical and non-surgical or sub-surgical populations (van Kampen et al., 2013). Different approaches have been used previously to stratify patients by using for example activity question within the SRS-22r, to classify patients into active and inactive groups (Raad et al., 2018), or by using MIC of the SRS-22 subtotal score to classify patients into successful and unsuccessful groups (Bago et al., 2012). Future studies may estimate the cut-off score of the SRS-22r, such as SDC value, to enable discrimination between AIS, which would be clinically meaningful.

The main limitation of this study is the loss at follow up, which may introduce bias into the results. This study is a registry-based study with a robust reminder in place, however, the follow up rate was only 41%. Previous studies have reported similar poor response rate (Cunningham et al.,

2020, Jasani et al., 2016, Wang et al., 2020). This was attributed to several reasons such as patients may feeling well and not requiring evaluation, or they found alternatives treatment, and thought they are not obligated to complete questionnaires. Also, it due to low compliance which may be related to the design and content of the PROM that used to collect data (Cunningham et al., 2020). A comparison between study cohort and those lost at follow up revealed no difference in their pre-surgery characteristics, Therefore, findings of this study are therefore valid and the sample representative to individuals with AIS treated at this spine centre.

Although preoperative self-image is unchangeable, it undergoes significant transformations after surgery, particularly evident at the one-year follow-up. This highlights that individuals with AIS place more attention on their back shape and spinal deformity. Surgeons can use this information to help manage patients' expectations, discussing realistic expectations and potential misconceptions to help align surgical outcomes with patient needs. Additionally, the study findings may help surgeons identify those individuals at higher risk for post-operative dissatisfaction based on pre-operative self-image. This may facilitate implementation of targeted support to enhance coping behaviours and improve overall patient satisfaction and surgical outcomes.

In this study, those with AIS with low scores pre-surgery attained the SDC. This result was attributed to the ceiling effects of SRS-22 that may limit its sensitivity to change. Future studies should address this shortcoming of the SRS-22 and evaluate questions within the questionnaire that cause ceiling effects and may limit discriminative ability (Bago et al., 2012).

2.17 Chapter summary

The purpose of this chapter was to evaluate the changes in surgical outcomes for individuals with AIS using the SDC values of the SRS-22r questionnaire at short term and long-term follow-up

periods. The chapter also aims to compare individuals who achieved SDC in the pain and function domains at 1 year following surgery with those who did not, and to identify factors that influence the achievement of SDC in these domains. Participants were classified into "successful" and "unsuccessful" groups based on whether their change in scores exceeded the SDC value or not. The differences between these groups were assessed using statistical tests, and factors influencing the achievement of SDC were analysed using univariate and logistic regression analysis. The results showed that most SRS-22r domains, except self-image and satisfaction, decreased at 6 weeks following surgery. However, at 1 year, self-image showed the greatest improvement, followed by function, while pain decreased slightly. The successful group had lower pre-surgery scores and was statistically different from the unsuccessful group in all domains, except function and subtotal domains. Age and low pre-surgery function scores were associated with achieving SDC in the function domain at follow-up. Achieving SDC in the pain domain was significantly associated with age, gender, length of hospital stays, low pre-surgery pain scores, and high pre-surgery function scores. The self-image domain showed the largest improvement compared to other domains, indicating the importance of addressing individuals body image concerns. The study also highlighted the need for a better measurement instrument that is sensitive to change and can accurately detect improvements in function, especially near the ceiling effect.

Chapter Three

PHYSICAL FUNCTIONING IN ADOLESCENT WITH IDIOPATHIC SCOLIOSIS: A SYSTEMATIC REVIEW OF OUTCOME MEASURE AND THEIR MEASUREMENT PROPERTIES

This chapter reports in full the contents of a published manuscripts by the thesis author protocol (Alamrani et al., 2020) and research article (Alamrani et al., 2021). It includes verbatim text from the published manuscripts and some changes in the introduction were employed for the purpose of this thesis.

Publications and Conference presentations

- **Alamrani, S.,** Rushton, A. B., Gardner, A., Bini, E., Falla, D., & Heneghan, N. R. (2021b). Physical functioning in Adolescents with Idiopathic Scoliosis: A Systematic Review of Outcome Measures and Their Measurement Properties. Spine (Phila Pa 1976). doi:10.1097/brs.0000000000003969 (**Appendix 4**)
- **Alamrani, S.,** Rushton, A., Gardner, A., Falla, D., & Heneghan, N. R. (2020). Outcome Measures evaluating Physical functioning and their measurement properties in adolescent idiopathic scoliosis: a protocol for a systematic review. BMJ Open, 10(4), e034286. doi:10.1136/bmjopen-2019-034286 (**Appendix 5**)
- **Alamrani, S.,** Rushton, A. B., Gardner, A., Bini, E., Falla, D., & Heneghan, N. R.(2020). Physical functioning in adolescents with Idiopathic Scoliosis: A Systematic Review of Outcome

Measures and Their Measurement Properties. World Physiotherapy Congress online. 9-11th April 2021. (Platform presentation).

- **Alamrani, S.,** Rushton, A. B., Gardner, A., Bini, E., Falla, D., & Heneghan, N. R. (2020). Measurement properties of Outcome measures evaluating Physical functioning in adolescents with idiopathic scoliosis : A systematic review. Physiotherapy UK 13-14th November 2020. Birmingham. United Kingdom. (Poster presentation).

- **Alamrani, S.,** Rushton, A. B., Gardner, A., Bini, E., Falla, D., & Heneghan, N. R. (2020). Measurement properties of Outcome measures evaluating Physical functioning in adolescents with idiopathic scoliosis : A systematic review. 4th Saudi Spine Society annual conference. 7-9th November 2020. (Platform presentation).

3.1 Abstract

One of the important aspects in HRQOL is PF, being the ability to perform physical activity of daily livings. Different measures are suggested to evaluate PF for individuals with AIS. Yet, no systematic review has evaluated the measurement properties of such measures in individuals with AIS. This systematic review followed PROSPERO registered (CRD42019142335), and published protocol (*see Appendix 5*), to identify measures used to assess PF in individuals with AIS, and then to evaluate their measurement properties.

A two-staged search strategy were performed on electronic databases from inception up to September 2022. Search one revealed a list of measures which was used in search two to identify studies that evaluate measurement properties of those measures. Two independent reviewers determined study eligibility, assessed risk of bias using COSMIN checklist, performed data extraction, and assessed the quality of evidence using modified Grading of Recommendations, Assessment, Development and Evaluation (GRADE) approach.

Twenty-eight PROMs, 20 PBOMs, and 10 body structure and function measures were identified in search one. Search two revealed 20 study of PROMs, 1 study for PBOMs, and 3 for body structure and function measures. Construct validity, reliability, and responsiveness of most measures has been established in individuals with AIS, but not the content validity or internal consistency (moderate quality of evidence). Construct validity was sufficient for the Timed Up and Go test and body structure and function measures (very low to low quality of evidence).

Currently, PF is evaluated with a variety of measures in individuals with AIS. Based on COSMIN methodology, none of the measures identified in this review can be recommended with

confidence for evaluating PF in individuals with AIS. High-quality research is required to establish measurement properties of PF outcome measures in individuals with AIS.

3.2 Introduction

As stated in *Chapter One, sections 1.41.6,* PF is one of the important domains of HRQOL and one of the core outcome domains that should be assessed and reported in clinical trials (Williamson et al., 2012, Dodd et al., 2018). It is known as the physical ability to perform physical activities of daily living ranging from performing simple activity i.e., walking, to complex actions i.e., self-care (Painter et al., 1999, Dodd et al., 2018). Individuals with AIS may experience limitations in their functional ability including difficulties in movement, maintaining body position, and participating in activities requiring strength and endurance (Du et al., 2016a). Research have shown reductions in spinal mobility, lumbar movement, and muscle endurance among individuals with AIS who have undergone operative or conservative management compared to healthy controls (LaMontagne et al., 2004, Rullander et al., 2013, Rullander et al., 2017, Danielsson et al., 2006, Wilk et al., 2006, Aghdasi et al., 2020). Thus, assessment of PF is necessary to identify those who are at risk of disability and to predict the future use of health and social care (Tomey and Sowers, 2009, Painter et al., 1999).

As discussed in *Chapter one*, *section* 1.5, the PF can be assessed by PROMs, PBOMs and body structure and function measures (Tomey and Sowers, 2009). The radiographs and curve angle measurement are the gold standard for assessment of effectiveness of interventions for AIS (Joarder et al., 2023). The radiographs can give an indication about dysfunctions in structure but fail to fully capture functional limitations (Reiman and Manske, 2011). While the SRS-22r is the reference standard and recommended PROM for assessment of PF for individuals with AIS per COS study (de Kleuver et al., 2017). However, the COS study included all forms of spinal deformities (de Kleuver et al., 2017), and the heterogeneity of the population limits applicability to individuals with AIS as a discrete population. Although PROM are relevant, it should be used cautiously, as it influenced by

patients' perception of their abilities to perform activities and lack sensitivity to change (Reiman and Manske, 2011). On the other hand, measures such as PBOMs eliminate the self-report confusion and determine both the ability and capacity of what an individual can perform (Painter et al., 1999).

Adequate measurement properties of OMs are important to avoid the risk of bias and ensure accuracy in the evaluation of test results (Mokkink et al., 2018a). As stated in *Chapter One, section 1.4,* the COSMIN group developed a taxonomy of measurement properties to improve the selection of OMs, and they provide guidelines for conducting a systematic review for PROM, which can be adapted for other OMs (Mokkink et al., 2018b).

While many variants of SRS are available i.e., SRS-7, SRS-23, SRS-30 and SRS16, there is a lack of unified all-encompassing PROM which prevents data pooling and synthesis. This leads to avoidable inefficiencies in the research production and reporting, along with difficulties in effectively using research findings (Chalmers and Glasziou, 2009). To address this issue, a critical evaluation of the psychometric properties of different PROMs is necessary to determine the most appropriate options while ensuring it's have sufficient measurement properties for this specific population (Mokkink et al., 2018b, Mokkink et al., 2018a, Matza et al., 2004, Patrick et al., 2011b).

In the absence of existing evidence, this chapter present a systematic review with a primary objective to identify OMs used to assess PF in individuals with AIS, and secondary objective to summarize their measurement properties. Review findings can inform clinicians and researchers on the best available tools for the assessment of PF in individuals with AIS. Furthermore, findings will inform future research drawing on a range of measures of PF to investigate HRQOL in individuals with AIS.

3.3 Methods

3.3.1 Protocol and Registration

A two-staged systematic review was conducted according to a registered (PROSPERO: CRD42019142335) and published protocol (Alamrani et al., 2020). Reported in line with the Preferred Reporting Items for Systematic Review and Meta-Analysis (PRISMA) statement (Page et al., 2021). This review followed COSMIN guidelines for conducting systematic review of PROMs (Prinsen et al., 2018).

3.4 Search one: Inventory of outcome measures

Search one was performed to identify studies that used any type of OMs to assess PF in individuals with AIS. The aim of this search was to have a greater insight of the literature around OMs used for evaluating PF of individuals with AIS.

3.4.1 Eligibility criteria

3.4.1.1 Study design

To identify OMs used for assessment of PF of individuals with AIS, all study designs were included. These being randomised clinical trials, cohort studies, cross-sectional studies, and case-control studies. At this stage, no limitations were applied on the type of OM, language, or location of the study. Since the aim of this search is to identify all types of OMs, the search did not include any search terms about measurement properties (Mokkink et al., 2018b).

3.4.1.2 Participants

Studies that included participants diagnosed with AIS were included, aged 10 to 18 years old, had a diagnosis of scoliosis with unknown cause and curve size measured by Cobb method ≥10 ° (Konieczny et al., 2013). No restrictions were applied to the curve severity, evaluation settings and management.

3.4.1.3 Outcome

The outcome for this systematic review was the OM used to evaluate PF, which is the ability to perform simple task/activity e.g. walking, or perform complex actions e.g. self-care (Painter et al., 1999), assessed by one of the following type of OMs :

- Self-report measures in the form of questionnaires or scales (Reiman and Manske, 2011), or subscale within the questionnaire designed to evaluate PF and used for AIS and/or
- Performance-based OM to evaluate PF assessed by clinician/observer while an individual is performing a functional task, measured by time or distance, e.g. sit to stand test (Beaudart et al., 2019) and/or
- Body structure and function measures defined as measure of the physiological function of body system i.e. muscle endurance or/and the anatomical parts of the body e.g. range of motion (Tomey and Sowers, 2009).

3.4.2 Information sources

An electronic search of databases was conducted including MEDLINE (1946–November 2019), PsycINFO (1967–December 2019) and EMBASE (1974–December 2019) through OVID

interface, CINAHL (1937– December 2019) and SPORT discus (1800–December 2019) through EBSCO interface, Web of Science (1900– December 2019) and PubMed (1997–December 2019). The Web of Science database was searched for conference proceedings for the last 5 years. As well as an electronic search on the key journals including Spine, the Spine Journal, Spine Deformity, Scoliosis and Spinal Disorders and European Spine Journal. Further, searching for the grey literature, including British National Bibliography for report literature, open-grey, dissertation abstracts and Electronic Thesis Online Service (EThOS) was conducted.

3.4.3 Search strategy

A comprehensive and systematic search was completed by SA with a support and consultation from librarian. To ensure that all relevant studies were included, the type of the OM was not specified at this stage. Initial search terms were developed for MEDLINE and then adapted with relevant syntax and subject headings for the other databases. Terms and keywords used for this search is presented in the published protocol (Alamrani et al., 2020), and in the (see *Appendix* 7)

3.4.4 Data management

Search records were imported into Endnote V.X9 (Philadelphia, PA: Clarivate). Using Endnote, the abstracts and full texts were stored, exact duplicates were identified and removed. Two independent reviewers (SA & EB) assessed studies based on the title and abstract for eligibility. In case of insufficient information, full text articles were retrieved and screened for eligibility. Both reviewers screened all records independently. The two reviewers discussed findings and reached consensus on eligibility of studies. Third reviewer (NRH) was consulted in case of disagreement.

The percentage of agreement between reviewers was estimated using the k statistic (Belur et al., 2018) (IBM SPSS Statistics for Windows, Version 25.0. Armonk, NY: IBM Corp).

3.4.5 Data collection process

The data was extracted from eligible studies using a standardised form by (SA) and reviewed by the second reviewer (EB). The information was based on the first aim of review; to identify OM used to assess PF in AIS. Corresponding authors were contacted to request further details when information is not clear or unavailable in the studies. A second and final reminder was then sent two weeks apart. If no response was received after 1 month, the study was excluded.

3.4.6 Data items

Information from the included studies regarding type of OM and area of assessment were extracted from each study by SA and then reviewed by EB. An inventory for OMs that has been used to evaluate PF for individuals with AIS was created, and then classified according to its type (PROM, PBOM, Body structure and function).

3.5 Results

3.5.1 Study characteristics

The PRISMA flow diagram shows results of searching databases for search one of this review, selection process and reasons for exclusion are presented in *Figure* 3.1. A total of 503 studies met the eligibility criteria, consisting of (n=58) OMs that has been used to evaluate PF for individuals with AIS. These OMs were then listed and classified into 28 PROMs, 20 PBOMs and 10 body structure

and function OMs. The PROMs list was classified into generic PROM when the questionnaire is not specifically designed for individuals with AIS, or disease specific when the questionnaire/scale is designed to individuals with AIS.

The ICF model (WHO, 2001) has been used to aid classification when the type of the measure is not clear. Some PBOM was classified as body function using the ICF, however, since the PBOM has been defined in this review as assessment of task or activity, those measures were assigned into the PBOM list. (See *Tables 3.1, 3.2 and 3.3* which present the lists of OMs identified in search one).

Different methods have been identified and used to evaluate PF such as e.g., radiographs, and laboratory based or anthropometric measures. These measures were excluded because it has been extensively investigated in literature (Prowse et al., 2016, Langensiepen et al., 2013, Navarro et al., 2019, Wade et al., 2013, Fong et al., 2010, Wu et al., 2020, He and Wong, 2018).

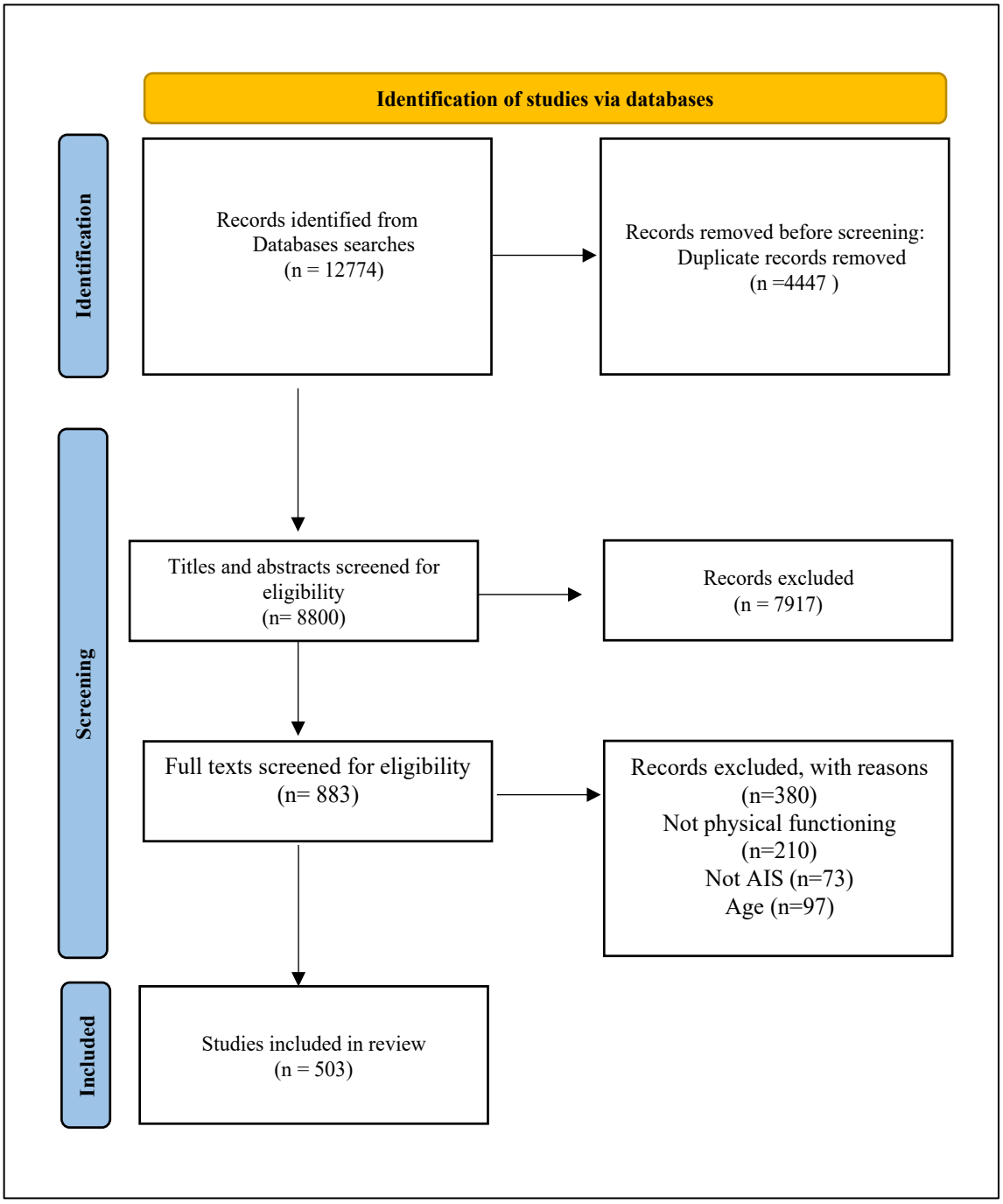


Figure 3.1:: PRISMA flow diagram for search one. Adapted from (Page et al., 2021)

Table 3.1: List of PROMs generated from search one

NO.	Generic PROM	NO.	Disease-specific PROM
1	Short Form Health Survey (SF-36)	**1**	Scoliosis Research Society (SRS-30)
2	Short-Form Health Survey (SF-12)	**2**	Scoliosis Research Society (SRS-24)
3	Oswestry disability Index (ODI)	**3**	Scoliosis Research Society (SRS-23)
4	Roland and Morris Disability Index (RMDQ)	**4**	Scoliosis Research Society (SRS-22)
5	Pediatric Patient-Reported Outcomes Measurement Information System (PROMIS)	**5**	Scoliosis Research Society-22 revised (SRS-22r)
6	Competence Scale of the Youth Self-Report and Profile	**6**	Scoliosis Research Society (SRS-18)
7	Paediatric Outcomes Data Collection Instrument (PODCI)	**7**	Scoliosis Research Society (SRS-7)
8	Paediatric Quality of Life Inventory (PedsQL)	**8**	Quality of Life Profile for Spinal Deformities (QLPSD)
9	Functional Rating Index (FRI)	**9**	Scoliosis Quality of Life Index (SQLI)
10	Hannover Functional Ability Questionnaire (HFAQ)	**10**	Brace Questionnaire (BRQ)
11	Patient Specific Functional Scale	**11**	Italian Spine Youth Quality of Life questionnaire (ISYQOL)
12	Paediatric Evaluation of Disability Inventory (PEDI)	**12**	Scoliosis Japanese Questionnaire-27
13	EuroQoL 5-dimension (EQ-5D)	**13**	Sports Activity Questionnaire (SAQ)
14	Activity Scale for kids	**14**	Scoliosis Research Society (SRS-16)
15	Child Health Questionnaire-CF87		

Table 3.2: List of PBOMs generated from search one

No	PBOMs	Area of assessment	ICF classification
1.	Short Physical Performance Battery	Gait speed, chair stand and balance tests to assess function and disability	d4 mobility
2.	Kraus-Weber Test (K-W test)	Strength and flexibility of key postural (core) muscles	d4 mobility
3.	Six Minutes' Walk Test	Gait / Aerobic Capacity	d4500 Walking short distances
4.	Two minutes' walk test	Gait / Aerobic Capacity	d4500 Walking short distances
5.	Shuttle Walk Test	Gait / Aerobic Capacity	d4500 Walking short distances
6.	Incremental Shuttle Walk Test	Gait / Aerobic Capacity	d4500 Walking short distances
7.	Timed Up and Go Test	Balance/ Functional Mobility/ Gait	d498 Mobility, other specified
8.	Single limb Hop test	Balance/ Strength /Functional Mobility	b235 Vestibular functions
9.	Y-Balance Test	Balance	b2351 Vestibular function of balance
10.	Sit and reach test	Flexibility	b710 Mobility of joint functions
11.	Romberg Test	Proprioception	b235 Vestibular functions
12.	Tandem (Sharpened) Romberg test	Balance	b235 Vestibular functions
13.	Limit of Stability Test	Dynamic postural stability/balance	b235 Vestibular functions/ b755 Involuntary movement reaction functions
14.	Sensory organization test	Dynamic postural stability/balance	b235 Vestibular functions/ b755 Involuntary movement reaction functions
15.	Berg Balance Scale	Balance	b755 Involuntary movement reaction functions
16.	Fukuda stepping test (Unterberger step test)	Balance/vestibular	b235 Vestibular functions
17.	Timed Unipedal stance test	Balance	b235 Vestibular functions/ balance
18.	Biering Sorensen test	Lumbar trunk muscle endurance	b740 Muscle endurance functions
19.	Non-dynamometric trunk performance tests	Lumbar trunk muscle endurance	b740 Muscle endurance functions
20.	Trunk endurance test	Lumbar trunk muscle endurance	b740 Muscle endurance functions

Table 3.3 : List of body structure and function measures generated from search one

No.	Body structure and function measures	Area of assessment / Purpose	ICF classification
1.	Modified Schober's Test	Lumbar ROM	b710 Mobility of joint functions
2.	Trunk-Pelvis-Hip Angle test	Mobility of lumbo-pelvic-hip complex	b710 Mobility of joint functions
3.	Side bending, axial rotation, flexion, extension of spine	Spinal ROM	b710 Mobility of joint functions
4.	Fingertip to floor distance	Flexibility	b7101 Mobility of several joints
5.	Shoulder Range of Motion	Shoulder Range of Motion	b710 Mobility of joint functions
6.	Temporomandibular Range of Motion	Temporomandibular Range of Motion	b710 Mobility of joint functions
7.	Hand grip strength	Muscle strength	b730 Muscle power functions
8.	Thomas test	Flexibility	b710 Mobility of joint functions
9.	Lasegue test/ Lasegue sign/ single leg rises test	Flexibility	b710 Mobility of joint functions
10.	Dega wall test	Flexibility	b710 Mobility of joint functions

Abbreviations: ICF: International Classification of functioning, disability, and health, ROM: Range of Motion

3.6 Search two: Measurement properties of OMs

Using lists of OMs generated form search one. A second search was conducted to identify studies that evaluated measurement properties of these OMs.

3.6.1 Eligibility criteria

3.6.1.1 Study design

Any study that has evaluated one or more measurement properties of the identified OMs in search one was included. Only full-text studies available in English were included. Conference abstracts were excluded due to the inability to effectively evaluate the quality of the study (Mokkink et al., 2018a).

3.6.1.2 Participants

Participants aged between 10 and 18 years of age, with a diagnosis of AIS and curve size (Cobb angle) ≥10° measured by cobb method (Konieczny et al., 2013) were included. Studies with mixed cohorts, >50% of participants should be diagnosed with AIS for the study to be included. When there is missing of information about the number of participants with AIS, authors of studies were contacted. Studies without original participant data (e.g., systematic review) were excluded.

3.6.1.3 Outcome

Any study that has evaluated any measurement properties of the measurement properties of the OMs identified in search one was eligible. The measurement properties have been defined and explained in detail at *Chapter One.* In short, reliability including (internal consistency, test–retest,

inter-rater and intra-rater), measurement error, validity including (content validity, structural validity or criterion validity), and responsiveness (Mokkink et al., 2010),

3.6.2 Exclusion criteria

Studies conducted in non-English speaking populations were excluded. Systematic reviews focusing on outcome measures were also excluded to prioritize original research. Studies providing normative data, which involved participants without scoliosis were excluded. Additionally, studies that provided indirect evidence on the measurement properties, such as assessing alternative tests or using the outcome measure solely for measuring the outcome, were excluded.

3.6.3 Information sources

Similar to search one, a comprehensive search was performed and adapted to each database format. Limited to English, humans, and full text availability. The search was conducted on the following databases from inception till September 2022. An electronic search of databases was conducted including MEDLINE (1946– September 2022), PsycINFO (1967– September 2022) and EMBASE (1974– September 2022) through OVID interface, CINAHL (1937– September 2022) and SPORTdiscus (1800– September 2022) through EBSCO interface, Web of Science (1900– September 2022) and PubMed (1997– September 2022). The Web of Science database was searched for conference proceedings for the last 5 years. As well as an electronic search on the key journals reference list and grey literature.

3.6.4 Search strategy

Using the list of OMs determined from search one, (SA) conducted the search. The search terms were consisting of the name of the OM/s, the individuals with AIS and the measurement properties. The recommended search filters specifically designed for retrieving articles on measurement properties was adapted and used at this search. The search terms were first developed for MEDLINE and adapted with relevant syntax and subject headings for the other databases (Terwee et al., 2009). It combined name of population, name/s of instrument/s, measurement properties and the exclusion filter (Mokkink et al., 2018b). Each category of OMs was searched separately i.e., PROM both generic and disease specific, PBOMs, and body structure and function measures. The details and examples of search strategy are listed in the (see *Appendix* 7).

3.6.5 Data management

Search records were imported into Endnote V.X9 (Philadelphia, PA: Clarivate). Abstracts and full texts were stored in Endnote. The duplicates were identified, and exact duplicates were removed through the Endnote software. All records were screened by two independent reviewers (SA & EB) and assessed for eligibility based on the title and abstract. Full text article was retrieved when there is insufficient information to determine study eligibility. The two reviewers discussed findings and reached consensus on eligibility of studies. Third reviewer (NRH) was consulted in case of disagreement. The percentage of agreement between reviewers was estimated using the k statistic (Belur et al., 2018) (IBM SPSS Statistics for Windows, Version 25.0. Armonk, NY: IBM Corp).

3.6.6 Data collection process

The data was extracted from eligible studies using a standardised form by (SA) and reviewed by the second reviewer (EB). The information was based on the second aim of review; to identify studies on measurement properties of OM that used to assess PF in AIS. Corresponding authors were contacted to request further details when information is not clear or unavailable in the studies. A second and final reminder was then sent two weeks apart. If no response was received after 1 month, the study was excluded.

3.6.7 Data items

At this stage of review, the data extracted from the included studies were related to the study and participants characteristics, outcome measures and measurement properties. Corresponding authors were contacted in case of missing information.

3.6.8 Risk of bias in individual studies

Two independent reviewers (SA, EB) assessed the risk of bias for all included studies. Any disagreement was resolved through discussion, and a third reviewer (NH) was consulted in case of disagreement. The COSMIN checklist for assessment of risk of bias and methodological quality in individual studies was used (Mokkink et al., 2018a), It was revised and specifically designed for use in systematic reviews of PROMs to evaluate studies on measurement properties (Mokkink et al., 2018a). However, COSMIN recommended it to adapt the checklist for other measures (Prinsen et al., 2018).

The methodological quality of each study for each measurement property was assessed separately (Mokkink et al., 2018b). Each measurement property is rated on a 4-item check- list as either: very good, adequate, doubtful or inadequate quality (Mokkink et al., 2018b). Then, the overall methodological quality of the measurement property was rated based on "the worst score counts principle" i.e. that the overall quality of the study for a specific measurement property is based on the lowest rating of any items in the standards' box (Mokkink et al., 2018b).

3.6.9 Data analysis and synthesis

The necessary homogeneity in studies results (study population, outcome measures, statistical analysis) were insufficient, thus meta-analysis was not performed. Therefore, a narrative synthesis was conducted according to COSMIN guidelines (Mokkink et al., 2018b). Two independent reviewers preformed the analysis, and third independent reviewer (NRH) was consulted in case of disagreement.

A summary of the steps that was performed in the analysis and the synthesis of the study results are presented below:

1. The COSMIN risk of bias checklist was completed for each individual study

2. Results from developmental studies and any associated content validity studies was rated against the 10-established criteria for evaluating content validity (relevance, comprehensiveness, comprehension). The overall results of each aspect was rated against the updated criteria for good measurement properties as sufficient (+), insufficient (–) or indeterminate (?) (Terwee et al., 2018b).

3. Results of other measurement properties (e.g. reliability) of each study was also rated against the updated criteria for good measurement properties as sufficient (+), insufficient (–) or indeterminate (?) (Mokkink et al., 2018b).

According to COSMIN guidance, a set of hypotheses was predefined to evaluate the methodological quality of studies that evaluated both construct validity (including both convergent and discriminative validity), and responsiveness (Mokkink et al., 2018b). These hypotheses are used to determine the degree that scores of measurement properties are consistent with the predefined hypotheses (Prinsen et al., 2018b).

- Correlation would be found between instrument measure similar constructs (i.e., PF/activity/function) (≥0.50)
- Correlation of scores of OMs measuring related, but dissimilar construct would be lower (0.3–0.5).
- Correlations with instruments measuring unrelated constructs would be <0.3. (Abma et al., 2016).

4. The pooled overall results was then synthesised for each outcome measure and per measurement property and, rated against the established criteria (Mokkink et al., 2018b). The pooled or summarized evidence was rated as sufficient when at least 75% of the results met the criteria (Mokkink et al., 2018b).

3.6.10 Confidence in cumulative evidence

The quality of the overall evidence was graded using a modified Grading of Recommendations Assessment, Development and Evaluation (GRADE) approach. (Mokkink et al.,

2018b). Two independent reviewers evaluated each summarised result of each measurement property per outcome measure in each OMs category. Quality of evidence was rated either high, moderated, low, and very low, using risk of bias ratings. All studies were considered of high quality and could be downgraded up to three levels (Mokkink et al., 2018b).

- Risk of bias for content validity studies, downgraded one level (serious), if the available content validity studies are of doubtful quality, two levels (very serious), if there were no content validity study or only inadequate quality and the developmental study of doubtful quality, three level (extremely serious), when there were no content validity study and the developmental study was of inadequate quality (Terwee et al., 2018b).

- Risk of bias for other measurement properties, down-graded one level (serious) when there are multiple studies of doubtful quality or only one study of adequate quality, or two level (very serious) when there are multiple studies of inadequate quality, or there is only one study of doubtful quality, or three level (extremely serious) if there is only one study of inadequate quality (Mokkink et al., 2018b).

The evidence was downgraded up to two levels for:

- Inconsistency—serious variation between results (e.g., measurement properties rated as sufficient and insufficient in different studies).

- Imprecision—downgraded one level if total sample size in the studies was <100 and two levels if <50.

- Indirectness- downgraded one level and two level (Serious and Very serious) if studies included in the review were partly performed in participants with another condition.

3.6.11 Formulating recommendations

The recommendation of an OMs was based on the COSMIN guidelines as well as considering interpretability and feasibility aspects of the measurement tool.

1. The OM should have evidence of sufficient content validity (any level) and at least low-quality evidence for sufficient internal consistency could be recommended for use and the results of this tool is trustworthy (Mokkink et al., 2018b).
2. The OMs categorized not in 1 or 3, can be recommended for use but require further research to assess its quality (Mokkink et al., 2018b).
3. The OM had high-quality evidence for an insufficient measurement property it could not be recommended for use (Mokkink et al., 2018b).

3.7 Results (Measurement properties studies)

The PRISMA flow diagram shows results of searching databases for search two of this review, selection process and reasons for exclusion are presented in Figure 3.2. Twenty studies for measurement properties of PROM, one study for PBOM and three studies for body structure and function measures were identified (four studies were identified when the search was updated up to September 2022).

The agreement obtained between reviewers was excellent (SA & EB) for titles/abstracts (95%, Kappa=0.92) and substantial agreement was obtained for full-text articles (90%, Kappa=0.78) (Cohen, 1960). Eleven authors responded from twenty-one who were contacted for clarification of participant ages, the language of PROM used, or for the need of missing information/data. The third reviewer (NRH) was consulted four times.

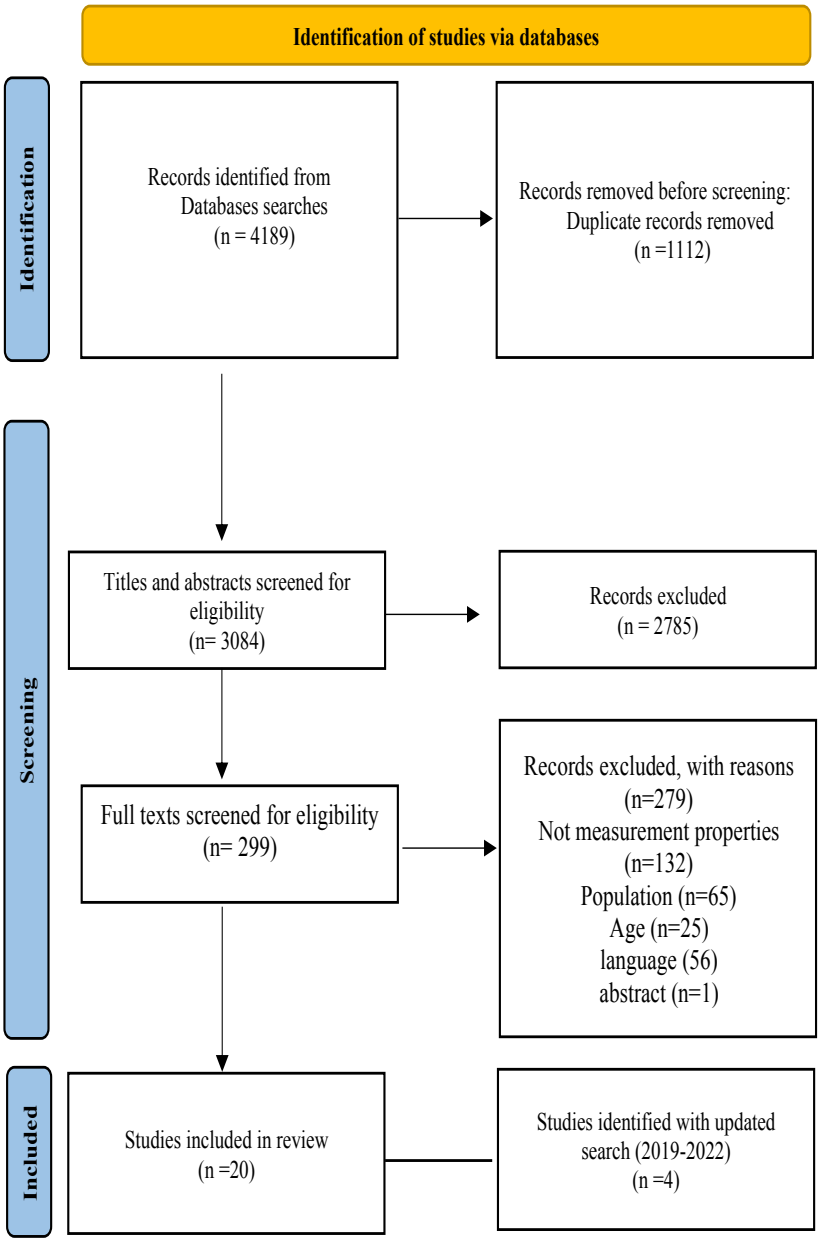


Figure 3.2: PRISMA flow diagram for search two. Adapted from (Page et al., 2021)

3.7.1 Characteristics of studies

Detailed information about studies and participant characteristics including country, age, gender, sample size, curve type, severity, management, and the score obtained by the included OM are shown in *Table* 3.4.

Table 3.4: : Studies and participants characteristics

Patient Reported Outcome Measure									
Reference	**Name of OM**	**Country**	**Age (Mean ±SD) Range**	**Gender (n)**	**Sample size (n)**	**Curve type (%), (n)**	**Curve size degree ± SD, (n)**	**Type of intervention (n)**	**Score (mean ± SD)**
Feise *et al.* (Feise et al., 2005)	SQLI	Canada	14.9 ± 2.4 (10-18)	F (70) M (14)	84	NR	Unbraced 26.1°± 10° Braced 34.3°± 8.7° Postsurgical 31.0°±11.4°	Postsurgical (16) Braced (30) Unbraced (24) Control (14)	81.1± 15.7
Parent *et al*. (Parent et al., 2007)	SQLI	Canada	14.7 ±1.9 (8-20)	F (95)	95	Main thoracic (29) Double thoracic (4) Double major (23) Triple major (2) Thoracolumbar/lumbar (20) Thoracolumbar/lumbar, main thoracic (17)	<30° (34) 30°–50° (44) >50° (17)	Surgery	NR

Bastrom *et al.* (Bastrom et al., 2015)	SRS-24, SRS-22	USA	14.8±2 (10-21)	F (81%)	829	Lenke 1(43%) Lenke 2 (20%) Lenke 3 (7%) Lenke 4 (4%) Lenke 5(16%) Lenke 6(10%)	Pre-surgery 55°±13 Post surgery 20°±9	Pre- and post-surgery	Pre-surgery 45°Cobb SRS-22 (4.6±0.5) SRS-24 (4.1±0.5) >80° Cobb SRS-22 (4.2±0.7) SRS-24 (3.8±0.7) Post-surgery <11°Cobb SRS-22(4.6±0.5) SRS-24 (4.17±0.5) >29°Cobb SRS-22 (4.61 ±0.5) SRS-24 (4.17±0.6)
Asher *et al.* (Asher et al., 2003b)	SRS-22	USA	16.4 (10.6 – 47.3)	F (48) M (10)	58	Single (36) Double (19) Triple (3)	63°	Surgery	Function (0 months) 4.1 Function (3 months) 3.3 Function (6 months) 3.9 Function (12 months) 4.2 Function (24 months) 4.3
Asher *et al.* (Asher et al., 2003c)	SRS-22	USA	Control 13 (10.7-15.4) non-surgical 14 (9.9 - 16) non-surgical untreated	Control F(15) M(4) Non-surgical F(57) M(11) Non-surgical untreated	Total (119) Control (19) Nonsurgical (68) Untreated (54) Braced (14) Pre-surgery	Thoracic, Thoracolumbar, Lumbar; double Triple	Largest cobb angle Non-surgical untreated 27° Braced 31° Pre-surgery 61°	Brace, pre-surgery, control	Control (4.5±0.35) Nonsurgical (4.4±0.36) non-surgical untreated (4.4±0.37) Non-surgical braced (4.5±0.32) Pre-surgery (4.2±0.42)

			14 (10.8-16) non-surgical braced 13 (9.9 - 15.2) Pre-surgery 14 (10.6–15.8)	F(44) M(10) Non-surgical braced F (13)M(1) Pre-surgery F (31) M(1)	(32)				
Parent *et al.* (Parent et al., 2009)	SRS-22	Canada	13.5–20 (153) Total (18.6 ± 9.2)	F (153)	153	NR	Maximal cobb angle 37.6 ±15.5 30° (58) 30°–50°(66) 50° (4)	Observation (107) Brace (32) Pre-surgery (22) post-surgery (62)	Observation (4.3 ± 0.59) Brace (4.5 ± 0.59) Pre-surgery (4.2 ± 0.58) post-surgery (4.1 ± 0.60)
Carreon *et al.* (Carreon et al., 2010)	SRS-22	USA	14.3 ± 1.9 (10 –18)	F (735) M (152)	887	NR	53°±18°	Pre & 1-year post-surgery	Pre-surgery 4.15 ± 0.55 Post-surgery 4.23 ± 0.46
Verma *et al*. (Verma et al., 2014)	SRS-22	USA & Ghana	15.4	F (100) M (60)	160	NR	Ghana 67.2° USA 52°	Pre-surgery	Ghana 3.7 ± 0.8 USA 4.2 ± 0.4
Berliner *et al.* (Berliner et al., 2013)	SRS-22r	USA	13.8 (11.0 - 17.2)	F (115) M (40)	155	Non-surgical Thoracic (56.5%) Thoracolumbar (38.7%) Lumbar (4.8%) Pre-surgical	Total 43.1° non-surgical 21.9° Presurgical 57.2°	Non-surgical & pre-surgical	0° –19° (4.5± 0.47) 20° –40° (4.4 ± 0.37) 41° –50° (4.1± 0.69)

						Thoracic (65.2%) Thoracolumbar (34.8%) Lumbar (0%)			51° –60° (4.2 ± 0.54) >60° (4.3± 0.55)
Kelly *et al.* (Kelly et al., 2019)	SRS-22r	USA	14.6 (10–22)	F (1,034) M (247)	1,281	Lenke 1(552) Lenke 2 (272) Lenke 3 (93) Lenke 4 (46) Lenke 5 (196) Lenke 6 (120)	NR	1, 2year Post-Surgery	Activity MCID (0.08) MDMD (0.24)
Glattes *et al*. (Glattes et al., 2007)	SRS-22r, CHQ-CF87	USA	14.1 ± 2.7 (8-18)	F(58) M(12)	Total (70)	NR	29.8° ± 12.3°	Pre-surgery	SRS-22r (4.5±0.65) CHQ-CF87 (91±15.6)
Fedorak et al. (Fedorak et al., 2019)	PROMSIS, SRS22r	USA	14.4 ±2.1 (11.4–17.4)	F (78.8%) M(21.2%)	113	Thoracic (67%) Thoracolumbar (21.7%) Lumbar(11.3%)	Largest curve 28.8°±14.3 Thoracic kyphosis °34.1 ±14.9 Lumbar lordosis 54.8°± 13.3	Observed, Pre-or post-bracing (69.0%) Brace (27.4%) Surgery (3.5%)	PROMSIS, Mobility (50.93 ±9.80) SRS-22r, Function (4.5±0.5)
Roberts et al. (Roberts et al., 2011)	SRS-30	USA	F (14.0) M (15.2)	F (83.4%) M(16.5%)	744	Risser grade M (mean 3.5) F (mean 3.2)	F (53.3°) M (55.9°)	Pre-surgery, 2yr. post-surgery	Pre-surgery F(4.2)M(4.2) Post-surgery F (4.3) M (4.4)
Lubicky *et al.* (Lubicky et al., 2011)	SRS-30	USA	15.6 ±1.7	F (75%)	356	NR	NR	Pre-surgery, 2yr. post-surgery	Pre-surgery (4.18 ± 0.55) post-surgery (4.34 ± 0.51)

Sarwahi *et al.* (Sarwahi et al., 2018)	SAQ	USA	15 (13 – 17)	F (71) M (24)	95	NR	Pre-surgery 51.08° Post-surgery 15.98°	NR	NR
Lerman *et al.* (Lerman et al., 2002)	PODCI	North America	Parent 15.2 (11.7–18.8) Patient 15.3 (11.7–20.9)	Parent F (88) M (9) Patient F (86) M (9)	102	Thoracic (17) Thoracolumbar (6) Lumbar (7) Double curve (17)	10-29° (n=23) 30-49° (n=20) >50°(n=4)	1-year Post surgery	Upper extremity (96.8± 9.9) Transfer (97.6± 4.7) Sport & Physical Function (85.5±17.5) Global function (89.4±9.8)
Mannion *et al.* (Mannion et al., 2022)	SRS-16	Different countries	15.0 ± 2.2	F (75%)	English (2713) Spanish (270) German (264) Italian (223) French (148)	NR	NR	NR	SRS (3.6 ± 0.6) CFI (0.96) RMSEA (0.06)
Bouton et al. (Bouton et al., 2022)	PROMIS	USA	14.6	F (79.8%)	986	Thoracic(559) Thoracolumbar (427)	Thoracic (35.9) Thoracolumbar (29.2)	NR	Small (r = 0.491) Moderate (r =0.671) Large (r = 0.658)

Yau *et al.* (Yau et al., 2020)	PROMIS	USA	13.0 ± 2.2	F (135) M (71)	206	NR	NR	Brace 17 (8%)	Function SD (4.9 ± 0.3) SA (4.6 ± 0.4) Mobility SD (49.6 ±9.0) SA(56.7 ± 5.4)
Mitchell (Mitchell et al., 2022)	PROMIS, SRS-22	USA	14±1.6	F (166)	200	Proximal thoracic (43) Main thoracic (125) Thoracolumbar (17) Lumbar (15)	33±15	Observation (48) Bracing (94) For surgery (32) Post-surgery (26)	Function (SRS22) Mobility (r = 0.64) Physical Activity (r = 0.34)
Performance Based Outcome Measure									
Gao *et al.* (Gao et al., 2019)	TUG	USA	Mild AIS 14.9 ±1.7 Moderate AIS 16.4±3.3 Severe AIS 15.3±3.1	NR	AIS (30) Control (30)	Right-sided Thoracolumbar	Mild AIS 19.9°±4.3 Moderate AIS 31.8°±4.2 Severe AIS 53.4°±16.1	Pre-treatment	TUG (Seconds) Mild AIS (6.8±1.5) Moderate AIS (6.9±0.9) Severe AIS (6.5±0.8) Healthy control (6.0±0.6)

Body structure and function outcome measure									
Hresko *et al.* (Hresko et al., 2006)	MST	USA	14.2 ± 1.9 (11.3-18.6)	F (37)	37	Thoracic Lumbar	Thoracic 40°±20° Lumbar 31°±12°	Pre-treatment	5.7 ± 2.2 cm
Eyvazov *et al.* (Eyvazov et al., 2017)	MST FTF test, Axial rotation, LSB, ΔC7-PSIS	China	15.7 ± 4.1	M (12) F (46)	58	Lenke 5 (Thoracolumbar/ lumbar)	Group A 25° ± 7.1° Group B 49.8° ± 13.6° Total 34°± 9.2°	Pre-treatment	Modified Schober's (cm) Group A: (20.6 ± 1.4) Group B: (20.3± 1.2) FTF test (cm) Group A: (10.1±11.2) Group B: (11±10.3) ΔC7-PSIS (27.6±1.8%) LSB (degrees) Group A: (66.6±13.4) Group B: (57.8±14.3) Axial rotation (degrees) Group A: (90.1±21.9) Group B: (5.9±19.6)

Stepien *et al.* (Stępień et al., 2018)	TPHA test	Poland	AIS (12.7 ± 2.6) Control (11.8 ±2.5)	F (98)	Control (49) AIS (49)	Risser sign Grade 0 (14) Grade 1 (11) Grade 2 (6) Grade 3 (3) Grade 4 (9) Grade 5 (6)	Thoracic 27.7° ±13.4° Lumbar 25.8°±10.5°	Physiotherapy	AIS Left TPHA - 10.93°±4.64° Right TPHA - 2.37°± 8.30° Control Left TPHA -11°± 3.30° Right TPHA - 8.64°±4.70°

Abbreviations: AIS: Adolescent Idiopathic scoliosis, CHQ-CF87: Child Health Questionnaire- Child Self-Report Form 87, C7-PSIS:Cervical 7 to Posterior Superior Iliac Spine, F: Female, FTF: Fingertip To Floor Test, LSB: Lateral Side Bending, M: Male, MCID: Minimal Clinically Important Difference, MDMD: Minimal Detectable Minimal Difference, MST: Modified Schober Test, NR: Not Reported, OM: Outcome Measure, PODCI: Paediatrics Outcomes Data Collection Instrument, PROMSIS: Patient-Reported Outcomes Measurement Information System, SAQ: Sport Activity Questionnaire, SD: Standard Deviation, SRS: Scoliosis Research Society, SRS-22r: Scoliosis Research Society-22Revised, SQLI: Scoliosis Quality of Life Index, TPHA: Trunk Pelvis Hip Angle test, TUG: Timed Up and Go Test, USA: United State of America.

3.7.2 Characteristics of OMs

The identified OMs were 10 PROMs (7 disease-specific and 3 generic), 1 PBOM, and 6 Body structure and function OMs. There was no separate PROM for evaluating PF except the Sport and Activity questionnaire (SAQ), whilst most of the identified measures were included as scale within the PROMs. Majorities of PROMs were developed in USA, followed by Canada. It has been designed to evaluate HRQOL including PF for individuals with AIS before and after surgery. The responses options vary between 1-5 responses.

The most widely used PROM was the SRS-22 (Asher et al., 2003a) then, its revised version SRS-22r (Asher et al., 2000). Both questionnaires have been translated and adapted into different languages. Detailed information about these OMs is presented in *Table 3.5*

Table 3.6 presents the characteristics of the identified PBOM and body structure and function OMs. The Timed Up and Go test (TUG) is the only PBOM identified in this review its measurement properties have been tested for individuals with AIS. It evaluates the average time that patient need to perform standing from a chair, walking 3 meters, return and sit down for 2 to 3 trials (Gao et al., 2019). The body structure and function OMs were measuring mainly the average angle in the spine when patient bend, flex or rotate using goniometer or tape measure (Stępień et al., 2018, Eyvazov et al., 2017, Hresko et al., 2006).

Table 3.5: Characteristics of patient-reported outcome measures

PROMs	Country	Sub-scale items(n)	Target population	Mode of administration	Recall period	Response options	Scoring system	Available translations
SRS-24 (Haher et al., 1999)	USA	General Function (3) Function after surgery (2) Function-activity (3)	AIS	Self-administrated	Now, post-surgery	5 response options	1-5	None
SRS-22 (Asher et al., 2003a)	USA	Function/Activity (5)	AIS	Self-administrated	Now, post-surgery	5 response options	1-5	Turkish (Alanay et al., 2005),Italian (Monticone et al., 2004) , Spanish (Bago et al., 2004) , Japanese (Hashimoto et al., 2007), Traditional Chinese (Cheung et al., 2007), Simplified Chinese (Li et al., 2009), Polish (Glowacki et al., 2009), French (Beausejour et al., 2009, Lonjon et al., 2014),Thai (Leelapattana et al., 2011), Norwegian (Adobor et al., 2010)

SRS-22r (Asher et al., 2006)	USA	Function/Activity (5)	AIS	Self-administrated	Now, post-surgery	5 response options	1-5	German(Niemeyer et al., 2009), Greek (Antonarakos et al., 2009), Dutch(Schlosser et al., 2014), Chinese(Cheung et al., 2007), Brazilian(Camarini et al., 2013), Italian(Monticone et al., 2010) ,Thai (Sathira-Angkura et al., 2012), Arabic (Haidar et al., 2015), Persian(Mousavi et al., 2010) , Swedish(Danielsson and Romberg, 2013)
SRS-30 (SRS.Com)	USA	Function/Activity (5) post-surgery questions (2)	AIS	Self-administrated	Now, post-surgery	Function/Activity (5 response options) Post-surgery (3 response options)	Function (1-5) post-surgery (1-3)	Finnish (Kyrola et al., 2017) Brazilian (Carrico et al., 2012)
SRS-16 (Mannion et al., 2022)	USA	Function/Activity (4)	AIS	Self-administrated	Non-Surgical	Function/Activity (5 response options)	Function (1-5)	English, Spanish, Italian, German and French (Mannion et al., 2022)
CHQ-CF87 (Glattes et al., 2007)	USA	Physical Functioning (9)	Generic	Self-administrated	NR	4, 5, 6 Response options	0-100	-

SQLI (Feise et al., 2005)	Canada	Physical activity (5)	AIS	Self-administrated	Four weeks	5 Response options	0-4	-
SAQ (Sarwahi et al., 2018)	USA	Total (24) School, Gym, carry backpack, Bend over, Running	AIS	Self-administrated	Post-surgery	NR	NR	-
PROMIS (Fedorak et al., 2019)	USA	Mobility	Generic	Self-administrated	7-day	5 response options	Mean T-score 50, SD 10	-
PODCI (Lerman et al., 2002)	North America	Upper Extremity Functioning, Transfers& basic Mobility Sport & Physical Function Global function	Generic Paediatric orthopaedic conditions	Self-administrated Parent-report Adolescents report	NR	3-6	0-100	-

Abbreviations: CHQ-CF87: Child Health Questionnaire- Child Self-Report Form 87, NR: Not Reported, PODCI: Paediatrics Outcomes Data Collection Instrument, PROMIS: Patient-Reported Outcomes Measurement Information System, SAQ: Sport Activity Questionnaire, SRS: Scoliosis Research Society, SRS-22r: Scoliosis Research Society-22Revised, SQLI: Scoliosis Quality of Life Index,

Table 3.6 : Characteristics of performance-based and Body structure & function measures

Outcome Measure (Reference)	Activity	Required Equipment	Number of trials	Parameter measured
TUG (Gao et al., 2019)	Stand from chair, walk 3m, return, sit down	Chair, stopwatch, walking space	3 trials	Average of time in seconds
MST (Hresko et al., 2006, Eyvazov et al., 2017)	Marks on PSIS, keep knees straight, bend forward and touch the floor	Tape measure	2-3 trials	Average of distance in cm
FTF test (Eyvazov et al., 2017)	Stood upright, bend forward, and touch the floor	Tape measure	2 trials	Average of distance in cm
C7-PSIS distance (Eyvazov et al., 2017)	Stand upright, maximally flex, and extend neck, distance measured between C7 spinous process and PSIS	Tape measure	2 trials	Average of distance in cm
LSB angles (Eyvazov et al., 2017)	In upright posture, knees straight, bend to the side without rotation	Goniometer	2 trials	Average of angles in degrees between lines joining PSIS and C7
Axial rotation (Eyvazov et al., 2017)	Seated position, locked both arms in front of body with fixed pelvic, shoulder rotation controlled by a goniometer holder device	Goniometer	2 trials on left and right side	Average of angles in degrees
TPHA (Stępień et al., 2018)	Supine, flex & pull lower limbs, then move limbs to the left or right side	Plurimeter	Three times on each side of body	Average of angles in degrees

Abbreviations : C7-PSIS:Cervical 7 to Posterior Superior Iliac Spine, MST: Modified Schober Test, FTF: Fingertip to Floor Test, LSB: Lateral Side Bending, TPHA: Trunk Pelvis Hip Angle test, TUG: Timed Up and Go Test

3.7.3 Risk of bias

The measurement properties that have been evaluated are development (n=1) (Feise et al., 2005), structural validity (n=1) (Mannion et al., 2022), internal consistency (n=3) (Feise et al., 2005, Glattes et al., 2007), reliability (n=5) (Glattes et al., 2007, Feise et al., 2005, Sarwahi et al., 2018, Stępień et al., 2018), measurement invariance (n=2) (Verma et al., 2014, Roberts et al., 2011), measurement error (n=2) (Carreon et al., 2010, Kelly et al., 2019), , responsiveness (n=2) (Kelly et al., 2019, Asher et al., 2003b), construct validity (n=22) (Lerman et al., 2002, Feise et al., 2005, Fedorak et al., 2019, Lubicky et al., 2011, Bastrom et al., 2013, Asher et al., 2003c, Parent et al., 2009, Glattes et al., 2007, Berliner et al., 2013, Kelly et al., 2019, Gao et al., 2019, Hresko et al., 2006, Eyvazov et al., 2017, Bouton et al., 2022, Mannion et al., 2022, Mitchell et al., 2022, Yau et al., 2020). Results of risk bias assessment are presented in the (*Appendix 7).*

3.7.4 Measurement Properties and synthesis of evidence

Table 3.7 shows the summary of findings table for results of measurement properties and the overall evidence for measurement properties against COSMIN and modified GRADE approach.

Table 3.7: Summary of findings table for the measurement properties of outcome measures

Measurement property	Outcome measure (Subscale)	Summary result	Overall rating	Quality of evidence
Structural validity	SRS-16	CFI = 0.96 RMSEA = 0.06	+	Low (One study adequate quality, indirectness)
Internal consistency	SRS-22r (Activity)	∝ = 0.82	?	Moderate (Imprecision)
	SQLI (Physical activity)	∝ = 0.82 (0.76–0.88)	?	Moderate (Imprecision)
	CHQ-CF87 (Physical function)	∝ = 0.89	?	Moderate (Imprecision)
Reliability	SRS-22r (Activity)	ICC=0.76 (0.56– 0.80)	+	Low (One study adequate quality, Imprecision)
	SQLI (Physical activity)	ICC= 0.46 (0.29 –0.63)	–	Low (One study adequate quality, Imprecision)
	CHQ-CF87 (Physical function)	ICC=0.73 (0.20– 0.85)	+	Low (One study adequate quality, Imprecision)
	SAQ	Kappa k ≥ 0.70	+	Very low (One study of doubtful quality)
	TPHA Test	ICC= 0.85 (0.95-0.98)	+	Moderate (Imprecision)
Cross-cultural validity\ Measurement invariance	SRS-22 (Activity)	No multiple group factor analysis performed	?	Very low (one study inadequate quality, Imprecision)
	SRS-30 (Function/Activity)	No multiple group factor analysis performed	?	Moderate (one study adequate quality)
Measurement error	SRS-22 (Activity)	SDC (0.24) >MIC (0.08)	–	Moderate (one study of adequate quality)
	SRS-22r	SDC (0.41) > MIC (0.08)	–	Moderate (one study of adequate quality)

Construct validity	SRS-24 (Function)	2 hypotheses confirmed	+	High (One study very good quality
	SRS-22 (Activity)	2 out of 9 hypotheses confirmed	–	Moderate (Inconsistency)
	SRS-22r (Function)	4 hypotheses confirmed	+	Moderate (Inconsistency)
	SRS-30 (Function)	1 hypothesis confirmed	+	High (One study very good quality)
	SQLI (Physical activity)	2 hypotheses confirmed	+	Moderate (Imprecision)
	PODCI (functional scales)	2 hypotheses out of 5 confirmed	–	Moderate (One study adequate quality)
	PROMIS (Mobility)	2 hypotheses confirmed	+	Moderate (Two studies adequate quality)
	TUG test	2 hypotheses out of 3 confirmed	+	Moderate (One study adequate quality)
	MST, FTF Test, C7-PSIS	3 hypotheses not confirmed	–	Moderate (Imprecision)
	LSB, Axial rotation	2 hypotheses confirmed	+	Moderate (Imprecision)
Criterion validity	MST	Not all information for '+' reported	?	Moderate
Responsiveness	SRS-22 (Activity)	4 hypotheses confirmed	+	Very low (One study of doubtful quality, Imprecision)
	SRS-22r (Function)	1 hypothesis not confirmed	–	Low (One study doubtful quality)

Abbreviations: CHQ-CF87: Child Health Questionnaire- Child Self-Report Form 87, C7-PSIS:Cervical 7 to Posterior Superior Iliac Spine, FTF: Fingertip To Floor Test, LSB: Lateral Side Bending, MST: modified Schober Test, PODCI: Paediatrics Outcomes Data Collection Instrument, PROMSIS: Patient-Reported Outcomes Measurement Information System, SAQ: Sport Activity Questionnaire, SRS: Scoliosis Research Society, SRS-22r: Scoliosis Research Society-22Revised, SQLI: Scoliosis Quality of Life Index, TPHA: Trunk Pelvis Hip Angle test, TUG: Timed Up and Go Test

3.7.4.1 Patient Reported Outcome Measures

3.7.4.1.1 Scoliosis Research Society (SRS -24)

The SRS-24 represents the initial version of the SRS questionnaires (Haher et al., 1999). Comprising three sub-scales and 8 items, it assesses function in terms of general function, post-surgery function, and function/activity (Haher et al., 1999). Unfortunately, the developmental study did not report participants' ages, and the authors were not reachable, leading to an inability to evaluate the quality of the developmental study, consequently excluding it from this review (Haher et al., 1999).

A study of very good quality provided high-quality evidence, demonstrating that the construct (discriminative) validity of the functional scales of the SRS-24 was sufficient for individuals with AIS (Bastrom et al., 2015). The study results aligned with predefined hypotheses, revealing a significant difference in functional scores between individuals with AIS in pre- and post-surgery status (Bastrom et al., 2015).

3.7.4.1.2 Scoliosis Research Society (SRS-22)

The SRS-24 underwent refinement to become the SRS-22, involving the elimination or modification of items with low internal consistency, and an expansion of response options to five (Asher et al., 2000). However, the study underpinning this refinement was excluded from this review due to participant ages exceeding 18 years (Asher et al., 2000).

The responsiveness of the function scale of the SRS-22 was deemed sufficient, with confirmation of four predefined hypotheses (Asher et al., 2003b). However, this conclusion was based on very low-quality evidence due to study quality concerns and imprecision (Mokkink et al., 2018b).

The measurement invariance of the scale was rated indeterminate since no multiple group factor analysis or differential item functioning was performed, precluding an evaluation of methodological quality (Verma, 2014).

Moderate-quality evidence suggested that the construct validity of the functional scale of SRS-22 was insufficient based on results from three studies (Parent et al., 2009, Asher et al., 2003c, Bastrom et al., 2015). The evidence was downgraded due to inconsistency between these studies following COSMIN guidance (Mokkink et al., 2018b).

The measurement error of the SRS-22 functional scale was rated as insufficient because the Small Detectable Change (0.24) exceeded the Minimal Important Change (MIC) (0.08) (Carreon et al., 2010). The evidence was of moderate quality based on one study of adequate quality (Mokkink et al., 2018b).

One study of adequate quality indicated sufficient reliability for the function scale of the SRS-22, with an Interclass Correlation Coefficient (ICC) of 0.76 (0.56–0.80) (Glattes et al., 2007). However, the evidence was downgraded to low-quality due to imprecision (sample size <100 participants) (Mokkink et al., 2018b). Internal consistency was rated indeterminate because the criteria for sufficient structural validity were not met (Mokkink et al., 2018b).

One study of adequate quality indicated a sufficient reliability of the function scale of the SRS-22 i.e., Interclass Correlation Coefficient (ICC) 0.76 (0.56– 0.80) However, the evidence was downgraded to low-quality evidence for the imprecision (sample size <100 participants) On the other hand, the internal consistency (Glattes et al., 2007) was rated indeterminate because the criteria of sufficient structural validity was not met

3.7.4.1.3 Scoliosis Research Society (SRS-22r)

To enhance the internal consistency of the function domain in the SRS-22, modification was made to questions 15 (pertaining to financial difficulties) and 18 (related to going out with friends) (Asher et al., 2006). The internal consistency of the function domain was assessed in patients with an average age of 18.9 years, revealing a Cronbach's alpha (α) of 0.86. Notably, this decreased to 0.39 and 0.61 in younger patients with average ages of 13.8 and 16.4 years, respectively. These findings were consistent with results from studies that translated and adapted the SRS-22 into Spanish and Turkish (Alanay et al., 2005, Bago et al., 2004). This refinement eventually led to the development of the SRS-22 revised version, which has been used interchangeably with the original SRS-22. In some measurement properties studies, it was unclear which version of the questionnaire was employed. To address this ambiguity, communication was initiated with authors to confirm the specific version of the SRS questionnaire utilised in their studies.

Construct validity for the function scale of the SRS-22r was evaluated through two ways: convergent validity, involving correlation with another scale measuring a similar construct (Mokkink et al., 2018b), and discriminative validity, assessing the scale's ability to distinguish between different levels of condition severity (Asher et al., 2003c). The function scale demonstrated satisfactory convergent validity as evidenced by a robust correlation (Pearson r = 0.73) with the mobility scale of the Child Health Questionnaire-Child Self-Report Form 87 (CHQ-CF87) (Glattes et al., 2007). Whilst, when examining discriminative validity, the predefined hypothesis was not supported, indicating insufficient discriminative validity (Berliner et al., 2013). This lack of confirmation led to a downgrading of the evidence for construct validity due to inconsistent results in the assessment of discriminative validity.

The responsiveness of the functional scale of the SRS-22r was considered insufficient, as the results from responsiveness testing did not align with the predefined hypotheses. This assessment was supported by evidence of low quality (Kelly et al., 2019). Similarly, the functional scale of the SRS-22r exhibited insufficient measurement error, specifically with a Smallest Detectable Change (SDC) of 0.41, which exceeded the Minimal Important Change (MIC) of 0.08 (Kelly et al., 2019). This observation is based on moderate-quality evidence, derived from one study of adequate quality (Mokkink et al., 2018b).

3.7.4.1.4 Scoliosis Research Society (SRS-30)

The SRS-30 is a version of the SRS questionnaire, combining questions from both SRS-24 and SRS-22. However, only one study identified in this review delved into the evaluation of its construct validity (Lubicky et al., 2011). Notably, a significant difference in activity/function scores (0.50) was noted across all types of instrumentations before and after surgery for patients with AIS in this study (Lubicky et al., 2011). Consequently, the hypothesis was confirmed, and the construct validity was deemed sufficient, supported by high-quality evidence. The measurement invariance of the SRS-30 was rated as indeterminate since no multiple group factor analysis was conducted (Roberts et al., 2011).

3.7.4.1.5 Scoliosis Research Society (SRS-16)

In a recent study, the structural validity of the SRS-16, which comprises 4 items in each domain instead of the usual 5, was assessed (Mannion et al., 2022). Confirmatory factor analysis was employed to scrutinize the factor structure of the twenty non-management items of the SRS, involving 3618 young patients with spinal deformity and conducted across various languages (English, Spanish, German, Italian, and French). The 16-item version exhibited commendable model fit, as indicated

by a comparative fit index (CFI) of 0.96 and a Root Mean Square Error of Approximation (RMSEA) of 0.06. This performance was notably superior to the 20-item version ($p < 0.001$) across all language versions. The study's findings were considered sufficient as they met the criteria outlined by COSMIN, although supported by a low quality of evidence (Mokkink et al., 2018b).

3.7.4.1.6 Scoliosis Quality of Life Index (SQLI)

The SQLI, a modified version of the SRS-22, was developed to address challenges in certain questions for younger individuals with AIS. Notably, questions like "Which of the following best describes the appearance of your trunk?" and "(Are you and/or your family experiencing financial difficulties because of your back?)" posed difficulties for younger respondents (Feise et al., 2005). The SQLI questionnaire was adapted by focusing on the physical activity domain instead of the function domain found in the original questionnaire (Feise et al., 2005).

In this review, the SQLI was the only questionnaire that has been evaluated for its content validity. Despite being rated as sufficient based on reviewers' assessments, the evidence supporting this conclusion was considered very low quality. This was due to the scale's comprehensibility being tested exclusively among healthy school children with an average age of 9.9 years old (Feise et al., 2005). According to COSMIN guidance, these children may not be representative of the population of interest (Terwee et al., 2018c).

The internal consistency of the physical activity scale of SQLI was rated as indeterminate because it doesn't meet the criteria for at least low evidence for sufficient structural validity. The reliability of the same scale was deemed insufficient as the Intraclass Correlation Coefficient (ICC) was less than 0.70 (ICC = 0.46, 95% CI: 0.29–0.63) (Feise et al., 2005). The evidence was downgraded due to a serious risk of bias and imprecision. The construct validity of the physical

activity scale of SQLI was considered sufficient, with two hypotheses confirmed. This evidence was of moderate quality.

3.7.4.1.7 Patient-Reported Outcomes Measurement Information System (PROMIS)

The PROMIS has a mobility scale to evaluate PF and it has been used for individuals with AIS (Fedorak et al., 2019). Multiple studies have investigated the construct validity of PROMIS, encompassing both discriminative and convergent validity. The hypotheses formulated by the research teams were consistently confirmed in the majority of these studies. The overall results of the construct validity assessment were deemed sufficient, primarily due to the scale's correlation with the function scale of the SRS-22r (Pearson r=0.53–0.65) (Fedorak et al., 2019, Bouton et al., 2022, Yau et al., 2020, Mitchell et al., 2022). It's noteworthy that PROMIS stands out as the only scale rated as high in the evaluation. This elevated rating is attributed to the inclusion of multiple studies of very good quality (Mokkink et al., 2018b).

3.7.4.1.8 Paediatrics Outcomes Data Collection Instrument (PODCI)

The Paediatric Outcomes Data Collection Instrument (PODCI) is a generic questionnaire featuring functioning scales designed to evaluate upper extremities, transfer, and basic mobility in adolescents and paediatric individuals with orthopaedic conditions (Lerman et al., 2002). However, it was rated insufficient construct validity for the functional scales of PODCI supported with a moderate quality of evidence (Lerman et al., 2002). The study found that none of the predefined hypotheses were confirmed, as there was no identified difference in functional scores among individuals with AIS concerning different ages, varying sizes of Cobb angles, or different curve types (Lerman et al., 2002).

3.7.4.1.9 Child Health Questionnaire-Child Self-Report Form 87 (CHQ-CF87)

The Child Health Questionnaire-Child Form 87 (CHQ-CF87) is a generic questionnaire that includes a scale for assessing PF in the paediatric population (Glattes et al., 2007). The PF scale of CHQ-CF87 fell short of meeting the criteria for sufficient structural validity (Mokkink et al., 2018b). Consequently, the internal consistency of the PF scale was deemed indeterminate (Glattes et al., 2007). On the other hand, the reliability of the same scale was found to be sufficient supported by evidence of low quality, with an ICC of 0.73 (95% CI: 0.20–0.85) (Mokkink et al., 2018a).

3.7.4.1.10 Sport Activity Questionnaire (SAQ)

The Spinal Appearance Questionnaire (SAQ) is a questionnaire crafted through a test-retest method (Sarwahi et al., 2018). In the context of COSMIN definitions, this study is classified as a reliability study (Mokkink et al., 2010). Consequently, in this review, the SAQ has been evaluated primarily as a reliability study. The reliability coefficient for the SAQ, as measured by Kappa (K) ≥ 0.73, shows sufficient reliability. However, the evidence was downgraded due to the limitation of having only one study of doubtful quality (Mokkink et al., 2018a).

In conclusion, according to COSMIN method for a systematic review of PROM. The PROM should exhibit any level of sufficient content validity and low level of evidence of sufficient internal consistency to be recommended for use (Mokkink et al., 2018a, Mokkink et al., 2018b). In the current review, none of the PROMs identified met these criteria, thus none of the PROMs are recommended for use with individuals with AIS. Furthermore, none of these PROMs had a high evidence of insufficient measurement properties. Therefore, these PROMs can be used but it requires further assessment of their quality of its measurement properties to be recommended for use for individuals with AIS (Mokkink et al., 2018a).

3.7.4.2 Performance Based Outcome Measure (PBOM)

3.7.4.2.1 Timed Up and Go test (TUG)

The TUG test is the only PBOM identified in this review. Its construct validity was assessed to determine if there were differences in the time to perform the TUG test among individuals with AIS with varying curve severity (Gao et al., 2019). The TUG test demonstrated sufficient construct validity, revealing that individuals with mild and moderate curves spent a longer time performing the TUG compared to those with severe curves (Gao et al., 2019).

3.7.4.3 Body structure and function measures

3.7.4.3.1 Trunk Pelvis Hip Angle (TPHA)

The TPHA test, measuring the mobility of the lumbo-pelvic-hip complex, exhibited sufficient inter-rater reliability (ICC > 0.942) (Stępień et al., 2018). Despite this high reliability, the evidence was rated of moderate quality, downgraded due to imprecision stemming from a sample size less than 100, following COSMIN criteria (Mokkink et al., 2018a).

3.7.4.3.2 Modified Schober Test (MST)

The MST has been evaluated for its validity as a surrogate to radiographic methods for measuring spinal motion (Hresko et al., 2006). Very low-quality evidence reduced the criterion validity of MST indeterminate, as required information of the correlation with radiographs was not reported. Furthermore, its construct validity was deemed insufficient, as none of the hypotheses were confirmed, supported by moderate-quality evidence (Hresko et al., 2006).

3.7.4.3.3 Fingertip To Floor Test (FTF) test, Axial rotation, Lateral Side Bending (LSB), Δ7th cervical vertebra to Posterior Superior Iliac Spine (C7-PSIS)

The construct validity of the FTF test and the distance between the 7th cervical vertebra to C7-PSIS was rated insufficient with moderate-quality evidence (Eyvazov et al., 2017). No significant difference in scores of these tests was found between AIS with mild and severe curves. Conversely, the construct validity of LSB) and Axial Rotation was rated as sufficient, as differences were identified between AIS with varying curve severity (Eyvazov et al., 2017).

3.7.5 Interpretability and feasibility

Information about interpretability and feasibility aspects of functional scales included in this review are available in (*see Appendix* 7*)*. Majority of these scales had a high ceiling effect ranging between 20% to 44% and minimal floor effects (Bastrom et al., 2015, Glattes et al., 2007, Fedorak et al., 2019). An exception to this is the physical activity scale of SQLI, which displayed minimal ceiling and floor effects (Parent et al., 2007, Feise et al., 2005). The Minimal Clinical Important Difference (MCID) reported for activity domain for SRS-22 is 0.08 (Carreon et al., 2010). While the Minimum Detectable Measurement Difference (MDMD) or the SDC of activity for SRS-22r is 0.24 (Kelly et al., 2019). There was no information reported in the studies identified in this review about response shift and the percentage of missing items. Furthermore, limited information was available about feasibility aspects of these OMs. Most of the included PROMs are completed within 2-3 minutes, and it could be concluded that it these PROMs are easy to complete, available in different settings, and available free of charge.

3.8 Discussion

This is comprehensive, rigorous, two search staged systematic review designed to synthesis the evidence on the measurement properties of OMs used to evaluate PF for AIS, as one of the core outcome domains that should be evaluated and reported (Dodd et al., 2018). The COSMIN guidelines for systematic review of PROMs were adhered to ascertain a high-quality and trustworthy results (Prinsen et al., 2018).

Search one enabled generation of a list of OMs that has been used in AIS to evaluate PF. Using this list, search two was conducted and revealed a few measurement properties studies. That comprised of nine PROMs, just one PBOM, and six measures of body structure and function. None of the identified PROMs had evidence for sufficient content validity and sufficient internal consistency [34]. Thus, PROMs identified in this review have the potential to be recommended for use but are yet to have the measurement properties investigated. The current evidence showed limited information on the measurement properties of PBOM and body function and structure OM in AIS. Therefore, more research studies are needed to enhance the use of these OMs in research and clinical practice.

3.8.1 Patient Reported Outcome Measures

This review highlights an important gap in the evidence on the content validity of the PROMs that routinely used in scoliosis literature to evaluate quality of life as well as PF of AIS. As COSMIN suggested, the content validity is the first and most important measurement property to consider when selecting any PROM (Terwee et al., 2018c). It is a measurement property that reflect that the scale items are comprehensive, comprehensible, and relevant to the population of interest and measure

what is supposed to measure (Terwee et al., 2018a, Brod et al., 2009). It has been recommended that qualitative interviews should be performed with both patients and health care professionals to assess content validity of the PROM (Terwee et al., 2018a). Current review revealed a lack of adequate developmental process for all of the identified PROM, which in turns underpinning its content validity. They were developed in a population that may not truly represent individuals with AIS, as their mean age was larger than individuals with AIS i.e. 25 years old (Asher et al., 2000, Asher et al., 2003a, Asher et al., 2006, Haher et al., 1999).

The only scale that has been investigated for its developmental process was the physical activity scale of SQLI (Feise et al., 2005). The SQLI is a modified version for the SRS-22 questionnaire. The Modification was on the time frame to be 4-week period and replacement of two questions at the function domain , that was related to social behaviour rather than PF (Feise et al., 2005). The questionnaire comprehensibility has been tested among healthy school children only; their mean age was 9.9 years old (Feise et al., 2005). As COSMIN suggested those children were considered not representative to the population of interest (Terwee et al., 2018a). The scale was rated sufficient for its content validity based on reviewers' ratings only. The relevance, comprehensiveness, and comprehension domains were not assessed because of the lack of qualitative interviews with individuals with AIS (Mokkink et al., 2018a). Therefore, more research is required to evaluate the content validity of this scale and it is appropriateness to AIS aged 10-18 years old.

Majority of the identified measurement properties studies in this review tested construct validity. Almost all of the physical functional scales of the identified PROMs have displayed sufficient ratings against COSMIN criteria and the predefined hypotheses were confirmed. That is being able to discriminate between AIS with different ages, curve severity, and management (Asher et al., 2003c, Asher et al., 2003a, Feise et al., 2005, Bastrom et al., 2015), also it correlates with

other OMs measuring similar construct (Glattes et al., 2007, Bouton et al., 2022). Therefore, these scales can be used to evaluate PF of AIS with different disease characteristics and management approaches supported with moderate to high quality of evidence.

The internal consistency of the PROM demonstrates the degree of relatedness between the items of the scale (Prinsen et al., 2018). Sufficient rating for the internal consistency require a sufficient rating for structural validity according to the criteria of good measurement property (Mokkink et al., 2018b). In this review, none of the identified scales had sufficient ratings for internal consistency due to a lack of evidence of sufficient structural validity. On the other hand, three scales for the (SRS-22, CHQ-CF87, SAQ) questionnaire has been rated sufficient for their reliability (Glattes et al., 2007, Sarwahi et al., 2018). Reliable scales shows how much patients scores are consistent/similar when they have not changed for a repeated measurement and under several conditions (De Vet et al., 2011).

Another measurement property that is related to reliability is measurement error, which is the error either systematic or random of a patient's scores' that is not related to the actual changes in the measured construct (Mokkink et al., 2018a, De Vet et al., 2011). Change in patient scores within the limit of agreement or smaller than SDC are considered as measurement error (Mokkink et al., 2018a). This measurement property was evaluated for the function scales for the SRS-22 and SRS-22r only. Both scales had insufficient ratings for the measurement error per criteria of good measurement property.

3.8.2 Performance based Outcome Measures

Overall, there is a tendency in the literature to use PROMs rather than PBOMs; twenty studies were identified evaluating measurement properties of PROMs compared to one study of PBOM (Gao

et al., 2019). Although PROMs are relevant to patients, it should be used cautiously, since factors such as pain (Bastrom et al., 2013) and psychological distress (Sanders et al., 2018), which often reported by the AIS population has an influence on their self-reporting of functional ability (Tomey and Sowers, 2009, Painter et al., 1999). Therefore, it is questionable whether assessment of PF is performed by PROMs only, are they providing adequate information about the actual functional performance of this population?

Although a number of PBOMs has been identified in search one, only one PBOM has been identified, and its measurement property has been tested in AIS. The TUG test is a performance based test used to evaluate functional mobility including balance, and walking ability (Gao et al., 2019). However, more evidence-based studies are still needed to accurately evaluate the important and meaningful activities of daily livings of individuals with AIS.

3.8.3 Body Structure and Function Measures

Radiographs, measured using the Cobb angle, are the gold standard measure for evaluating the spinal curvature (Cobb, 1948). While the measurement properties of this measure have been investigated extensively in the literature (Langensiepen et al., 2013), little interest was given to other forms of OM, such as MST and FTF test. These tests considered an inexpensive, easy, quick measure that does not require children to be exposed to ionising radiation. When adequate measurement properties of these OM established, it could be used as a surrogate to the radiographs.

3.9 Strength and limitations

This review used a two-search strategy; search one enabled producing a list of OMs, which was utilised in search two for identifying measurement properties studies. Furthermore, all types of

OMs were included in this review, and it was not specific to one category of OM. The ICF framework was used to appropriately classify the PBOM and body structure and function measures. Risk of selection bias was minimized by involving two independent reviewers for all stages. Utilising the COSMIN methodology is another strength of this review, where it is currently preferred approach for the systematic review of measurement properties.

However, the ratings of studies were determined using the lowest score principle, which may result in underestimating a study's final methodological quality score. Another limitation of this review is the inclusion of English studies only which may introduce language bias, potentially excluding valuable studies from non-English publications. Lastly, the generalisability of the results from this review to other OMs for adults with scoliosis or different forms of scoliosis may be limited.

3.10 Clinical implications

This review has highlighted a substantial gap in the content validity of often utilised PROMs for individuals with AIS. This raises concerns about the appropriateness of these measures for the target population. Until additional evidence is established, clinicians should approach the use of PROMs with caution, recognizing potential influences from factors such as pain and psychological distress commonly reported by individuals with AIS (Bastrom et al., 2013). To ensure a more comprehensive evaluation of PF in individuals with AIS, it is recommended that clinicians consider incorporating a combination of measures. This may include not only PROMs but also alternative functional assessments measures.

3.11 Future research

The existing body of literature lacks sufficient research on the measurement properties of commonly used PROMs in individuals with AIS. This includes important aspects like content validity, internal consistency, and structural validity. The same knowledge gap also applies to PBOMs, and measures related to body structure and function. Additionally, many of the scales reviewed showed high ceiling effects, which limit their ability to accurately assess changes in the functional status of individuals with AIS. To address these issues, it is crucial to establish robust measurement properties of these OMs. This will enable researchers to make appropriate choices when designing studies and ensure standardization across different countries and populations.

3.12 Chapter summary

This chapter aimed to assess the currently used OMs for evaluating PF in individuals with AIS and their measurement properties. Two comprehensive searches were conducted to achieve this objective. The review identifies a variety of OMs used for evaluating PF among individuals with AIS. Majority of measurement properties studies identified were for PROMs with a paucity of information on the measurement properties of PBOM and body structure and function measures. According to COSMIN suggestions, none of the identified measures can be recommended for use in individuals with AIS. More measurement properties studies are required to support recommendation of these measures for research and clinical practice.

Chapter Four

Enhancing Content Validity of the Scoliosis Research Society Questionnaire (SRS-22r): Insights from a Qualitative Cognitive Debriefings Study

This chapter reports in full the contents of a published manuscript by the thesis author protocol (Alamrani et al., 2021a). It includes verbatim text from the published manuscript and some changes in the introduction section were employed for the purpose of this thesis.

Publication and conference presentation

- **Alamrani, S.,** Gardner, A., Falla, D., Russell, E., Rushton, A.B. and Heneghan, N.R., 2021. Content validity of Scoliosis Research Society questionnaire-22 revised (SRS-22r) for adolescents with idiopathic scoliosis: protocol for a qualitative study exploring patient's and practitioner's perspectives. BMJ open, 11(12), p.e053911. (**Appendix 3**)
- **Alamrani S.,** Gardner A, Falla D. Russell E, Rushton AB, Heneghan NR. Enhancing content validity of the scoliosis research society questionnaire (srs22r): insights from a qualitative cognitive debriefings study (Under review).
- **Alamrani S.,** Gardner A, Falla D. Russell E, Rushton AB, Heneghan NR. Content Validity of The Scoliosis Research Society (Srs22r): Qualitative Cognitive Debriefings Study. Submitted to Physiotherapy UK- 1st November 2023. Birmingham. United Kingdom. (Poster presentation)

4.1 Abstract

The SRS-22r is a commonly used tool for evaluating the HRQOL of individuals with AIS. However, its relevance, comprehensibility, and comprehensiveness for this unique population has not been evaluated. This study aimed to explore the content of the SRS-22r questionnaire from the perspectives of both individuals with AIS and HCPs.

A qualitative study was conducted according to a published protocol, reported according to the COnsolidated criteria for Reporting qualitative study (COREQ) guidelines. Purposive samples including individuals with AIS (8 female, 3 male) with a mean age of 14.9 years (SD=1.8) and HCPs (3 surgeons, 2 nurses, 2 physiotherapists) with mean clinical experience of 11.8 years (SD=4.9). Participants responded in semi-structured interviews to think-aloud and verbal probing techniques. Interviews were audio and video recorded, transcribed verbatim, and analysed using thematic content analysis.

Collectively participants challenged the relevance of some questions, e.g. those related to attractiveness and financial difficulties. Incomprehensible questions were because of outdated language (e.g., down in the dumps), adult-oriented questions, (e.g., household), and unfamiliar language (e.g., narcotics, bedridden). Additionally, participants reported that a 6-months recall period was excessive and required revision. Participants suggested adding questions about school and sports activities.

Findings of this study highlights areas for potential improvement in the SRS-22r content and structure to better reflect language, age and HRQOL concepts of individuals with AIS. This study provides valuable evidence to guide the development of more appropriate and relevant tools to assess the HRQOL of individuals with AIS.

4.2 Introduction

As stated in *chapter one* , *section 1.2,* the (AIS) is a complex spinal deformity affecting adolescents aged 10 to 18 years old with a prevalence between 2-3% (Negrini et al., 2018). It has a significant impact on many aspects of adolescent life including back pain, respiratory difficulties, and mental health problems (Du et al., 2016a, Mitsiaki et al., 2022, Amăricăi et al., 2020). The SRS-22r is a widely used PROM that has been used to evaluate HRQOL for individuals with AIS (*see Appendix 8*). It has been translated into numerous languages and has been culturally adapted for use in different countries and populations (Climent et al., 2005, Schlosser et al., 2014, Lee et al., 2011, Bago et al., 2004, Simony et al., 2016, Danielsson and Romberg, 2013, Mousavi et al., 2010)

Good research practice recommends that when designing PROMs for children and adolescents, a variety of factors need to be addressed, such as health-related vocabulary, reading level, length of the questionnaire, recall period and response options (Matza et al., 2013), as well as critically being relevant to their age, reflecting their activities and experiences (Matza et al., 2004, Matza et al., 2013, Rothman et al., 2009). The SRS-22r has been selected in a (COS) as a PROM for young adults with spinal deformities (de Kleuver et al., 2017). Findings from *Chapter Three* reveals that the content validity of the SRS-22r is insufficient. Also, *Chapter Two* results shows that the SRS-22r is insensitive to change and may not be suitable to individuals with AIS because of the low response rates and missing data at different time points (Jasani et al., 2016). As stated in *Chapter one, section 1.5.2,* the content validity is the property of a PROM which indicate that the questionnaire contents reflect the constructs to be measured for specific populations and contexts (Brod et al., 2009). It may affect other important measurement properties such as internal consistency, structural validity, and responsiveness (De Vet et al., 2011).

Cognitive debriefing is a process of gaining feedback on the PROM contents from the population of interest and their specialist HCPs to assess 1) relevance, 2) comprehensiveness, and 3) comprehensibility of the PROM contents (Terwee et al., 2018a, Ryan et al., 2012). It is a critical step in PROM development usually carried out prior to quantitative psychometric testing (Patrick et al., 2011b). Qualitative cognitive interviewing provides evidence of the content validity, by eliciting potential errors, and the pattern of interpretation amongst a number of relevant respondents. If the interviews reveal any misunderstanding, then modification of the questionnaire is needed (Rothman et al., 2009). This chapter addresses the third aim of this thesis by conduct cognitive debriefing interviews, specifically to evaluate:

1- The relevance, comprehensiveness, and comprehensibility of the contents of the SRS-22r from the perspectives of those with AIS.

2- The relevance, comprehensiveness, and comprehensibility of the contents of the SRS-22r from the perspectives of HCPs dealing with those with AIS.

4.3 Methods

4.3.1 Design

This is a qualitative study design which is part of a larger study that aims to evaluate the content validity of the SRS-22r questionnaire (*see Appendix 5*). Results are presented and discussed in two separate chapters (*Chapter 4: Cognitive debriefing, and Chapter 5: Concept elicitation*). This approach was chosen to provide a more detailed presentation and analysis of the results, and to enhance clarity and comprehension for the reader.

4.3.2 Qualitative approach and research paradigm

This study is grounded in pragmatic view of world. This approach suggests that objective reality is grounded in the environment and accessible through human experience (Kaushik and Walsh, 2019). It focuses on practical applications and problem-solving in achieving desired inquiry outcomes, whether through single, multiple, or mixed methods (Creswell and Poth, 2016).

Descriptive phenomenology was chosen as methodology for this study as it emphasises the detailed examination of individual lived experiences (Willis et al., 2016). This approach allows in-depth exploration of the experiences and perspectives of participants. Further, emphasizes the suspension of preconceptions of the researcher. This aligns with the goal of approaching the study free from pre-existing perceptions about the questionnaire, ensuring that the interpretations are grounded in the participants' experiences (Willis et al., 2016) .

4.3.3 Research team and reflexivity

A female experienced musculoskeletal physiotherapist researcher and PhD student (SA) conducted the interviews, with support from the co-authors who have expertise in conducting spinal and qualitative research. Further, a spinal surgeon with experience working with AIS was involved along with a patient and public involvement (PPI) representative. No relationship was established with participants prior to the commencement of the interviews. Participants were informed about the professional background of the interviewer and that the study was part of a PhD thesis

4.3.4 Data collection methods

In this study, in depth-semi structured interviews were conducted with individuals with AIS and HCPs. The traditional four-stage model for cognitive debriefing include assessment of (1) comprehension, (2) retrieval of information, (3) judgment or estimation, and (4) selection of a response to the question (Willis and Artino, 2013). This was adapted to cover the three aspect for a content validity study per COSMIN guidance (Terwee et al., 2018b). This study was designed using COSMIN framework as the guiding methodology for assessing the content validity through cognitive debriefings (Terwee et al., 2018b). According to COSMIN initiatives, cognitive debriefings are a component of content validity assessment where individuals from the target population and professionals are asked to provide feedback on relevance, comprehensibility, and comprehensiveness of the items in a PROM (Terwee et al., 2018a, Terwee et al., 2018b). Relevance ensures that the PROM items correspond to the actual experience of the respondents regarding the impact of their health condition, and its treatment and the context of use. Comprehensibility assesses respondents understanding of the PROM items and identifies any potential misinterpretations. Comprehensiveness confirms that the PROM content covers all important areas relevant to the target population, ensuring that no key concepts are missing (Terwee et al., 2018b, Terwee et al., 2018c).

4.3.5 Setting

All interviews were conducted virtually via the Zoom or Microsoft Teams platforms based on participant preference. The interviews took place between January to April 2022 for the individuals with AIS group, and July to September 2022 for the HCPs group.

4.3.6 Ethical approval

Ethical approval to conduct the study was granted from the Health Research Authority and Health and Care Research Wales (REC reference: 21/WM/0076). Written informed assent/consent was obtained from all participants (*see Appendix 9*).

4.3.7 Participants

According to the COSMIN group's recommendations for ensuring a high quality of a cognitive interviewing study, a generally accepted sample size for assessing the validity of items is typically between 7 to 10 participants per item (Terwee et al., 2018a). This is also to allow achievement of adequate agreement among participants considering the diversity of participant characteristics and the complexity of the questionnaire (Brod et al., 2009).

4.3.7.1 Individuals with AIS

A purposive sample of individuals with AIS were recruited (Cobb angle > 25°; age 10–18 years old and had access to a video/audio call platform). To capture all relevant experiences, participants were purposefully selected to represent various age categories (10-12, 13-15, 16-18 years old) and different management approaches (observation, brace, pre-surgery, post-surgery). This ensures that a comprehensive range of experiences is included in the study. Individuals with other forms of scoliosis, and those who were unable to speak English fluently were excluded.

4.3.7.2 HCPs group

A purposive sample of HCPs who were part of the health care team for individuals with AIS (i.e., physiotherapists, nurses, and surgeons) were recruited. They were eligible if they had clinical and/or research experience in managing AIS.

4.3.8 Deviation from the protocol

Originally, cognitive debriefing among HCPs was planned through focus group discussion. However, due to HCPs time constraints, this proved unfeasible. Consequently, individual semi-structured interviews were conducted (*See Appendix 5*). Notwithstanding this change, the individual interviews provided detailed insights into the SRS-22r questionnaire content (Brod et al., 2009).

4.3.9 Interview procedure

Timing of the interview was selected based on participant preference. The topic guide for interviews included the SRS-22r questions, a think aloud standard question, and verbal probing questions (Table 4. 1). Verbal probing questions were used to elicit more detailed information from participants about three aspects of the questionnaire as recommended by COSMIN (i.e. relevance, comprehensibility, and comprehensiveness) (Willis and Artino, 2013, Terwee et al., 2018a, Prinsen et al., 2018). Both groups were invited to think aloud (read and answered the SRS-22r questions), and then verbal probes were asked. Participants were unknown to the interviewer (SA) prior to the interview in all cases. For those with AIS, the interviews were observed by a parent in three instances. All other interviews were held without anyone else present. Participants were interviewed only once, and all interviews were audio and video recorded, and field notes were collected. Audio recordings

were transcribed verbatim by an official transcription service, and the transcripts were sent to the participants

4.3.9.1 Individuals with AIS group

Participants were recruited by a research nurse who outlined the study to them. Those who showed interest in the study were given an age relevant participant information sheet (PIS) as well as an assent (for those 10-16 years of age) or consent form (for those 16-18 years of age). Those who agreed to participate in the study signed the assent/consent from and provide their contact information. The lead researcher (SA) then contacted the participant or parent/carer to confirm eligibility, discuss the PIS, answer any questions about the study and arrange the time of interview. The demographic information alongside the SRS-22r questionnaires were completed via web survey prior to commencement of the interview.

4.3.9.2 HCPs group

Participants were invited to participate via email. The PIS and consent forms were sent to those who were interested in participation in the study. Those who agreed to participate signed and returned consent form electronically and they completed their demographic information prior to commencement of interviews via web survey.

for member checking. This is to promote accuracy, trustworthiness, and participant empowerment, as well as to enable for the addition of any further details. Participants were given two weeks to review the transcripts and suggest any changes / additions.

Table 4.1: Topic guide for cognitive interview

Introduction: In this part of interview, we want to discuss the contents of the SRS-22r if it is suitable to adolescent with idiopathic scoliosis. I want you to read each question one by one to me. Then, please tell me in your own words what do you understand?	
Purpose	**Example of probing questions**
Relevance	Do you think this question is relevant (important) to you?
Comprehension	<u>Questions</u> Could you tell in your own words what did this question is asking you about ?/ What went on in your mind when you read the question?/ How did you come to this answer?/ Are there any words that you don't understand?/unclear to you?
	<u>Response options</u> Please read each response choice and tell me what it means to you? What caused you to choose this response?/ was it easy or hard for you to choose an answer?/ Do you think the response options are appropriate to the question ?/ What are your suggestions to change the response options?
Recall period	Do you remember what happens to you during this time frame ?/Do you think the time frame is appropriate to you ?
Comprehensiveness	What other experiences do you have that are not covered in this questionnaire? Can you think of other areas that is missing in the questionnaire that you think it should be included in the questionnaire? / Is there anything we forgot to ask?

4.4 Data analysis

The main aim of cognitive interviewing is to confirm the relevance, comprehensibility and comprehensiveness of the SRS-22r by identifying potential problems and determine whether a question is interpreted as intended (Terwee et al., 2018b, Miller et al., 2014). This process helps to inform decisions around keeping, deleting, or modifying questionnaire items. Items that are consistently reported as comprehensible, and relevant by both groups (AIS and HCP) were suggested to be retained, others were suggested to be removed or modified (Knafl et al., 2007). If two or more participants raised concerns with the same question, it was considered as a criterion indicating that the item needed to be modified or revised (Brod et al., 2009). To determine comprehensiveness, participants were asked if the PROM content adequately covered the construct targeted by the PROM subscale. If any concepts were identified as missing, participants were asked to provide examples of these missing constructs. The identified missing constructs were subsequently presented in the results.

The data from AIS group interviews were analysed using thematic content analysis per COSMIN suggestions (Terwee et al., 2018b). In this study, this approach was slightly modified because individuals with AIS were not talkative, tended to give short answers and were reluctant despite prompting to elaborate on their responses and give further details. Therefore, their qualitative data were organised by counting and summarised in an Excel spreadsheet and presented in figures in this study. This was supported with quotations from participants when possible. This approach has been used previously in a recent study (Arbuckle et al., 2022). To examine variation in responses across sub-groups of participants with AIS, a stratification analysis was performed. This was to avoid any potential bias in the results, and to identify and issues that may be unique to a specific group of participants (Brod et al., 2009).

Data from the interviews of the HCPs was coded and analysed using thematic analysis (Braun and Clarke, 2006, Terwee et al., 2018b). This involved several steps, including familiarisation with the data through repeated reading of the content, generation of initial codes, grouping of similar codes into themes, review of themes to ensure they accurately reflect the data, and finally, defining and naming these themes and producing the report (Braun and Clarke, 2006).

4.5 Trustworthiness of the results

Many strategies have been employed to enhance trustworthiness of the study results (Nowell et al., 2017). The interviews were conducted by an experienced musculoskeletal physiotherapist researcher (SA), who was supported by co-authors with expertise in spinal problems and qualitative research. A spinal surgeon with experience working with AIS and a patient and public involvement (PPI) representative (ER) were integral to the study team. All participants were informed about the professional background of the interviewer and the nature of the study being part of a PhD thesis. The research team participated in data analysis enhancing the robustness of the interpretations (Nowell et al., 2017). The PPI representative was part of the study management group, and her feedback had been sought on the study protocol, the topic guide as well as was involved in interpretation and validation of the study findings.

4.6 Results

4.6.1 Participants

A total of 18 individuals participated in the study, participants with AIS (n=11) and HCPs (n=7). The mean duration of the interviews was 49 minutes (SD= 9.7). (*See Table 4.1*) for full details

of participant characteristics. One participant from the AIS group felt unable to participate at the beginning of their interview and withdrew their consent to be in the study; their data were withdrawn.

Table 4.2: Description of study participants

Characteristics	Individuals with AIS	HCPs
Sample size (n)	11	7
Age, years, mean (SD)	14.9 (1.8)	42 (7.9)
Gender, (n)	F (8) M (3)	F (4) M (3)
Large curve size, mean (SD)	47.5° (18°)	NA
Management (n) **Observation** **Brace** **Pre-surgery** **Post-surgery**	 2 2 4 3	NA
SRS-22r, mean (SD) **Function** **Pain** **Self-image** **Mental health** **Satisfaction** **Subtotal** **Total**	 4.3 (.6) 3.8 (.9) 3.4 (.8) 3.4 (.9) 3.5 (.7) 3.8 (.6) 3.7 (.5)	NA
Highest qualification, (n) **Bachelor** **Master** **Doctorate**	NA	 2 4 2
Abbreviations: SRS: scoliosis research society questionnaire, HCPs : Health Care Professionals, F: Female, M: Male, SD: Standard Deviation, NA: not applicable,		

4.6.2 Cognitive interviews (AIS group)

Participants provided feedback on the three aspects (i.e., relevance, comprehension, and comprehensiveness) of the questionnaire, and findings are presented visually and supported with a quotation example.

4.6.2.1 Relevance

In general, participants with AIS found that the SRS-22r questions were relevant, however, some items were reported as irrelevant by at least two participants (*see Figure* 4.1). Two questions from the self-image domain were reported as irrelevant by three participants, (6: How do you look in clothes? and 19- Do you feel attractive with your current back condition?)

> *"I don't know how much the back condition has to do with it. Some people might not feel attractive but it's just not due to their back. They just don't feel attractive in general"* (P9, Observation, 14)

Two participants reported that question (13- have you felt calm and peaceful during past 6 months?) in the mental health domain was irrelevant. Additionally, one participant stated that question (16- In the past 6 months have you felt down hearted and blue?) was also irrelevant.

> *'I just don't see how helpful it is in terms of knowing how calm and peaceful I've been'* (P6, Pre-surgery, 17)

One question in the function domain was reported as irrelevant by two participants (15- Are you and/or your family experiencing financial difficulties because of your back?).

"This is the one question that I never really understood. Because we've never put money towards getting better .... Everything's been on the NHS". (P8, Post-Surgery, 16)

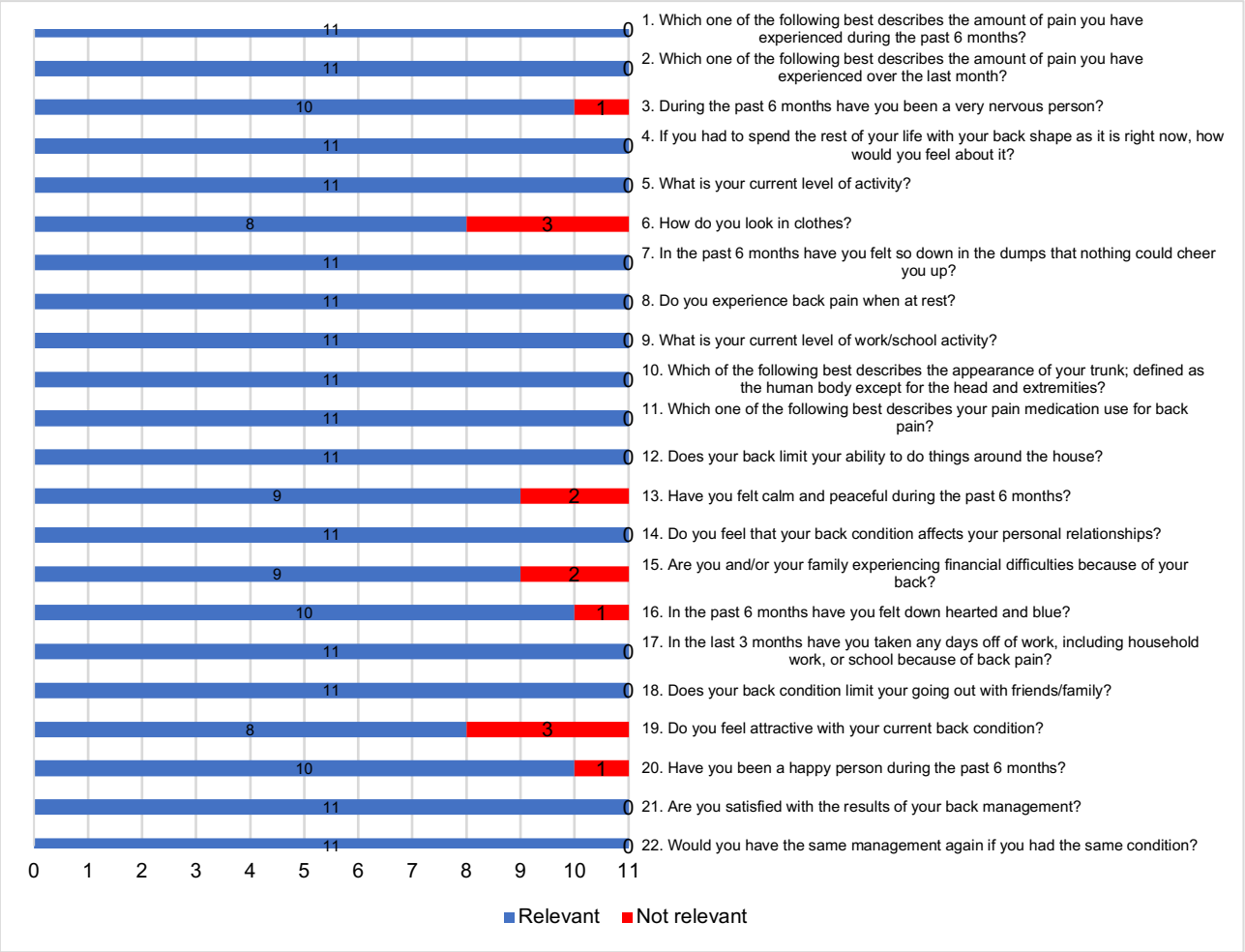


Figure 4.1: Summary of cognitive debriefing results: relevance of the SRS-22r questions

4.6.2.2 Comprehension

Compared to the relevance, more participants reported difficulty in understanding the questions (*see Figure* 4.2). Participants reported that the terminology used in mental health

questions as incomprehensible. For example, four participants found (Q20: Have you been a happy person during the past 6 months?) as incomprehensible.

> *"I don't really understand because... I just don't…. I think like what does it mean by am I generally happy? Am I happy with my back? Am I happy in my life?"* (P8, Post-Surgery, 16)

Two questions in the self-image related domain were reported as incomprehensible. For example, 6 out of 11 participants found Q10 as ambiguous and confusing (Q10- Which of the following best describes the appearance of your trunk; defined as the human body except for the head and extremities?)

> *"I didn't understand what trunk mean". "I don't really understand what that's got to do with my back really".* (P4, Observation, 12)

Two participants reported difficulty in comprehension of the satisfaction questions. Function and pain domain questions were reported to have relatively better comprehension compared to other questions.

> *"What… does that mean? I guess like physiotherapy, is that back management? …it is weird to see… when I think of management, I think like, ...managing like a club or a school ... so, I do not really link it straight to treatment, it is a bit confusing"* (P6, Pre-Surgery, 17)

“

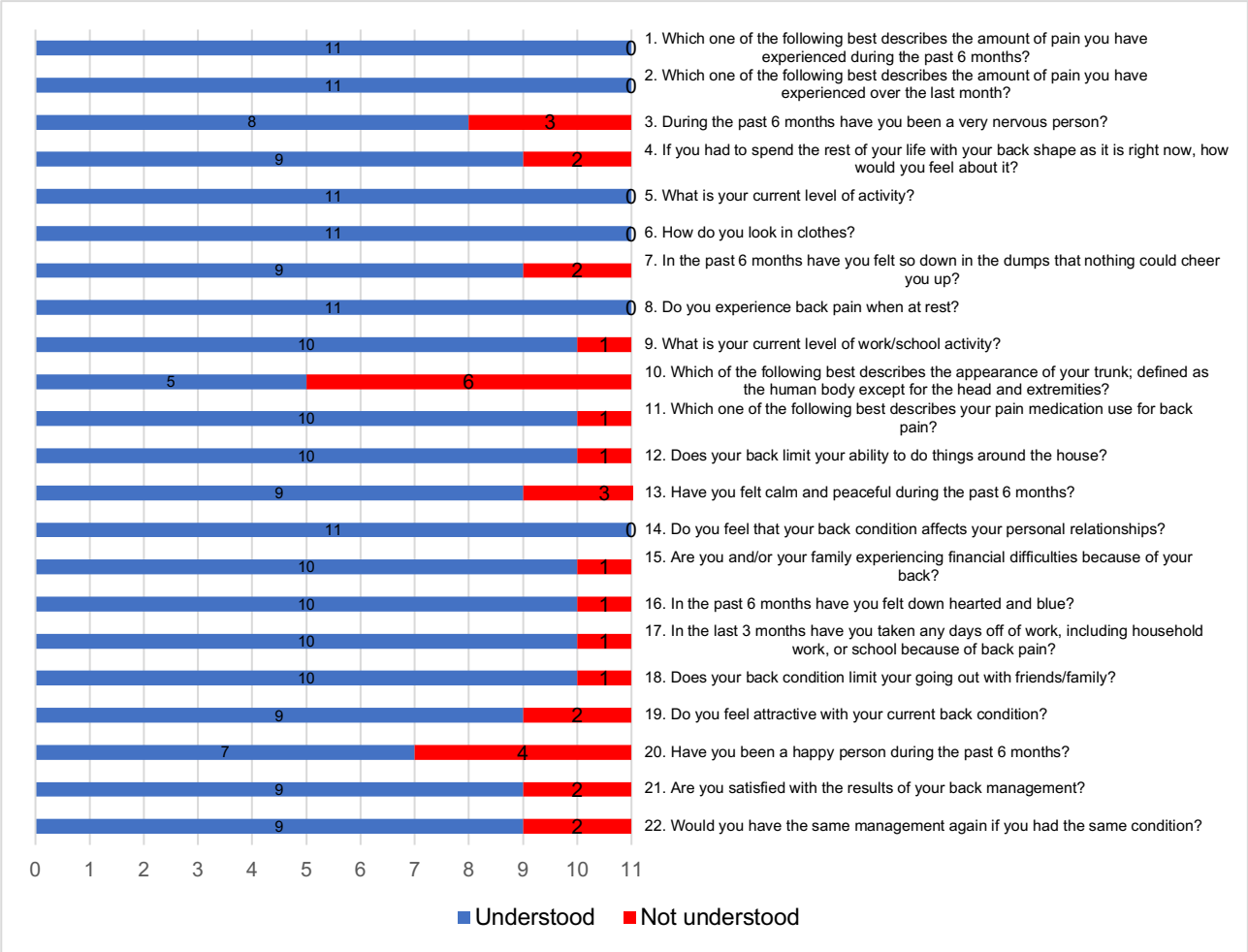


Figure 4.2: Summary of cognitive debriefing results: Comprehension of the SRS-22r questions

Many problems were identified regarding the comprehension of the response options compared to the questions themselves (*see Figure* 4.3). Lack of understanding of words such as ‘bedridden’ and‘ labour’ was reported by five participants. Also, six participants reported difficulty in understanding what was meant by ‘narcotics.

"This is a difficult question because I would say light or moderate labour, but I'm not doing light or moderate sports alongside it. I can't do sports …. The question is easy to understand and it's relevant, but the answers are a bit tricky". (P8, Post-Surgery, 16)

"I can't differentiate between non-narcotic and narcotic medication. I've literally not heard of them, is it some sort of back pain thing?" (P5, Post-Surgery, 16)

They also found that the response options denoting a moderate or severe answer as unclear and inconsistent between questions.

"I'd say it's weird that a moderate to severe, but not a mild to moderate, for example" (P1, Observation, 15)

"I don't know if it's relevant, but the order of them, from none to severely is flipped from the previous question…. That's just inconsistency" (P8, Post-Surgery, 16)

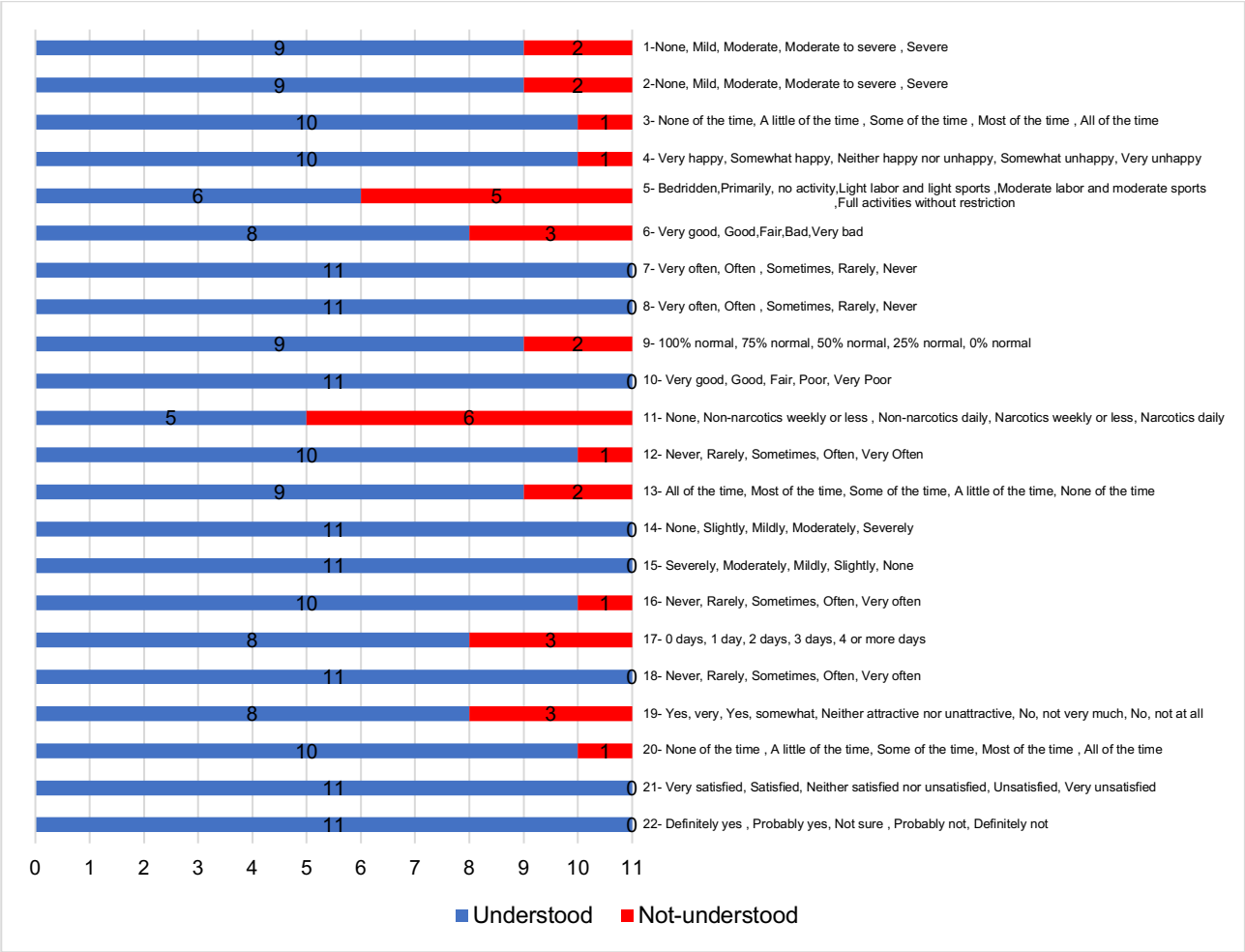


Figure 4.3: Summary of cognitive debriefing results: Comprehension of the SRS-22r response options

Regarding recall periods, two participants who had surgery found that a recall period for the past 6 months for questions 1 and 13 as an excessively long long-time scale over which to assess their pain experience, as it differed a lot during this time. (*See figure* 4.4.)

> *"I didn't know it said six months. Again, it's a difficult question because in the last two or three months I've been doing a lot better because of the surgery, the question is easier to understand but the time period, for me, is a bit awkward?"* (P8, Post-Surgery, 16)

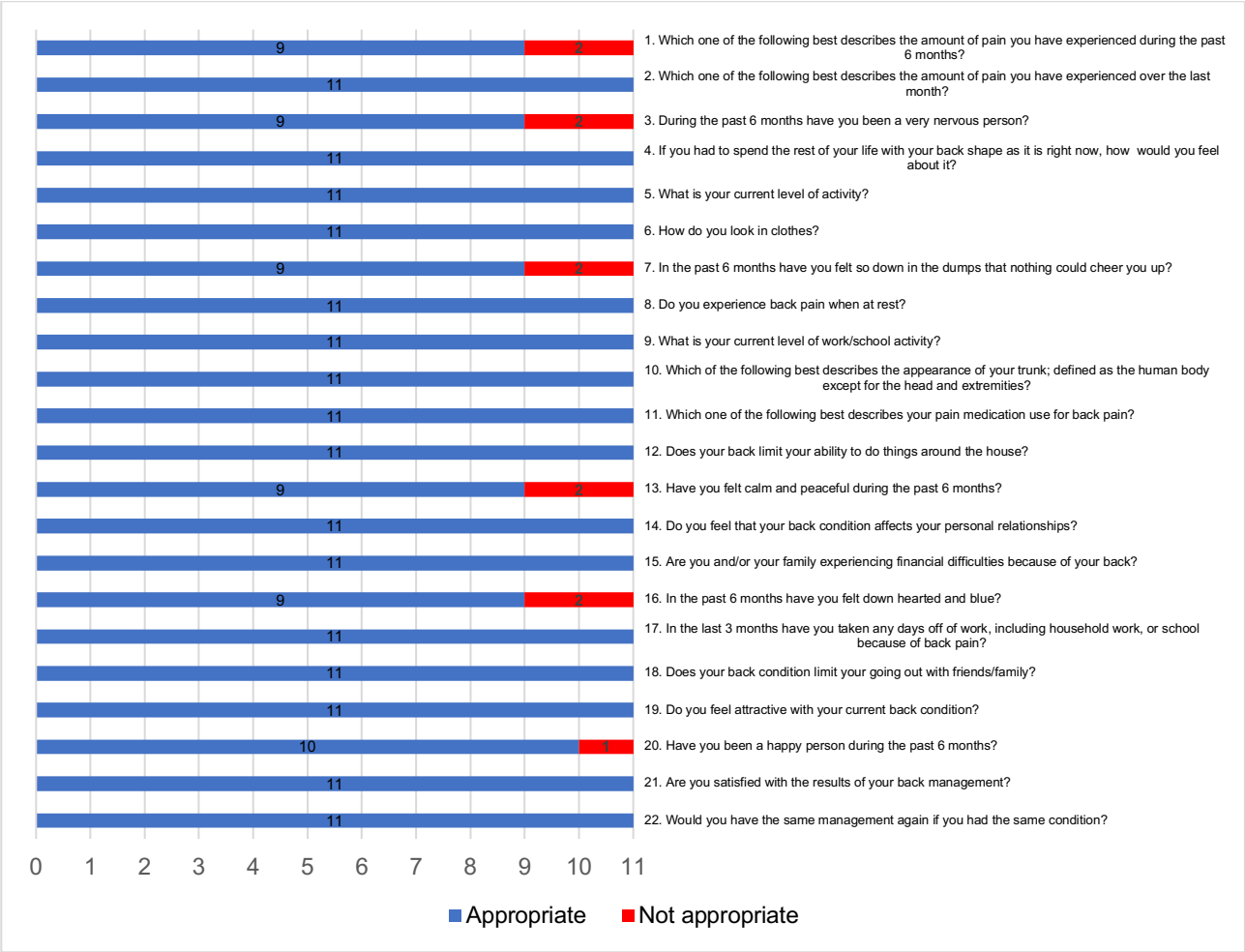


Figure 4.4 :Summary of cognitive debriefing results: appropriateness of recall periods of the SRS-22r questions

4.6.2.3 Comprehensiveness

Participants reported that the questionnaire covered most of the major areas relating to their HRQOL. However, they suggested addition of questions related to performing of activities such as sport and school activities. Adding questions about knowledge of scoliosis and treatment options was also suggested.

"What type of sports it can limit or what other activities, something to do with friends, what you can and can't do with them, possibly to do with family and how they think about it and how you think others view it, how many breaks you would have to take during school days or how maybe it affects your friends and how they think of it?" (P1, Observation, 15)

"I guess asking if you've had a variety of options presented to you instead of just surgery or back brace. I think asking if they've really explored the options before going into surgery, I think is important" (P6, Pre-Surgery,17)

4.6.2.4 Stratification analysis

Table 4.2 shows the distribution of problems reported by participants according to their age and management approaches. Reported problems in the relevance and comprehension aspects of the SRS-22r items were distributed equally among participants with different characteristics. An exception to this is participants managed by observation did not report any concerns with relevance of SRS-22r items. Recall periods problems were reported by participants who are 16 y/o and at post-surgery status. Suggestion to include more items about activities and knowledge were reported by two participants who were 15 and 17 y/o manged by observation and waiting surgery, respectively.

Table 4.3 : Distribution of SRS-22r reported problems according to participants characteristics

Classification	Relevance (n)	Comprehension (Questions & response options) (n)	Recall periods (n)	Comprehensiveness (n)
Age categories				
10-12	1	2	-	-
13-15	2	2	-	1
16-18	4	4	2	1
Management approaches				
Observation		2	-	1
Brace	2	2	-	-
Pre-surgery	2	2	-	1
Post-surgery	2	2	2	-

4.6.3 Cognitive interviews (HCPs group)

Participants critically evaluated the content of the SRS-22r questionnaire, and three main themes emerged. *See Figure* 4.5. presents the themes and subthemes which emerged from interview data for each aspect assessed in the questionnaire.

4.6.3.1 Relevance

Three subthemes emerged from data related to the relevance or otherwise of the SRS-22r questions for those with AIS.

Specificity: Mental health questions were reported as being generic questions with no relation to the back or to individuals with AIS.

"*Q20: Have you been a happy person during the past six months?*"

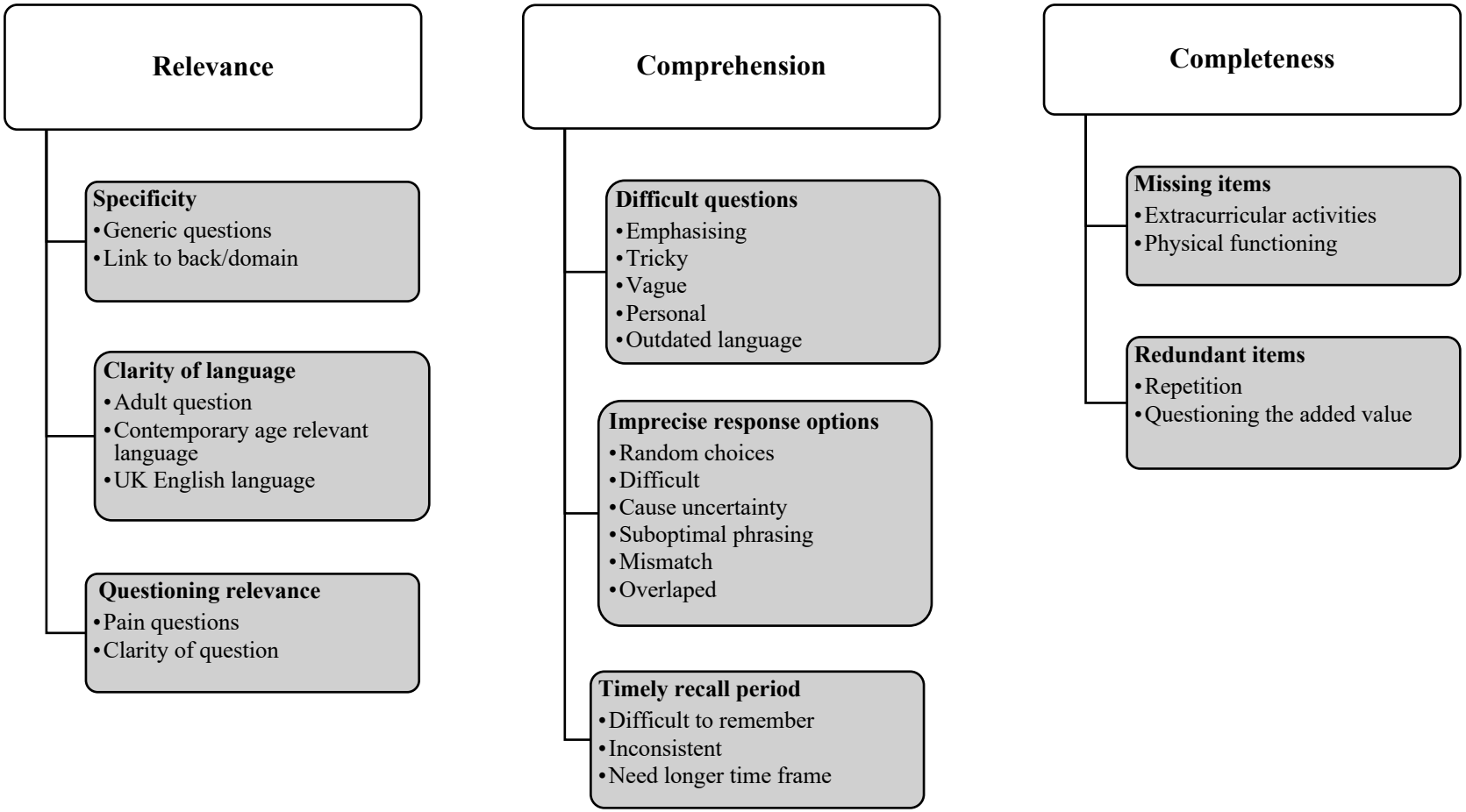
"Well, no, we've been in a pandemic. It's been horrible. It's got nothing to do with the spine. It is what we are asking and specifically in relation to your scoliosis in your back…It's too generic" (P7, Nurse).

"I don't love that question. I don't think it tells us whether it's about their back or anything else that's happening in their life". (P2, Physiotherapist)

Participants reported that some questions do not relate to a particular domain. For example, question 18 (Does your back condition limit you going out with friends and family?) is a question from the function domain but was assumed to be a self-image question.

"That's interesting, isn't it? I would have said it's definitely self-image and not activity" (P3, Surgeon)

Figure 4.5 :Themes and subthemes which emerged from the interviews with health care professionals



Clarity of language. Participants reported that some questions are more suited to an adult population and young people could struggle to respond/comprehend those questions. Further, the language use was identified as being a language of an older generation and not related to the language of contemporary adolescents. Participants also discussed that some questions might be not understood by children living in United Kingdom as the language used in the SRS-22r is US English.

> *"Q12: Does your back limit your ability to do things around the house?"*
>
> *" You're talking to a teenager. I'm sure they do the washing and the laundry (laughs), the washing up. It's just one of those questions that I think is very much set at an adult population. That's a very adult population kind of question" "* (P1, Nurse)
>
> *"Again, does that reflect that it's an Americanism rather than a UK language specific? Everyone assumes that UK English and American English are the same, and they are not "(*P3, Surgeon)

Questioning relevance. Participants reported that AIS is not generally a pain generating condition, therefore they questioned the relevance of pain questions. Further, they questioned the relevance of information obtained from the asking of some questions as no clear relevance to AIS was identified.

> *Q13: Have you felt calm and peaceful during the past six months? "I would question the relevance of the question and the wording. I am not really sure what they are trying… whoever it was that developed this, I'm not really sure what they are trying to find out from this question"* (P7, Nurse)

4.6.3.2 Comprehension

Difficult questions. Participants discussed many problems relating to the wording of questions as being negative, emphasising, sensitive and personal. They also found that some questions were

complicated and difficult to understand by an adolescent as the language used in those questions is outdated and vague.

> *"Q4: "If you had to spend the rest of your life with your back shape as it is right now, how would you feel about it? "I think that's emphasising, "Oh gosh, you look awful, wow, you've got to do something about that"* **(**P1, Nurse)

> *Q10: "Which of the following best describes the appearance of your back? Yeah, it's just your back isn't it. I suppose you want your chest. Well, these are really tricky."* (P2, Physiotherapist)

Improper response options. Participants found that some response options were unclear, leading to uncertainty and difficulty in understanding, due to poor word choices or arbitrary selection. They also noted that some response options did not match the question, or there was an overlap.

> *"'narcotics' implies something illegal, doesn't it, I don't know whether children or adolescents would hear of it, or whether or not parents might look at it and be a bit horrified, but I don't think that's quite a nice word to use"* (P5, Physiotherapist)

Timely recall period. The recall period of 6 months was identified as a long –time, and the participants with AIS found it difficult to remember all events that had occurred during that period. Participants reported inconsistency between the recall periods across questions asked in the questionnaire, i.e., the majority of questions assessed a 6-month period except for one question where it was 3 months. The response options for the question that asked about the person missing work or school days were found to need a longer period.

> *"Four days or more, I have some patients and they're literally spending half of their time at home. I mean four days is not even… a drop in the ocean. I think that actually is maybe the timeframes, they need to be made bigger"* **(**P1, Nurse)

4.6.3.3 Completeness

Missing items. Inclusion of items pertaining to extracurricular activities and physical functioning was identified as valuable or relevant by participants. Questions around patients own goals or expectations from management was also identified as being relevant.

> *"Are you able to do all the extracurricular activities you want to do? Be that after school clubs or be that going to the gym when you want to go to the gym or going horse riding"*(P7, Nurse)

> *"Maybe you could ask a few more functional questions, like, "Can you enjoy swimming?" or, "Can you enjoy cycling in the park?"* (P4, Surgeon)

> *"That's important. We're not asking them at all what their goals are and whether they've achieved them. I think it just needs to be some more subjective measurements on what's your goal. How much of the time can you achieve your goal or at all".* (P2, Physiotherapist)

Redundant items. The participants of this study mentioned a repetition of the mental health questions. They questioned the added value of asking the same questions more than one time.

> *"Again, this is a very similar question to one previously asked, so I don't know if it's… I don't know. It's like asking the same question again"* (P6, Surgeon)

> *"I don't know if it adds any value, because if the previous question has given you the answer about how satisfied they are, then that's probably more relevant information, compared to that one."* (P5, Physiotherapist)

4.6.4 Modifications suggested to SRS-22r

Both groups of participants suggested and provided examples of modifications and improvement to the content of the questionnaire, by replacing words or deleting some of the questions. All suggestions made by the participants, based on each domain of the SRS-22r questionnaire, are summarised in Table 4.3.

- **Pain**

Participants suggested that the response options for pain questions should be presented in a numerical or visual pain scale format to make it easier to comprehend. The recall period should be shortened or replaced with "since symptoms started" to improve accuracy. In addition, terms such as "narcotics" and "household work" should be removed as they are not relevant to adolescent life. Examples of questions and response options are presented in Table 3.

- **Function**

Participants suggested asking individuals with AIS about activities related to their activity and exercises they performed at school. Additionally, they recommended removal of the “financial difficulties” question and use of the terms “bedridden”, “around the house” and “labour” from response options. Suggested examples of questions and response options are presented in Table 2.

- **Self-image**

Participants of this study raised many concerns regarding questions within this domain. They suggested revising the wording of the questions and response options, to make them more focused and understandable to those completing the questionnaire; given that the current questions within this

domain introduced and emphasised negativity. Participants suggested replacement of “If you had to spend the rest of your life with your back shape as it is right now, how would you feel about it?” by “How does the shape of your back make you feel?”. Also replacing “Do you feel attractive with your current back condition?” with "Do you feel different from other people due to your current back shape?”. Some participants suggested deletion of “How do you look in clothes”, while other suggested replacement with “How confident do you feel about your body?”. It was suggested that the term “trunk” should be replaced by “postural appearance” or include a picture that shows the back. Other suggestions that the term “personal relationship” to be replaced with “friendship” to make it more relevant to adolescents.

- **Mental health**

Both groups of participants reported that the questions of this domain are not related to the back and that there is a lot of repetition, and as such this domain should be minimised to one or two questions. For example, “Which emotions have you felt over the last six months because of your back? The response options could then include calm, peaceful, nervous, anxious, etc. This would then be followed by question about the frequency of these feelings.

- **Satisfaction**

Participants suggested replacement of the word “management” by “care” or “treatment” and “back” by “scoliosis”, and that the response options would only be “yes” or “no”. Some participants also suggested that one question would be sufficient to assess satisfaction about .

Table 4.6: Examples provided by participants to refine the questions, response options, and recall periods of the SRS-22r

Domains	Questions	Response options	Recall periods
Pain			
1- Which one of the following best describes the amount of pain you have experienced during the past 6 months? • **None** • **Mild** • **Moderate** • **Moderate to severe** • **Severe**	• How much back pain have you been experiencing? • How much back pain have you been experiencing last month?	• VAS-type scale • Smiley faces • Pain scale 0-10	"Since your symptoms started"
2- Which one of the following best describes the amount of pain you have experienced over the last month? • **None** • **Mild** • **Moderate** • **Moderate to severe** • **Severe**	• How much back pain have you been experiencing last month? • Are your symptoms improving or worsening? • How many times did you feel pain because of your back?	• VAS-type scale • Smiley faces • Pain scale 0-10 • Once in a week, one to two times a week, or every day"	"Since your symptoms started"
8- Do you experience back pain when at rest? • **Very often** • **Often** • **Sometimes** • **Rarely** • **Never**	• No changes suggested	• VAS-type scale • Smiley faces • Pain scale 0-10	

11- Which one of the following best describes your pain medication use for back pain? • **None** • **Non-narcotics weekly or less (e.g., aspirin, Tylenol, Ibuprofen)** • **Non-narcotics daily** • **Narcotics weekly or less (e.g., Tylenol III, Lorcet, Percocet)** • **Narcotics daily**	• Do you need to use pain medication on a regular basis, if so, how often and what is it?' • Which pain relief do you take and the name of it? How often do you take this and what dose? • Do you take pain relief for your back? yes or no. How regularly? • Which painkillers are you taking for your back? How often are you taking them on average?	• None, • Non-opioids such as paracetamol, aspirin, or ibuprofen. • Weak opioids such as codeine • Strong opioids such as morphine • Every day, like four times a day, twice a day, every day, weekly, only when I have pain, never.	
17- In the last 3 months have you taken any days off of work, including household work, or school because of back pain? • **0 days** • **1 day** • **2 days** • **3 days** • **4 or more days**	• "Delete household work". • How many days of school have you had off because of your symptoms/back pain? • How does scoliosis impact your school life and how often you go there?	• None • 1 to 3 days • 1 -2 weeks • 1-2 months	
Function			
5- What is your current level of activity? • **Bedridden** • **Primarily no activity** • **Light labor and light sports** • **Moderate labor and moderate sports** • **Full activities without restriction**	• How often do you exercise?" • How much you do and how fully are you doing things?	• Once a week, two times a week, etc. • Short walks, longer walks of over an hour Running, Weekly sports clubs, 30 minutes of light walking a week • I don't play sport at school / I do PE, but I don't do clubs outside of school / I play sport on a Saturday / I'm always playing sport all the time	

9- What is your current level of work/school activity? • **100% normal** • **75% normal** • **50% normal** • **25% normal** • **0% normal**	• Delete /remove • Activities at school or activities that they would normally do outside of school, normal sports.?	• Have number rather than percentage , scale rather than categories.	
12- Does your back limit your ability to do things around the house? • **Never** • **Rarely** • **Sometimes** • **Often** • **Very Often**	• Does your back limit your ability to do day-to-day activities? • Does your back limit your ability to socialise with friends, or participate in hobbies? • Does your back pain limit your ability to take part in your usual activities? • Is your back stopping you from doing things around the house? put example about around the house		
15- Are you and/or your family experiencing financial difficulties because of your back? • **Severely** • **Moderately** • **Mildly** • **Slightly** • **None**	• This should be just taken out completely/delete		
18- Does your back condition limit your going out with friends/family? • **Never** • **Rarely** • **Sometimes** • **Often** • **Very often**	• How have the activities you did with your friends and your family changed because of your back? • Does having scoliosis prevent you going out with friends or family?		
Self-image			

4- If you had to spend the rest of your life with your back shape as it is right now, how would you feel about it? • **Very happy** • **Somewhat happy** • **Neither happy nor unhappy** • **Somewhat unhappy** • **Very unhappy**	• Would you consider changing your back shape? • Would you want an operation to change the shape of your back? • How willing are you to consider an operation? • How does the shape of your back make you feel? • As of today, how do you feel about the shape of your back?	• Scale zero to 10 (10 being very happy, zero being very unhappy), • Yes, No • 'Very happy' and 'very unhappy'	
6- How do you look in clothes? • **Very good** • **Good** • **Fair** • **Bad** • **Very bad**	• How do you feel in your clothes? • How self-conscious, how confident do you feel about your body? • What sort of clothes do you feel comfortable wearing? • How do you feel about your appearance?		
10- Which of the following best describes the appearance of your trunk; defined as the human body except for the head and extremities? • **Very good** • **Good** • **Fair** • **Poor** • **Very Poor**	• Which of the following best describes the appearance of your posture? • Draw picture to show trunk and ask which part of your body worries you or concerns you or that makes you sad?		
14- Do you feel that your back condition affects your personal relationships? • **Never** • **Rarely** • **Sometimes** • **Often** • **Very often**	• Does the shape of your back affect your friendship ? • Do you feel that your back affects your relationship with your friends? • Do you think your scoliosis affect your relationship?	• No, slightly, mildly, moderately yes completely.	

19- Do you feel attractive with your current back condition? • **Yes, very** • **Yes, somewhat** • **Neither attractive nor unattractive** • **No, not very much** • **No, not at all**	• Do you feel different from other people due to your current back shape? • How conscious do you feel about your postural appearance? • Do you like the way your back shape looks? • DO you feel different from other people due to your current back condition?	• zero to 10, very self-conscious, not self-conscious at all.	
Mental health			
3- During the past 6 months have you been a very nervous person? • **None of the time** • **A little of the time** • **Some of the time** • **Most of the time** • **All of the time**	• During the past 6 months have you felt anxious? • In the last months, how best describes how you've been feeling? Then a reason?	• Confident, anxious, depressed, etc..	Change into different time period
7- In the past 6 months have you felt so down in the dumps that nothing could cheer you up? • **Very often** • **Often** • **Sometimes** • **Rarely** • **Never**	• Have you felt depressed? Have you felt low in mood, low self-esteem? • In the past six months have you felt so low about your back that nothing could cheer you up?		Change into different time period
13- Have you felt calm and peaceful during the past 6 months? • **All of the time** • **Most of the time** • **Some of the time** • **A little of the time** • **None of the time**	• Are you low in mood or have you had any anxiety or worry issues? • Which emotions you felt over the last six months? How often do you feel these? what is the main source of your feelings?	• Calm, peaceful, nervous, anxious, happy	Change into different time period

16- In the past 6 months have you felt down hearted and blue? • **Never** • **Rarely** • **Sometimes** • **Often** • **Very often**	• Put all different emotions in one question, tick which emotions you've been feeling? • Does the shape of your spine or your spinal deformity, has it made you feel, or has it affected your mental health?	• None, 3 days, 1-2 weeks, 1-2 months	Change into different time period
20- Have you been a happy person during the past 6 months? • **None of the time** • **A little of the time** • **Some of the time** • **Most of the time** • **All of the time**	• Do you feel happy about your back?		Change into different time period
Satisfaction			
21- Are you satisfied with the results of your back management? • **Very satisfied** • **Satisfied** • **Neither satisfied nor unsatisfied** • **Unsatisfied** • **Very unsatisfied**	• Are you satisfied with the treatment (care) you've had?	• Yes, no	
22- Would you have the same management again if you had the same condition? • **Definitely yes** • **Probably yes** • **Not sure** • **Probably not** • **Definitely not**	• Would you recommend this treatment to somebody else who has scoliosis?"	• Yes, no	

4.7 Discussion

This study is the first qualitative evaluation of the content validity of the SRS-22r questionnaire for individuals with AIS which was achieved through cognitive debriefings. Through in-depth, semi-structured interviews with both individuals with AIS and their HCPs, this study sought to understand their perspectives on the content of the SRS-22r. Overall, individuals with AIS reported that two self-image and two mental health questions, along with the questions around financial difficulties, were irrelevant. Three subthemes emerged related to the relevance from the HCPs interviews: the questions were too general, the questions used irrelevant language (e.g., adult-oriented questions), and questions were unclear in relevance to AIS. Both individuals with AIS and HCPs expressed a variety of concerns regarding the comprehensibility of the questions posed, the response options and the length of the recall periods, highlighting issues such as vagueness and negative wording. Participants also noted that the SRS-22r lacked questions pertaining to sport and school activities, which based on their experiences are important concepts related to individuals with AIS. To address these concerns, participants provided examples of potential changes to the wording of both the questions and the response options that could improve the content of the SRS-22r questionnaire. However, it is important to note that any proposed revisions made to the questionnaire must be evaluated in a pilot study before formal adoption.

4.7.1 Relevance

Relevance assessment ensures that the PROM sufficiently assesses all important areas of the health condition and treatment (Matza et al., 2013, Terwee et al., 2018b). The problem with financial difficulties question has been identified previously, with a reduction in the internal consistency of the function domain when the questionnaire is adapted into other languages (Asher et al., 2006). This

issue seems relevant to individuals with AIS living in the UK, who are treated by the publicly funded NHS system with a 'free at the point of care' model of healthcare delivery. The HCPs highlighted that this question adds unnecessary worry to individuals with AIS who already felt anxious, and afraid due to the potential need for surgery alongside their future with scoliosis (Rullander et al., 2017, Essex et al., 2022, Motyer et al., 2022). This has also reported by the parents of individuals with AIS in other studies (Donnelly et al., 2004, Motyer et al., 2021a, Bull and Grogan, 2010). Attractiveness and clothes questions were reported as being irrelevant to the adolescent's experience with scoliosis as well as a mental health question, as they lacked necessary reference to the back by being too generic and not specific enough to AIS. This may be attributed to the fact that the question was originally derived from the SF-36 questionnaire, which was primarily designed for adults rather than adolescents with AIS (Asher et al., 2006, Asher et al., 2000). Contents of HRQOL questionnaires should be tailored to the experiences, activities, and environments that are specific to the target population's age (Matza et al., 2004, Lohr and Zebrack, 2009), especially for adolescents who have unique developmental and psychological needs (Matza et al., 2013, Matza et al., 2004). Recommended practice emphasises the use of age-appropriate language when developing questionnaires for children and adolescents, simplifying the language to meet their understanding level (Matza et al., 2013, Matza et al., 2004). This can be achieved by using the same language that individuals with AIS use during concept elicitation interviews (Matza et al., 2013).

4.7.2 Comprehension

Both group of participants encountered difficulties in comprehension of questions due to negative wording, emphasis aspects, sensitivity, or vagueness, all of which could potentially impede the participants understanding of the questions. When developing a questionnaire to determine the age appropriateness, it has been recommended to preform pilot testing and cognitive debriefings with

individuals from various age groups within the intended population (Matza et al., 2004). The mean age of participants in this study was 14.9 years old, which is considered an age where children can complete questionnaires independently (Matza et al., 2013, Matza et al., 2004). However, they needed assistance to complete some questions.

The type of response options for the SRS-22r involves a Likert scale, however, participants found it difficult to comprehend some of the response options, due to being either inconsistent or chosen arbitrarily. Since the SRS-22r has been developed for a broad range of ages, it might be appropriate to use a numerical pain scale, or pictures, instead of a Likert scale for pain severity questions, as it will be easier for an adolescent to understand. A recent review recommended using visually appealing scales to enhance acceptability among young individuals (Coombes et al., 2021). They reported that there is a preference in the literature to use scale constructed of 'faces' from sad to happy for response options for children at all ages (Coombes et al., 2021). The Likert scales were found more appropriate for older rather than young individuals (Matza et al., 2013). Further, the use of 3-point Likert scales functioned better in those 8–18 years old than 5-point Likert scales (Coombes et al., 2021).

It is essential to use appropriate language when discussing both time off school and pain medication with individuals with AIS. Responses that limit time off school to four days or more were deemed inappropriate for individuals with AIS. A study compared time to return to school following surgery among different orthopaedic conditions. Individuals with AIS underwent spinal fusion surgery were out of school the longest, with a mean of 42 days, and missed the greatest number of in-session school days (Willimon et al., 2019). Therefore, this study's participants suggested to include weeks and months along with days to accurately capture the extent of the absence.

The use of term “narcotic” in the response options for pain medications was reported as difficult to understand and inappropriate by both groups. It is crucial to use clear and age-appropriate language when discussing pain medication as using complex medical terminology may be confusing for children and their caregivers. The HCPs participants suggested use of the naming and classification of the analgesic (pain) ladder by the World Health Organization (WHO) (Anekar and Cascella, 2021), in which pain medications are classified according to the pain severity starting with non-opioids such as paracetamol for mild pain, weak opioids such as codeine for moderate pain, and strong opioids such as morphine for severe pain. This might facilitate understanding and communication between patients, caregivers, and HCPs (Anekar and Cascella, 2021).

The appropriateness of a six-month recall period for individuals with AIS was questioned by some participants in this study. The HCPs also noted that it could be challenging for adolescents to recall accurately and that they may require support from family members to do so. In a recent review of PROMs assessing children's HRQOL, it was found that many commonly used recall periods for PROMs were “the past four weeks” or “the past month” (Landraf et al., 2005). This indicates that the PROM developers were aware that a lengthy recall period is difficult for children when compared to adults (Matza et al., 2013, Matza et al., 2004). Moreover, PROMs that rely on patients to recall over a long period are likely to undermine content validity, as the response is likely to be influenced by the patient's state at the time of recall (Health et al., 2006). Thus, items with short recall periods are usually preferable.

4.7.3 Completeness

The SRS-22r covered key aspects of HRQOL related to individuals with AIS, such as pain, mental health, function, and self-image domains (Asher et al., 2006, Solans et al., 2008). However,

participants suggested including questions about sport and school related activities, which were identified as important to individuals with AIS (Kakar et al., 2017, Fabricant et al., 2012). A previous qualitative study reported that knowledge of scoliosis and the available treatment options is also important to this population (Motyer et al., 2021a, Motyer et al., 2022). Including such questions would provide valuable insights into the patient's level of understanding of their condition, which could have implications for their adherence to treatment plans, and overall management of their health condition (Motyer et al., 2021a).

4.8 Strength and limitations

This study utilised a rigorous methodology following COSMIN recommendations for evaluating quality of content validity study through cognitive debriefing interviews (Terwee et al., 2018b). It involved experts in conducting spinal and qualitative research along with a PPI representative. The study included individuals with AIS with diverse demographic and clinical characteristics, as well as HCPs with an average of 12 years of experience in managing individuals with AIS, allowing for a broad range of perspectives to be represented. Moreover, the fact that the majority of participants had no previous experience with the SRS-22r allowed for a more accurate assessment of their experience and opinions, without any influence from prior learning.

The study's main limitation was the recruitment of participants from a single centre in the UK, which may have introduced social desirability bias and limited the generalisability of findings. Ideally, cognitive debriefing is performed following concept elicitation to confirm that the concepts identified by the participants are adequately represented in the questionnaire (Alamrani et al., 2023a) However, the SRS-22r, which was developed by Haher et al. in 1999 (Haher et al., 1999), has been culturally adapted to various languages (Alamrani et al., 2021b), and has been used without prior

feedback from the relevant population. The traditional 4-stage cognitive model was not employed in this study (Willis and Artino, 2013). However, the COSMIN recommendations to assess cognitive debriefings was adhered (Terwee et al., 2018b). The cognitive debriefing approach is ideally an iterative procedure where a smaller initial round of interviews is conducted. Then, analysis is undertaken and subsequent alterations to questionnaire items are performed if needed. This would be followed by additional interviews with modified items facilitating ongoing refinement of questionnaire (Willis and Artino, 2013). However, in this study due to time constraints of the lead researcher (SA), only one round of interviews was conducted followed by the analyses. The refinement of questionnaire items and participants' suggestions for improvement of questions, response options and recall periods are presented in Table 4.4. Further testing with another round of cognitive debriefing interviews is still needed to further refine the SRS-22r questionnaire.

4.9 Implication for practice and future research

Findings have important implications for potential refinements of the SRS-22r, as it has identified potential issues in the relevance, comprehension, and comprehensiveness aspects of the existing questionnaire. It has highlighted areas where revisions and improvements are necessary, such as both the self-image and mental health domains. Findings also serve as a crucial reminder that the widespread use of a PROM does not automatically guarantee that it has a content validity. The testing of the content validity of this PROM improves the quality of the data collected from the patients as well as our understanding of the patient experience and outcomes. Future research can use our study findings to refine the SRS-22r or develop a new questionnaire that’s content is based on qualitative input directly from individuals with AIS.

4.10 Chapter summary

This chapter assessed the content validity of the SRS-22r with respect of its relevance, comprehension, and comprehensiveness. Using think aloud and verbal probing techniques, both individuals with AIS (n=11) and HCPs (n=7) participated in semi-structured interviews and critically evaluated the content of the SRS-22r. Findings provides important evidence that highlights areas for potential improvement in the SRS-22r content and structure. Some questions were considered to be irrelevant or unclear in relevance to AIS, and both groups reported issues regarding comprehensibility and the length of recall period. Participants also mentioned missing of questions about sports and school activities, both of which they thought were significant. Examples of proposed improvements to improve the questionnaire's content were provided.

Chapter Five

CONTENT VALIDITY OF THE SCOLIOSI RESEARCH SOCIETY-22 REVISED (SRS-22R): A QUALITATIVE CONCEPT ELICITATION STUDY

This chapter reports in full the contents of a published manuscript by the thesis author protocol (Alamrani et al., 2021a), and research article (Alamrani, et al., 2023). It includes verbatim text from the published manuscript and some changes in the introduction section were employed for the purpose of this thesis.

Publication and Conference presentations

- **Alamrani S.** Gardner, A., Falla, D., Russell, E., Rushton, A. B., & Heneghan, N. R. (2023). Content validity of the Scoliosis Research Society questionnaire (SRS-22r): A qualitative concept elicitation study. PloS one, 18(5), e0285538. https://doi.org/10.1371/journal.pone.0285538. (**Appendix 2**)

- **Alamrani, S.,** Gardner, A., Falla, D., Russell, E., Rushton, A.B. and Heneghan, N.R., 2021. Content validity of Scoliosis Research Society questionnaire-22 revised (SRS-22r) for adolescents with idiopathic scoliosis: protocol for a qualitative study exploring patient's and practitioner's perspectives. BMJ open, 11(12), p.e053911. (**Appendix 5**)

- **Alamrani S.,** Gardner A, Falla D. Russell E, Rushton AB, Heneghan NR. Content Validity of The Scoliosis Research Society (Srs22r): A Qualitative Concept Elicitation Study. Physiotherapy UK- 1st November 2023. Birmingham. United Kingdom. (**Poster presentation**).

5.1 Abstract

The SRS-22r is the common questionnaire used to evaluate HRQOL for young people with AIS. However, the impact of the AIS on HRQOL for adolescent as well as its associated management has not been evaluated before to inform development and content validity of the SRS-22r. Therefore, the aim of this chapter is to evaluate the content validity of the SRS-22r questionnaire by eliciting concepts that are important to HRQOL of individuals with AIS per their perspectives.

In-depth semi-structured interviews were conducted with a purposive sample of young people with AIS (Cobb angle ≥25˚, aged 10-18 years). Concept elicitation was used to evaluate the influence of AIS on participants' HRQOL. Participant information sheets and consent/assent forms were age relevant. Topic guide was informed by the SRS-22r and existing evidence. Interviews were audio and video recorded, transcribed verbatim, coded, and analysed using thematic analysis. Derived themes/codes were compared with SRS-22r contents (domain/items).

Eleven participants (mean age 14.9 years [SD=1.8]; 8 female) were recruited. The mean curve size was 47.5° [SD=18°] and participants had been managed via different approaches. Four main themes emerged with associated subthemes: 1) Physical effects related to physical symptoms (back hurt, stiffness) and body asymmetry (uneven shoulders), 2) Activity-related effects showed impact on mobility (sitting for long periods) , self-care (dressing), and school activities (focus during lessons), 3) Psychological effects revealed emotional (feel worried), mental (sleep quality), and body image effects (hide back from others), 4) Social effects (participation in school and leisure activities), and mental health support as well as from school, friends. A weak association was found between items of the SRS-22r and the identified codes.

The SRS-22r does not adequately capture important concepts that relate to HRQOL of adolescents with AIS. These findings support revision of the SRS-22r, or the development of a new patient reported outcome measure to evaluate HRQOL of adolescents with AIS.

5.2 Introduction

Findings from *Chapter three* shows that the SRS-22r is the most common questionnaire used among individuals with AIS, and it has been translated and culturally adapted into more than ten languages (Climent et al., 2005, Sathira-Angkura et al., 2012, Lee et al., 2011, Bago et al., 2004, Simony et al., 2016, Danielsson and Romberg, 2013, Antonarakos et al., 2009, Alanay et al., 2005, Cheung et al., 2007). The SRS-22r psychometric properties have been extensively studied (Asher et al., 2003b, Glattes et al., 2007, Asher et al., 2003a, Asher et al., 2006, Asher et al., 2000, Asher et al., 2003c). However, participants in these studies were older than 18 years, (range 19-34 years), and therefore they may not be representative of an AIS population. Furthermore, the mental health domain of the SRS-22r includes questions from the SF-36 survey, which is a generic questionnaire designed for adults (Asher et al., 2003b). Because adolescence is a distinct demographic age group with discrete emotional and social characteristics, it is essential that their PROM represents their developmental stage (Matza et al., 2013).

As has been stated in *section* 1.4, the COS for individuals with AIS has limitations in its applicability and development process (de Kleuver et al., 2017). It encompasses all individuals undergoing spinal deformity surgery, lacking specificity for individuals with AIS. The COS was not developed with the involvement of the targeted AIS population, and the non-English-speaking participants in the Delphi study raise concerns about the findings' applicability to English-speaking populations (de Kleuver et al., 2017). If the SRS-22r is recommended as a PROM within the COS, it should demonstrate good content validity, which ensures consistency with the population's perspective and experiences (Terwee et al., 2018b, Terwee et al., 2018c, James et al., 2018).

Qualitative research methods such as the use of concept elicitation methodology may usefully assess content validity by interviewing the population of interest to understand their perspectives regarding the impact of the disease on their own health condition (Toye et al., 2016, Brédart et al., 2014, Holman, 1993). A recent scoping review exploring the experiences of AIS and families about the diagnosis and treatment of scoliosis revealed a need for more qualitative research for this population (Essex et al., 2022). The majority of studies evaluated decision for treatment (Donnelly et al., 2004) and focused on the experience of surgery (Rullander et al., 2013), mainly its effects on the psychological aspects for the patient and parents (Bull and Grogan, 2010, Motyer et al., 2022).

To the best of our knowledge, no previous qualitative study has interviewed individuals with AIS to elicit concepts about their scoliosis and their impact on HRQOL. Evidence for content validity of the SRS-22r is therefore lacking. This chapter addresses the fourth objective of this thesis to determine whether the content of the SRS-22r questionnaire is relevant and important to participants with AIS, by exploring the impact of scoliosis and its associated treatment on HRQOL.

5.3 Methods

5.3.1 Design

This is a qualitative study design reported in accordance with the COREQ guidelines (Tong et al., 2007) (*see Appendix* 9). Additionally, it has been described in a published study protocol (See Appendix 2).

5.3.2 Qualitative approach and research paradigm

The world view of pragmatism was used in this study. It suggests that objective reality is grounded in the environment and accessible through human experience (Kaushik and Walsh, 2019). This approach emphasises the importance of selecting methods based on their appropriateness for addressing specific research questions rather than adhering strictly to philosophical debates (Ritchie et al., 2014).

The interpretive hermeneutic phenomenology was chosen as methodology as it allows an understanding that individual experiences are unique, rich and complex (Holroyd, 2007). The hermeneutic cycle facilitates an interplay between interpretation and tradition that extends beyond subjective vagueness or objective analysis (Holroyd, 2007). This is valuable in enhancing awareness for applying research in practical settings and gaining a deeper understanding of the context of illness and individual experiences (Holroyd, 2007). This approach facilitates an in-depth exploration of the lived experiences of individuals with AIS, enabling a comprehensive understanding and interpretive flexibility regarding HRQOL from the participants' standpoint.

5.3.3 Setting

Participants were recruited from a tertiary scoliosis centre in the United Kingdom. Semi-structured interviews were conducted virtually (using Zoom/ Microsoft Teams platforms). Written consents to participate in the study were collected from participants and parents by the research nurse, during their visit to the clinic. The lead researcher (SA) contacted the participant/parent, and the interview time was chosen based on participant/parent preferences. Data collection took place between January and April 2022.

5.3.4 Ethics approval

Ethical approval to conduct the study was sought from the Health Research Authority and Health and Care Research Wales approval (REC reference: 21/WM/0076), (*see Appendix* 9).

5.3.5 Reflexivity

The lead research (SA) conducted the semi-structured interviews. She is an experienced musculoskeletal physiotherapist researcher conducted the interviews, with support from specialist co-authors in spinal research (surgeon, physiotherapists), and qualitative research. No specific relationship was established prior to the commencement of the interviews and participants were informed about the professional background of the interviewer and that the study is part of a PhD thesis.

5.3.6 Participants

A heterogenous purposive sampling approach was used to recruit individuals with AIS aiming for n=10-15, which is deemed sufficient to reach concept saturation (Kerr et al., 2010). This approach enabled a wide range of experience and opinions to be explored and ensured diversity in the sample characteristics (Etikan, 2016). The sample size was decided by reaching saturation in the sub-themes ; therefore, a saturation table was created to ensure that concept saturation was achieved. At the point where two consecutive interviews failed to elicit any new information (*see Appendix* 9) (Kerr et al., 2010). Participants with a Cobb angle >25°, aged 10–18 years, and who had access to a video/audio call platform, were considered eligible. Individuals with other forms of scoliosis, and those who were unable to speak English fluently were excluded.

5.3.7 Semi-structured interview guide

A single semi-structured interview was conducted with individual with AIS. The topic guide for interview include assessment of the HRQOL. The HRQOL concept is a "complex, multidimensional concept, including social, emotional and physical functioning or well-being, related to the patient's health state" (Seid et al., 2004). To ensure that the concept of HRQOL was elicited from participants during the interview, a topic guide was developed using a hypothesised conceptual framework (Fig 5.1), as well as the recommended guidelines on the development of PROM for measuring HRQOL. The topic guide was age appropriate and consisted of open-ended questions that explored HRQOL as well as other concepts from the SRS-22r such as satisfaction. Ended by exploring other areas that are important to participants with AIS, such as participation in sport and other physical activities (*see Appendix* 9).

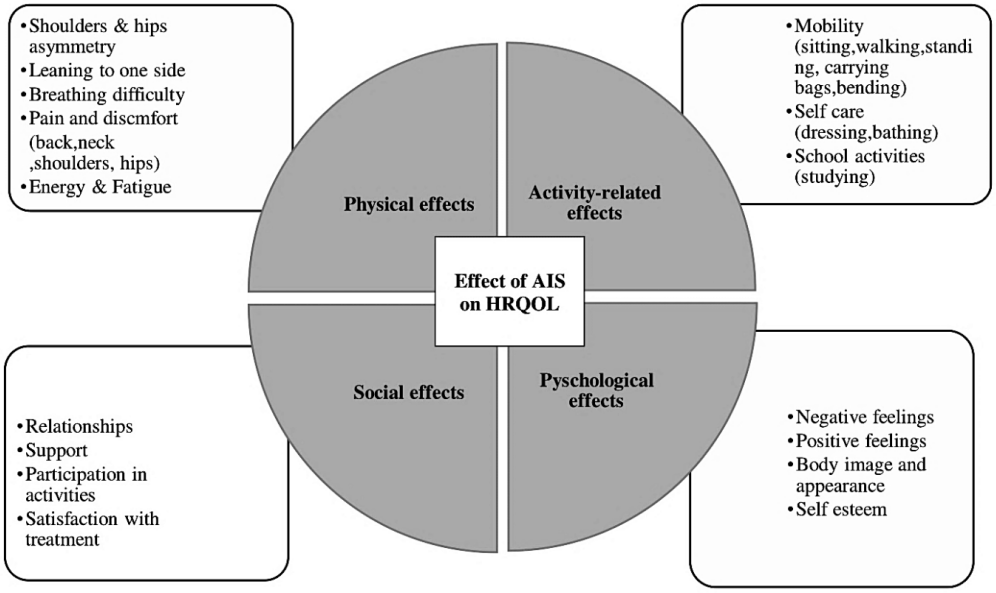


Figure 5.1:Hypothesised conceptual framework using HRQOL dimensions

5.3.8 Interview procedure

After conducting a pilot interview with an individual with AIS, adjustments were made to the topic guide to enhance the clarity of certain questions that cause confusion. Furthermore, details about confidentiality were explained and simplified. At the beginning of the interview, the interview format was explained, and any questions were answered. The confidentiality, voluntary participation and withdrawal process was also explained. Participants were encouraged to talk for as long as was needed during the interview, which lasted a mean of 49 minutes (SD = 9.7). Participants were unknown to the interviewer (SA) prior to the interview in all cases. The interviewer and participants were alone during interview in all but three instances, where a parent observed the interview. Interviews were audio and video recorded, and field notes were collected following the interview. Audio recordings were transcribed verbatim by an official transcription service. Interview transcripts were sent to participants to check accuracy with an opportunity for participants to add any further details if required. No revisions or amendments were received. Demographic and clinical data were collected electronically prior to the interviews.

5.3.9 Patient and public involvement (PPI)

This study was conceived as a direct consequence of gaps identified in a previous systematic review of physical functioning outcome measures amongst participants with AIS (Alamrani et al., 2021a). The PPI representative was part of the study management group, and her feedback had been sought on the study protocol, the topic guide as well as versions of the participant information sheet and consent forms. Further, the PPI representative was involved in data analysis and interpretation of study findings through provision of a plain English summary.

5.3.10 Data storage and management

All study investigators complied with requirements of the Data Protection Act 2018 with regards to the collection, storage, processing, and disclosure of personal information and upheld the Act's core principles. Personal data were coded and depersonalised and replaced with a participant identification number. and stored electronically on a password-protected computer at the University of Birmingham. Secure maintenance of the data ensured that the linking code were kept securely in a separate location using encrypted digital files within password- protected folders and storage media. Only the Chief Investigator and the study Research Fellow had access to the data as necessary for quality control, audit, and analysis. Data will be stored for 10 years in line with the University of Birmingham's Research Governance procedures only accessible for the research team.

5.3.11 Data analysis

When evaluating the content validity of an existing PROM, it is advisable to follow the same steps employed in the development of PROM (Rothman et al., 2009). A deductive thematic analysis was used to analyse the data into themes, which were predetermined by the topic guide or emerged from the data related to the concept (Brod et al., 2009). The process started by familiarization with the data, by reading and rereading it several times. Then, generation of initial codes by identifying and labelling interesting features or patterns was performed. Coding was performed by (SA) with guidance from the co-authors. Then, codes were grouped into potential themes based on similarities or pattern, followed by reviewing and naming themes. At the last step, a report is produced with complete themes, subthemes and participant quotes which are tabulated (A*ppendix 9*) to allow easy visualisation and interpretation of results (Terry et al., 2017). To ensure that the results were representative of the HRQOL among different participant subgroups (Rothman et al., 2009), the

coding was stratified according to participant characteristics i.e., curve severity and management approach (Rothman et al., 2009). Finally, themes that emerged were mapped to the SRS-22r contents (domain and items) (Rothman et al., 2009). The words and phrases in the derived themes and subthemes were compared to words used in the SRS-22r questionnaire (Rothman et al., 2009). If a poor agreement was evident this then shows inadequate coverage of the concept of interest. Adaption to the PROM, or the development of a new PROM, should then be undertaken to support its use (Rothman et al., 2009).

5.3.12 Trustworthiness

Quality was ensured by employing many strategies to enhance trustworthiness of the study results (Mays and Pope, 2000). The study was reported following COREQ guidelines (Tong et al., 2007). Furthermore, strategies to enhance rigor was followed including a) triangulation by comparing themes and subthemes identified with existing knowledge, b) peer debriefings as coding and analysis were assessed and discussed by co-authors, c) relevance in which the data obtained from studies are add to the previous knowledge, as well as d) attention to negative cases in which (SA) present a case and was discussed with (NH). All of these steps are in line with subtle realism world view (Mays and Pope, 2000).

5.4 Results

5.4.1 Participants

Demographic and clinical characteristics of participants are presented in Table 5.1. From 19 recruited and consented participants, 11 took part in the study. Four participants could not be contacted because they did not respond to the invitation from the researcher. Two withdrew due to school commitments and one withdrew because of parental withdrawal of consent. One participant withdrew once the interview had commenced as they denied speaking and participating because of feeling shyness. Therefore, their demographic data were discarded. The sample was representative of the population of adolescents with AIS, with the majority being female (n=8), aged between 10-18 years old.

Table 5.1: Demographic and clinical characteristics of participants

ID	Age (years)	Gender	Curve severity	Location of curve	Lenke type	Management	Duration Following surgery	Number of X-rays	Pain intensity (0-10)	Duration of pain	Physical activity level per day	SRS-22r score Mean (SD)
1	15	Female	51°	Thoracic	1	Observation	NA	2	5	> 6 mos.	< 60 min.	3.2 (.89)
2	17	Female	32°*	Thoracic	3	Post-surgery	20 mos.	12	5	> 6 mos.	> 60 min.	3.7 (.72)
3	12	Female	31°	Thoracolumbar junction	5	Brace	NA	5	3	> 6 mos.	> 60 min.	4.4 (.46)
4	12	Female	25 °	Thoracolumbar junction	5	Observation	NA	1	0	-	> 60 min.	4.6 (.81)
5	16	Female	40 °*	Thoracic	1	Post-surgery	10 mos.	8	5	> 6 mos.	60 min.	4 (.76)
6	17	Male	57 °	Thoracic	1	Pre-surgery	NA	6	4	> 6 mos.	< 60 min.	3.3 (.71)
7	16	Female	70 °	Thoracic	1	Pre-surgery	NA	2	8	> 6 mos.	< 60 min.	3.7 (.52)
8	16	Female	70 °	Thoracolumbar junction	1	Post-surgery	3 mos.	11	4	> 6 mos.	< 60 min.	3.4 (.68)
9	14	Male	40 °	Thoracic	1	Brace	NA	2	3	3 - 6 mos.	< 60 min.	4.1 (.68)
10	13	Female	86 °	Thoracic	1	Pre-surgery	NA	3	7	> 6 mos.	> 60 min.	2.8 (.66)
11	16	Male	40 °	Thoracic	1	Pre-surgery	NA	2	7	> 6 mos.	60 min.	3 (.52)

5.4.2 Themes and subthemes

A slight modification was made to the hypothesised conceptual framework (Fig 5.1), when compared to the updated conceptual framework (Fig 5.2) that was developed from the data. The identified themes were: (1) physical effects, (2) activity-related effects, (3) psychological effects, and (4) social effects. Some of the hypothesised subthemes were discarded as no data were collected to support their inclusion. Each theme and subthemes with appropriate code (participants quotes) are presented in the (*see Appendix* 9).

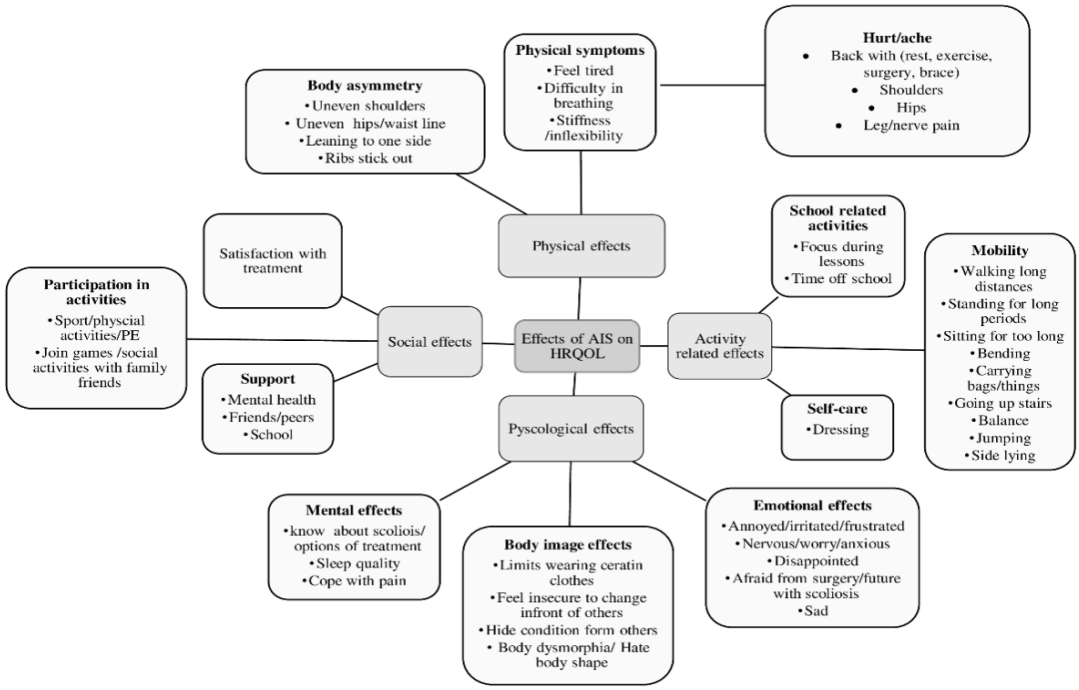


Figure 5.2:Updated conceptual framework

5.4.3 Physical effects

This theme was consistent with hypothesised themes across the two conceptual frameworks. In addition to various new subthemes were identified.

5.4.3.1 Physical symptoms

Back hurt was the main physical symptom reported by study participants. Those with severe curves expressed high pain intensity compared to participants with milder curves. Pain was associated with exercise performance such as stretching exercises as a form of sport (e.g., yoga), or in a form of treatment (e.g., physiotherapy). Participants noted that exercise was the leading cause of pain and the need for subsequent analgesia, which affected their participation in sport or physical education in school.

> *"Sometimes it hurts my back when I do some exercises.. ، when I play football, it hurts my back"* (P11-pre-surgery)

Management of AIS (e.g., surgery) contributed also to the feel of pain. It affected school attendance, performance and the need for analgesia for several months following surgery. Bracing also can contribute to the experience of pain, and it was described as uncomfortable, and causes bruises.

> *"Whereas now if I do get pain, it's because of the surgery" (*P8-post surgery)

Participants also reported shoulder and hip pain, which was associated with body asymmetry. Shoulder and hip asymmetry were thought to affect being able to find a comfortable sleeping position and having a good quality of sleep.

"I think just my shoulders and my hips sometimes because it's kind of like disjointed everything. They ache most of the time and no matter how many medications I can take for it, it will always be there because it just reoccurs" (P1-observation)

Nerve or leg pain was also reported by those with severe curves and waiting surgery, contributed to the feeling of weak legs and the increased need of taking pain medications.

*"When I get nerve pain, I have to take three [painkillers] to kill it" (*P10-Pre-surgery).

Participant reported feeling more tired when comparing themselves to their peers at school, and they felt that they needed to take more breaks in the day. Difficulty in breathing was also reported especially with running, and among those with severe curves.

"I definitely feel more tired than other people after I do a lot of things and I have to take more breaks than most of my friends when they do the same things" (P1-Observation).

Our study participants have not reported any limitation in range of movements; however, the terms "stiffened", and 'inflexibility' were used by some.

"I'm less flexible; I've never really been that flexible. I used to be really; I could do anything. But now it feels kind of stiffened and I've definitely lost quite a lot of flexibility" (P1-observation)

5.4.3.2 Body asymmetry

Participants did not see or report a back hump that reflects the asymmetry of the posterior torso and is a physical sign of scoliosis, whilst a parent had noticed it. Participants noted different shoulder and hip levels, and with severe curves they noticed the ribs being more prominent. They also reported feelings of leaning over to one side.

"My hips are about five centimetres apart so like one's there, one's there…" (P10-presurgery)

5.4.4 Activity-related effects

Three main themes were identified related to the effects of scoliosis on activity (mobility, self-care, school related activities). Compared with the hypothesised framework, multiple subthemes emerged from collected data for mobility but not for self-care.

5.4.4.1 Mobility

Sitting for too long was the frequently reported mobility problem by participants. They described that they cannot sit for a long time without feeling back pain and discomfort. This often led to a change in their position, either by standing or moving around. This was problematic in school, especially with lessons of a long duration without breaks, or where chairs had no back support. Further, "standing for long time" and "walking for long distances" were also reported as the cause of back pain. Some participants avoided joining activities with friends and family that require walking a long distance.

> *"Sitting down because I have two-hour lessons, instead of one hour. And sitting down for two hours is uncomfortable"...*" *Sometimes, I'm allowed a break like 10 minutes in between the lesson. But sometimes, I don't, and it can get kind of painful"* (P6-Presurgery)

Bending activities (i.e., picking up things from floor or vacuuming) was described as painful and challenging activity. Those who had surgery or wore brace found that it was difficult to tie their shoelaces or put socks on. Avoidance behaviour, asking for help or finding alternative ways to perform the activity was reported.

> *"When I keep having to bend down and pick things up, it's quite hard to sometimes clean my room and especially vacuum….and it hurts quite a lot at the end"* (P1-Observation)

> *"Bending over, that kind of thing; I tend to avoid bending over" (P2-post surgery).*

Activities such as jumping and running at a pace that is more than a jog, were reported as difficult, and would be avoided.

> *"The first time I realised that something was quite painful about it was when I was out with friends at a trampolining place. I started to jump, .this was the only time that it actually started to hurt. And I had to stop"* (P1-Observation)

Participants found that "carrying things" such as school bags could increase of back discomfort. Other reported that scoliosis "hindered their balance", which make them feel dizzy and unable to walk in a straight line. Some adolescents reported that they were unable to go upstairs, and they have been allowed to use the lift instated of stairs at school. Side lying and difficulties finding a comfortable position have an impact on sleep quality as well.

> *"Walking in straight... I can't walk in like... well, I feel like I'm not walking in a straight line"* (P7- Pre-surgery)

> *"Sometimes I do think it hinders my balance as well, so maybe that is why I can't stand up for very long periods of time without feeling dizzy."* (P1- Observation)

5.4.4.2 Self-care

Dressing was the only challenging activity identified in this theme, causing discomfort for some participants. Activities such as washing oneself was discarded as no data supported its inclusion. Those who had undergone surgery reported needing help in self-care activities in early post-surgery period, but not after several months following surgery. Dressing also included tying shoes laces and putting socks on, which remained challenging following surgery.

> *"Dressing, putting my socks on can be a bit of a task, or pulling up my trousers, but it's not that hard that it is a problem"* (P8-Post-surgery)

5.4.4.3 School-related activities

Participants reported that their focus and concentration during lesson time was impacted because of pain they felt with prolonged sitting. Thinking about pain and changing sitting position to find a comfortable spot affected concentration. Also, wearing a brace increased their discomfort and further affected their ability to focus.

> *"Sometimes it's definitely difficult to focus when my back is hurting that much and I'm trying to get on with the lesson. It is worse in science because we have these chairs, they don't have a back on and so I have nothing to lean against. It makes me struggle because I'm trying to focus, but then trying to make sure that my back is not bending or hunching over because it would hurt more"* (P1-Observation)

Participants reported that pain, surgery, and the follow-up appointments affected school attendance and performance. Surgery alone led up to 5 months away from school for one participant. For another participant she decided not to carry on schooling, because she had missed a lot of school days due to surgery.

> *"It did affect my GCSEs because I literally had it done right before my exams. I had two months off school, ...so I obviously missed a lot of school ...now I still have follow-up appointments, obviously you miss time off school... so, it does add up"* (P5-post-surgery).

5.4.5 Psychological effects

Three main subthemes were identified related to this theme. Hypothesized factors such as self-esteem and positive feelings has been discarded, as no data supported its inclusion.

5.4.5.1 Emotional effects

Many terminologies were used interchangeably by participants with AIS to describe their negative feelings and emotions about scoliosis and it associated treatment (Figure 5.2). While no positive feelings were elicited. Participants used terms like "sad" when they felt pain, when they couldn't explain their feelings to others, or when they think about surgery.

> *"It makes me sad because it reminds me of being in pain a lot before the surgery, because the biggest thing I wanted out of the surgery was to help my pain, even if it was just a little bit".* (P8-post-surgery)

Feelings nervous, worried, and anxious were used when they compared themselves with others, or when thinking about the need of surgery and how it will affect their future life. They felt worried also because their parents felt worried too.

> *"I feel a bit worried, like how it will affect me in the future and how it's going to impact on my life. Just a bit nervous about if it will get worse in the future or not"* (P11-pre-surgery)

Participants reported feelings of being "annoyed" and, "irritated" because of having scoliosis or because they still have back problems following surgery. Wearing a brace made them feel annoyed as well. Feeling "afraid" was used when describing their emotions to have surgery, or that they might need surgery in the future. Disappointed was expressed when they believed there was nothing that could be done to treat their condition.

> *"It can make me feel very annoyed, like it's just a bit like why me? Why?"* (P10-presurgey)

> "*I feel quite disappointed that there can't be anything done. And I understand why there cannot be anything done. Yeah... just disappointment is the biggest one"* (P1-Observation)

5.4.5.2 Body image effects

This theme has a significant impact on participants lives and many subthemes were found. Participants reported feeling insecure about their body shape, making them unable to change when being with their friends or peers. They tend to hide their back from others by wearing large clothes. They felt that scoliosis limits wearing certain clothes such as a swimming kit, which limits participation in swimming. The term "dysmorphia" was used by one participant describing how she feels about her body shape.

> *"I don't know if this is the right thing, but it's getting changed in front of people, I can't do that and that's the issue usually also in PE, … Or it's when I'm with friends and we're all getting ready for going out …. I just cannot get changed in front of them" (*P1-Observation).

This was also associated with preference of some of participants with AIS, to hide their condition from others.

> *"I didn't tell anyone about it because I didn't like it ... I distanced myself from people.. I just wanted to be on my own. But it is fine now" (P5-Postsurgey).*

5.4.5.3 Mental effects

Participants described different methods to cope with pain, including taking analgesia, changing positions, and taking rest when pain was associated with activity.

> *"It's usually just a case of I just have to lie down for a minute. Obviously when I am resting it does not hurt, that's literally the way to quickly cure it, is lie down" (*P5-post-surgery).

Participants reported that they felt shocked and confused, because of the lack of knowledge about scoliosis and its treatment. They reported that they felt anxious and worried which affect quality of sleep.

> *"I kind of had a stage of denial, I didn't believe that it was real, and I felt that they were wrong. But then I saw the x-rays and I was shocked, to be honest. I did not realise what it actually looked like and what it was meant to look like basically"* (P1-Observation).

5.4.6 Social effects

5.4.6.1 Participation

Sport and exercise participation was limited or discontinued due to back discomfort. Likewise, following surgery, some people were recommended to avoid contact sports, therefore their involvement in sports was limited.

> *"I've always been a big sporty person, which is why it impacted me so much having scoliosis. I went from being very competitive and fast and loved sports to not being able to participate"* (P8-Posturgery).

Scoliosis also has an influence on "recreation and leisure" activities with family and friends. Going out with family and friends was difficult because of the severe pain.

> " *Sometimes. When they were going trampolining, and I couldn't't go because it hurt that much. Or when it is a birthday party and everybody is doing piggybacks and I have to say, hmm, maybe that's not a good idea"* (P1-Observation).

5.4.6.2 Support

Friendship support was found to be important to participants and would assist in mental health, particularly during the recovery time following surgery.

> *"My school has been amazing.. has provided a chair with lumbar support. It's a very comfortable chair for me to sit in in exams, so I don't have to sit for two hours to do a mock on an uncomfortable chair".* (P8-post-surgery).

Mental health support" has been shown to be necessary since scoliosis has various effects on the psychological part of AIS life.

> *"I think, definitely, the treatment you get post-op; you don't get any mental support, and I think there should be more of it"* (P2-post-surgery)

5.4.6.3 Satisfaction

Treatment satisfaction was essential to study participants. They felt that having observation without intervention was frustrating, and that performing exercises was not an effective treatment. Some were dissatisfied when surgery was the only possible option associated with long waiting periods.

> "*So, I literally got diagnosed, they were like, "You've got this," and then they were like, "We can't help you with anything and you've just got to wait for surgery." I've basically been given nothing, absolutely nothing"* (P10-Pre-surgery).

5.4.7 Stratification analysis

Table 5.2 shows the stratification of data based on the participant's management approach (i.e., observation, brace, pre-/ post-surgery). Although, some participants do not express some codes it could be returned to individual variation in experience and personality. Overall, the majority of codes are expressed by participants across the three-management approach.

Table 5.2 :Stratification of subthemes according to participants management approach

Subthemes	Observation	Brace	Pre-surgery	Post-surgery
1a. Back hurt at rest	✓			✓
1b. Back hurt with brace		✓		
1c. Back hurt when exercising	✓	✓	✓	✓
1d. Back hurt because of surgery				✓
Hips hurt	✓	✓	✓	
leg ache/nerve pain			✓	
Shoulder aches	✓		✓	
Feel stiffened/inflexible	✓		✓	✓
Difficult breathing			✓	
Feel tired	✓		✓	
Uneven Shoulders	✓	✓	✓	✓
Uneven hips/waist	✓	✓	✓	
Leaning to one side			✓	
Ribs stick out			✓	
Time off school		✓	✓	✓
Focus during lessons	✓	✓	✓	
Ache with dressing	✓	✓	✓	✓
Jumping/jog	✓		✓	

Hinder balance/walking straight	✓		✓	
Bending	✓	✓	✓	✓
Carrying bags/things	✓		✓	
Going up stairs			✓	
Walking long distances			✓	
Sitting for long time			✓	✓
Side lying	✓	✓	✓	
Stand for too long	✓		✓	✓
Annoyed-irritated-frustrated	✓	✓	✓	✓
Sad	✓		✓	✓
Disappointed	✓			
Nervous-worry-anxious	✓		✓	✓
Afraid	✓		✓	
Feel insecure to change in front of others	✓		✓	
Hate body shape/dysmorphia			✓	
Limit wearing certain clothes			✓	
Hide back condition			✓	
Pain coping	✓	✓	✓	✓
Know about the scoliosis	✓	✓	✓	✓
Know about options of treatment	✓	✓	✓	✓
Sleep quality			✓	✓
School support		✓	✓	✓
Friends support	✓		✓	
Mental health support		✓	✓	✓
Participation In sport/physical activities/physical education	✓	✓	✓	✓
Participation in games /social activities with family friends	✓		✓	
Satisfaction about given treatment	✓	✓	✓	✓

5.4.8 Comparison analysis

Table 5.3 presents the comparison and association performed between the codes elicited from the interviews and the items of the SRS-22r. Only one question from the SRS-22r was linked to the physical effects codes. Whereas other physical symptoms and body asymmetry were not linked to the any of the SSRS-22r items. Similarly, for activity effects only one question from the SRS-22r was linked to the codes, while three questions were linked to psychological effects cods and two questions to the body image codes. Participation was also linked to the SRS-22r and the satisfaction theme. Only 10 questions from the SRS-22r were connected to the codes raised from the qualitative data. This may indicate that the SRS-22r may not accurately reflect the perspectives of those with AIS.

Table 5.3 : Comparison between SRS-22r contents with themes, and subthemes elicited from interviews

Qualitative data		**SRS-22r**
Themes	**Subthemes**	**Questions**
Physical effects	1a. Back hurt at rest	8- Do you experience back pain when at rest?
	1b. Back hurt with brace	
	1c. Back hurt when exercising	
	1d. Back hurt because of surgery	
Physical Symptoms	Hips hurt	
	leg ache/nerve pain	
	Shoulder aches	
	Feel stiffened/inflexible	
	Difficult breathing	
	Feel tired	
Body asymmetry	Uneven Shoulders	
	Uneven hips/waist	
	Leaning to one side	
	Rips stick out	
Activity-related effects School-related activities	Time off school	17-In the last 3 months have you taken any days off work, including household work, or school because of back pain?
	Focus during lessons	
Self-care	Ache with dressing	
Mobility	Jumping/jog	
	Hinder balance/walking straight	
	Bending	
	Carrying bags/things	
	Going up stairs	
	Walking long distances	
	Sitting for long time	

	Side lying	
	Stand for too long	
Psychological-related effects Emotional effects	Annoyed-irritated- frustrated	
	Sad	7-In the past 6 months have you felt so down in the dumps that nothing could cheer you up?
	Disappointed	16-In the past 6 months have you felt down hearted and blue?
	Nervous-worry-anxious	3-During the past 6 months have you been a very nervous person?
	Afraid from surgery/future with scoliosis	
Body image	Feel insecure to change in front of others	
	Hate body shape/dysmorphia	4- If you had to spend the rest of your life with your back shape as it is right now, how would you feel about it? 19. Do you feel attractive with your current back condition?
	Limit wearing certain clothes	
	Hide back condition	
Mental effects	Coping with pain	
	Know about scoliosis	
	Know about other treatment options	
	Sleep quality	
Social effects Support	Friends' support	
	School support	
	Mental health support	
Participation	Sport/physical activities/physical education	
	Join games /social activities with family friends	18-Does your back condition limit your going out with friends/family?
Satisfaction	Satisfaction about given treatment	21-Are you satisfied with the results of your back management? 22-Would you have the same management again if you had the same condition?

5.5 Discussion

This is the first qualitative study that has used in depth semi-structured interviews to evaluate the impact of scoliosis on the HRQOL of adolescents with AIS. The study was designed to evaluate the content validity of a currently used PROM (i.e., the SRS-22r) through a concept elicitation format, focusing on the adolescent experience with scoliosis and its associated treatment. Previous research studies have used quantitative methods to evaluate the validity of the SRS-22r with no attention being given to qualitative approaches. This study used words and phrases from participants with AIS in the generation of concepts, to ensure that the data are a true reflection of participants' perspectives. When the qualitative data from participants with AIS was compared to the contents of the SRS-22r, a poor match was found indicating a lack of content validity of the SRS-22r. Furthermore, the terminology used in the SRS-22r does not reflect the language used by participants with AIS. Findings from this study provide robust evidence for the need to revise the SRS-22r, or development of a new PROM using data similar to that generated from this study that represents the voice of adolescents with AIS and is language and age relevant.

5.5.1 Physical effects

Back hurt/pain was the most reported physical effect of scoliosis on HRQOL of participants, consistent with all previous reviews, where the prevalence of back pain amongst adolescents with AIS ranges between 37% to 42% (Théroux et al., 2017). Performing exercise improves HRQOL of AIS, by improving pain and function (Thompson et al., 2019, Monticone et al., 2014). However, most participants experienced back pain when exercising, which affected their adherence and participation in sport and PE at school. Consistent with previous studies, surgery contributed to back pain which may extend to two years post-surgery (Bastrom et al., 2013, Perry-Eaddy, 2018). This has

been identified as the major concern in the presurgical period (Motyer et al., 2021a, Motyer et al., 2022). The adolescents in this study used terms such as “hurt” or “ache” rather than “pain”, to describe their pain experience. This is not reflected in the terminology used by the SRS-22r (Asher et al., 2006), which may not be representative of this category of population. An important aspect of content validity is that the PROM should be age and language relevant (Matza et al., 2013, Haynes et al., 1995). Other symptoms were also reported such as difficulty in breathing, stiffness, hip, and shoulder pain in accordance with earlier reports (Motyer et al., 2022, Weinstein et al., 2008, Altaf et al., 2013). However, these symptoms are not assessed by the SRS-22r. Body asymmetry was also reported by participants as a major concern consistent with previous studies, where body asymmetry associated with scoliosis had a social and psychological impact on life (Aroeira et al., 2016). This aspect is not assessed by the SRS-22r. These findings reflect the strength of this study in using a concept elicitation interview to capture participant’s own perceptions of their condition to inform content validity (Patrick et al., 2011a).

5.5.2 Activity-related effects

Carrying out normal daily activities such as sitting, standing, and walking, if performed for a long time, was reported to aggravate symptoms. These activities were reported in a previous qualitative study in a presurgical period (Motyer et al., 2022). Scoliosis also affects standing stability and balance (Nault et al., 2002). Walking in a straight line was altered because of scoliosis, which has been assessed in earlier studies (Yang et al., 2013). Bending was limited generally, and for those who had a surgery in particular, since spinal mobility and flexibility is reduced following surgery (Danielsson et al., 2006, Nokariya et al., 2022). Participants reported discomfort when carrying backpacks, which has been explained by the increase in the compressive forces on the curve apex (Schmid et al., 2020), that may cause additional strains on muscles around the spine. Jumping was

avoided as they realised that it caused back pain (Motyer et al., 2022). Going up stairs was also reported as difficult, although it is not reflected in any scoliosis literature. The association between performing functional tasks such as climbing stairs and having back pain has been previously noted in other groups (Lima et al., 2018). Participants described that their focus during lesson time was affected because of a feeling of back hurt during sitting for prolonged time. A study on the association between pain and functioning in school found that adolescents with pain were more likely to have low school engagement, attendance and performance (Groenewald et al., 2020). Participants also reported that they missed school days because of feelings of pain, surgery or hospital appointments which affected their academic achievements. In some countries, surgery is performed during summer time to reduce interruptions of school performance (LaMontagne et al., 2004). However, for some people with AIS, time off school may extend to 6 months (Danielsson et al., 2001, Toye et al., 2016). Regarding self-care, participants reported that dressing caused aching and was a challenging. In summary, these findings demonstrated the significance of these aspects to HRQOL of adolescent with AIS, and therefore should be reflected in a PROM that evaluates HRQOL of this population.

5.5.3 Psychological effects

The emerged themes revealed that scoliosis has a significant impact on the psychological aspect of individuals with AIS life. Different negative feelings were reported associated with both diagnosis and treatment. Feelings of frustration, irritation, and annoyance as well sadness, when experiencing pain were described by participants (Toye et al., 2016). Disappointment was also discussed, relating to the treatment decisions, as mentioned in a recent review (Essex et al., 2022). Worry and fear about the need for surgery and a future living with scoliosis contributed to feelings of nervousness and anxiety, as discussed before (Rullander et al., 2017, Essex et al., 2022, Motyer et al., 2022), and is also reported by the parents of adolescents with AIS in other studies (Donnelly et al.,

2004, Motyer et al., 2021a, Bull and Grogan, 2010). Different questions in the SRS-22r evaluated mental health of adolescents, however, they are driven from the SF-36 which is an adult rather than a paediatric questionnaire (Alamrani et al., 2021a).

The effects of scoliosis on body image of adolescents with AIS is another key factor found in this study. Participants disclosed feeling insecure to change in front of others and hid their back. Scoliosis also limited their ability to wear certain clothes (Essex et al., 2022, Motyer et al., 2022). Body dysmorphia and hate of body shape were raised by one participant with a severe curve. This was associated with dissatisfaction about body image (Bertuccelli et al., 2022). The self-image domain in the SRS-22r assesses the effect of AIS on body image (Asher et al., 2006). Only two questions from this domain seems relevant to the driven subthemes.

A pain coping subtheme was revealed by our participants. A prior study found that coping with pain is dependent on the personality of the individual and affects the recovery process (LaMontagne et al., 2004). Since the HRQOL of adolescents with AIS may be related more to psychosocial effects than to physical effects and its consequences (Gallant et al., 2018), it is recommended to train clinicians who are caring of individuals with AIS, in stress reducing techniques including pain coping strategies (LaMontagne et al., 2004, Rullander et al., 2016). Information about scoliosis, and the options for treatment and associated consequences, has a psychological impact on participants life. This has been studied among adolescents with AIS and their parents prior to surgery (Motyer et al., 2021a, Motyer et al., 2022), wherein providing support and information to adolescents and their families helps in reducing stress and anxiety associated with scoliosis (Essex et al., 2022, LaMontagne et al., 2004).

Participants also reported that their sleep quality was affected by scoliosis. A recent study assessed the sleep profile of adolescents with AIS and revealed poor sleep quality associated with

high pain intensity (Yakut et al., 2022). Furthermore, following surgery reports indicated anxiety, nightmares and sleeping difficulties, both after the hospital visit and for a long time after the recovery period (Rullander et al., 2013). These results highlight the significant impact of scoliosis on the psychological aspects of life with AIS and therefore, should be included in a PROM evaluate their HRQOL.

5.5.4 Social effects

Participation in activities of daily life is important for the physical and psychological development of adolescents (Huus et al., 2021). In this study, participants reported reduced participation in social and sport activities consistent with previous studies (Kakar et al., 2017). Fear of pain or injury, and reduced self-image, may limit social participation, and has a negative impact on wellbeing (Rullander et al., 2013). Spinal fusion surgery has been shown to have a long term effect on an individuals' social life, and that providing support in the form of information and coping techniques increased the level of social participation (LaMontagne et al., 2004). In our study, participants reported the need for mental health support to overcome the psychological consequences of scoliosis and surgery. Earlier studies recommended providing psychological support to adolescents with AIS and their parents to minimize stress and uncertainty about surgery (Bull and Grogan, 2010, Rullander et al., 2013). Satisfaction about the treatment, assessed based on the answers of the SRS-22r was relevant to our study participants. Two questions in the SRS-22r assess satisfaction and were linked to the identified themes.

5.6 Strengths and limitations

This study is reported in line with published guidance of qualitative research (COREQ) (Tong et al., 2007), and is based on a published protocol (Alamrani et al., 2021a) to ensure rigor and comprehensiveness of the findings. Findings of this study using concept elicitation format allow capturing participant's own perceptions of their condition to inform content validity (Patrick et al., 2011a). Furthermore, patient perspective was central in this study, through involvement of PPI representative (ER) in the study design and interpretation of the results.

Limitations are in a number of areas with several factors that have influenced data collection and analysis, and thus may influence the findings reported. The sample was recruited from a single hospital in the UK limiting transferability of the findings. However, we assured diversity in population characteristics using a heterogenous purposive sampling technique (Etikan, 2016), which ensures the recruitment of participants with various demographic, clinical characteristics, and treatment approaches. The children/adolescents were not talkative, tended to give short answers and were reluctant despite prompting to elaborate on their responses and give further details. The assessment of concept saturation was performed following data collection, although, it has been recommended to be evaluated during data collection (Kerr et al., 2010). Coding of data was performed by one researcher (SA) and evaluated later by the research team. However, the coding framework and the codebook provide an evidence that coding is a true reflection of participants quotes (Patrick et al., 2011a). Ideally when designing a PROM, a theoretical model is developed based on the analysis of the data (Brod et al., 2009). However, in this study, a conceptual framework was used and modified based on the data analysis. These results still require further investigation and assessment, for example in another sample from another specialist centre in the UK, to ensure comprehensive understanding of the AIS and how its impacts the HRQOL of adolescent. Furthermore, qualitative

research is needed to assess the content validity of the developed PROM through conducting cognitive testing, to test its relevance, comprehension, and comprehensiveness from both adolescent and practitioner perspectives (Terwee et al., 2018a).

5.7 Chapter Summary

In this chapter the experience of individuals with AIS was explored through concept elicitation interviews. Eleven adolescents with AIS were purposively recruited with different age categories, curve severity and management approaches. Interviews which last approximately 45 minutes were recorded, transcribed, and analysed. The data revealed that scoliosis and its associated treatment has a broad impact on life, including physical, activity-related, psychological, and social effects. Evaluation of content validity of SRS-22r by comparing between the concepts and codes elicited from interview data, and the SRS-22r contents, revealed that the SRS-22r does not accurately reflect the words and phrases of adolescents with AIS, indicating a lack of its content validity. Themes and subthemes that have resulted from this qualitative study could be used to develop a new PROM for AIS or update the existing SRS-22r to enhance its use for this population.

Chapter Six

OUTCOME MEASURES FOR INDIVIDUALS WITH ADOLESCENT IDIOPATHIC SCOLIOSIS: A QUALITATIVE EXPLORATION OF HEALTHCARE PROFESSIONALS' PERCEPTIONS AND PRACTICES

This chapter reports in full the contents of a published manuscript by the thesis author protocol (Alamrani et al., 2021a), and research article (Alamrani, et al., 2023). It includes verbatim text from the published manuscript and some changes in the introduction section were employed for the purpose of this thesis.

Publication and Presentation

- **Alamrani,** S., Gardner, A., Falla, D., Russell, E., Rushton, A.B. and Heneghan, N.R., 2021. Content validity of Scoliosis Research Society questionnaire-22 revised (SRS-22r) for adolescents with idiopathic scoliosis: protocol for a qualitative study exploring patient's and practitioner's perspectives. *BMJ open*, *11*(12), p.e053911. (**Appendix 3**)
- **Alamrani S.**, Gardner A, Falla D. Russell E, Rushton AB, Heneghan NR. The use of outcome measures by healthcare professionals for young people with adolescent idiopathic scoliosis: a qualitative exploration. (Under review).
- **Alamrani S.**, Gardner A, Falla D. Russell E, Rushton AB, Heneghan NR. The use of outcome measures by healthcare professionals for young people with adolescent idiopathic scoliosis: a qualitative exploration. World Physiotherapy Congress. 2-4th June 2023. (Poster presentation).

6.1 Abstract

The results from *Chapter Two* and *Chapter Three* shows that the use of PBOMs among individuals with AIS is limited. The current knowledge around the current use of PROMs and PBOMs for AIS, as well as perceived barriers and facilitators of HCPs towards their use is limited. This *Chapter* objectives were 1) to explore current practice of HCPs when assessing outcomes for AIS, 2) to understand perceived barriers and facilitators of HCPs to use PROMs 3) to understand perceived barriers and facilitators of HCPs to use PBOMs.

A qualitative study recruited a purposive sample of HCPs from a tertiary hospital in the United Kingdom. Mean years of experience managing individuals with AIS was 11.8 years; and included surgeons, physiotherapists, and nurses, educated at Bachelor, Master, and Doctoral level. Consent to participate and demographic information were collected in advance of the interviews. In-depth, virtual semi-structured interviews were informed by a topic guide based on current evidence. Interviews of approximately 45 minutes were audio and video recorded and transcribed verbatim alongside written field notes. Data were coded and analysed using inductive thematic analysis, involving researchers with topic and methodological expertise and input from a patient representative.

Two themes emerged regarding current practice of using PROMs in routine practice and personal evaluations. Four themes emerged as barriers to using PROMs for individuals with AIS: priority and support (e.g., HCPs focus on providing care), practical challenges (e.g., inadequate PROMs), patient-related challenges (e.g., patient preferences), and the knowledge, education, and perceived value. Two themes emerged as facilitators: quality existing measure (e.g., sufficient psychometric properties), and priority and support (e.g., research department/culture). Themes for

barriers to use performance measures were practicality (e.g., need physical space) and perceived value and knowledge (e.g., PROMs are more important), while the one theme for facilitators was practical consideration (e.g., acceptability)

Although HCPs perceived the value of using OMs, current practice indicate limited use for individuals with AIS. The findings revealed different barriers and facilitators to implement PROMs in practice. Adoption of PBOMs is limited by lack of knowledge and perceived value and practicality, while considering practical factors can improve the use of these measures in practice.

6.2 Introduction

Findings from *Chapter Three,* showed that most of the studies investigating measurement properties of OMs in AIS, are for PROMs, with few studies evaluating PBOM, limiting their use in practice. The review identified only the Timed up and go test, in which its construct validity was evaluated in individuals with AIS. A clinical measure, such as radiographs and muscle strength, give an indication about impairment of structure or function, but does not fully capture the functional limitations of an individual (Reiman and Manske, 2011). Although PROMs are useful tools for gathering information about a patient's health status and HRQOL from their own perspective, their responses may be influenced by their perception of change as well as other variables such as pain and psychological stress, which are frequently reported in this population (Leszczewska et al., 2012, Motyer et al., 2022). On the other hand, PBOMs such as walking speed or endurance testing afford a more comprehensive assessment of function (Reiman and Manske, 2011). Performance measures can be used to evaluate the impact of interventions such as surgery and exercise on specific functional domains such as mobility, strength, endurance, and balance, which provide a holistic understanding of patient's condition (Ferguson et al., 2005). Nevertheless, variables that could improve or limit the usefulness of such measurements in individuals with AIS remain unknown.

Various barriers to the use of OMs have been identified in other fields, such as clinician self-judgment, time constraints, lack of knowledge and lack of resources (Kieser et al., 2020). Facilitators have also been identified, including knowledge and training to use an OMs, availability of the OMs, and guidelines recommending its use (Demers et al., 2019, Amini et al., 2021, Ntsiea et al., 2022, Briggs et al., 2020). Current research mostly focuses on PROMs, with little exploration of the use of PBOMs or the impact of population characteristics such as age, gender, and curve severity on the use

of OMs in AIS. To date, no research has explored current practice of using OMs for individuals with AIS, or HCPs perceived barriers and facilitators of all forms of OMs.

This chapter addresses the objectives of this thesis as follows:

1- To explore the current practice of the HCPs when assessing outcomes for individuals with AIS.

2- To understand the perceived barriers and facilitators of HCPs to use PROMs.

3- To understand the perceived barriers and facilitators of HCPs to use PBOM.

6.3 Methods

6.3.1 Design

This is a qualitative study design which is a component of a larger study that evaluated the content validity of the SRS-22r from the perspective of both individuals with AIS and HCPs (Alamrani et al., 2021a). It has been reported in a single chapter in this thesis to provide more detail and analysis of the results as well as to enhance clarity and comprehension for the reader. This study is reported in line with the COREQ checklist (Tong et al., 2007) (*See Appendix 10*).

6.3.2 Qualitative approach and research paradigm

The world view of pragmatism was used in this study. This approach emphasises the importance of selecting methods based on their appropriateness for addressing specific research questions rather than adhering strictly to philosophical debates (Ritchie et al., 2014).

Since the objectives of this study was to explore and understand the current practices of HCPs when assessing outcomes for individuals with AIS. The descriptive phenomenology was chosen as methodology for this study, as it focuses on describing the universal essence of an experience as lived by individual (Willis et al., 2016). This approach allows for an in-depth exploration of the essence of their experiences and practices, providing a detailed and rich description of the phenomenon (Shorey and Ng, 2022) .

6.3.3 Research team and reflexivity

A female experienced musculoskeletal physiotherapist researcher and PhD student (SA) conducted the interviews, with support from the co-authors who have expertise in conducting spinal and qualitative research. Further, a spinal surgeon with experience working with AIS was involved along with a patient and public involvement (PPI) representative. No relationship was established with participants prior to the commencement of the interviews. Participants were informed about the professional background of the interviewer and that the study was part of a PhD thesis

6.3.4 Settings

Due to COVID-19 pandemic restrictions that were in place at the time of data collection, the semi-structured interviews were conducted virtually via Zoom. Zoom is considered as a highly

satisfactory software alternative to face-to-face interviews, because of its ease of use, cost-effectiveness, and security options (Archibald et al., 2019). The timing of interviews was selected based on participants preferences and interviews lasted a mean of 45 minutes (SD=9.7). Interviews were conducted between July and September 2022.

6.3.5 Ethical approval

Ethical approval to conduct the study was gained from the Health Research Authority and Health and Care Research Wales approval (REC reference: 21/WM/0076). All participants provided their consent to participate before conducting the study, (see *Appendix* 10).

6.3.6 Participants

Participants were recruited from a tertiary scoliosis centre in the UK and invited to participate in the study via email. The purposive sample of HCPs included physiotherapists, nurses, and surgeons, who were part of health care team for individuals with AIS. Participants were considered eligible if they had experience in managing AIS.

6.3.7 Data collection methods

To explore the current use of OMs among HCPs including their perceived barriers and facilitators, a single semi-structured interview guide (*See Appendix 10*) was developed by the research team (Ritchie et al., 2014). Pilot interviews were not conducted; however, a comprehensive understanding of the topic was derived from existing knowledge and an extensive literature review, providing a robust foundation for formulating the interview protocol. No pilot interviews were

undertaken; however, the development of topic guide was strengthened by an input from a PPI representative.

All participants were all unknown to the interviewer prior to the interview. Only the participant and the interviewer were present during interview. Field notes were assimilated following the interview. Interviews were audio and video recorded, then transcribed verbatim through official transcription services. Transcripts were emailed to participants to allow corrections to be made or to add any further details. Participants were given two weeks to make any alterations or suggest any changes. Demographic information of the participants, along with the consent forms were collected electronically prior to commencement of interviews.

6.3.8 Data analysis

The lead researcher (SA) listened to the transcripts and performed the initial coding, which was reviewed and discussed with qualitative experienced co-authors, to enhance the credibility of the study findings. To ensure that participants' experiences and perspectives are accurately reflected, allocation of codes and theme alignment with participants quotes were evaluated. A saturation table was created to ensure that concept saturation was achieved (see *Table 6.1*), defined as the point where two consecutive transcripts failed to elicit any new themes (Kerr et al., 2010). Inductive thematic analysis following the Braun and Clarke framework was used to analyse the data (Terry et al., 2017). This process started first with familiarisation with the data by reading and re-reading it several times. Second, generation of initial codes, third, searching for the themes that reflect the data by grouping similar codes. Fourth, reviewing themes to ensure they accurately reflect the data. Finally, a process of defining, and naming themes and production of the report (Terry et al., 2017).

Table 6.1**: Saturation table showing that theme saturation was achieved**

Themes	Subthemes	Interview number						
Objective 1: Current practice		1	2	3	4	5	6	7
Routine practice	Standard practice in hospital	✓						
	To monitor outcomes	✓						
Personal evaluation	Personal questions	✓						
	Use patients' goals and needs		✓					
	Rating of outcomes		✓					
Objective 2: Barriers and facilitators to use PROMs								
Barriers								
Priority and support	Difficult to implement		✓					
	Focus on providing care		✓					
	Not informing practice	✓			✓			
Practical challenges	No time							
	Inadequate PROMs		✓					
	Logistic provisions			✓				
Patient-related challenges	Potential invalidity				✓			
	Patient preferences	✓						
	No discharge assessment		✓					
Knowledge, education, and perceived value	Lack of knowledge			✓				
	Questioning importance			✓				
	Need of education			✓				
Facilitators								
Quality exiting measure	Sufficient psychometric properties			✓				
	Updated and modernised	✓						
	Personalised and relevant	✓						
	Comparable and widely acceptable					✓		
	Simple and easy			✓				
Priority and support	Research department	✓						
	Scoring system			✓				
	Show difference in practice			✓				
Objective 3 : Barriers and facilitators to use Performance measure								
Barriers								
Perceived value and knowledge	Patient reported measure more important		✓					
	Performance indicators differ across individuals				✓			
	Unaware about use performance measure				✓			
Practicality	Need physical space and time			✓				
Facilitators								
Practical consideration	Relevant				✓			
	Simplicity and acceptability			✓				
No. of new codes appearing in each interview		8	7	10	5	1	0	0
% Of total new codes (Total =34)		23	20.5	29.4	14.7	2.9	0	0

6.3.9 Trustworthiness

Many strategies have been employed to enhance trustworthiness of the study results (Nowell et al., 2017). Credibility of the study was determined by using prolonged engagement with participants, promoting a deeper understanding of their experiences (Nowell et al., 2017). Member checking was also employed to validate findings (Nowell et al., 2017), by asking participants to review and confirm the accuracy of the study findings The research team, consisting of researchers with both topic and methodological expertise, participated in data analysis enhancing the robustness of the interpretations (Nowell et al., 2017). The transferability of the study findings was ensured by presenting findings with a focus on providing a "thick description" of the context and participants' experiences (Nowell et al., 2017).

6.4 Results

6.4.1 Participants

Of the 13 participants invited, seven agreed to participate in this study. Participants included four females and 3 males, had a mean age of 42 years (SD=7.9). Professions of participants included surgeons (n=3), nurses (n=2) and physiotherapists (n=2) with a mean of 11.8 years (SD= 4.9) of experience working with individuals with AIS. Their highest qualification was a doctorate (n=1), followed by a master's degree (n=4) and bachelor's degree (n=2). Different themes and subthemes were identified from participants for each objective of the study.

6.5 Current practice

Two themes related to current practice of using OMs were identified from participants' descriptions: routine practice and personal evaluation with associated subthemes.

6.5.1 Routine practice

Standard practice in hospital. Participants described that the current use of OMs is standard routine practice within the hospital, as per the national guidance of the British Spine Registry. The PROM used for individuals with AIS is the Scoliosis Research Society-22 questionnaire. Individuals with AIS routinely complete the questionnaire prior to the appointment with the HCPs. It also sent to the individual/parent email address automatically at specific time points to be completed remotely, to monitor changes over time.

> *"Actually, we use them for all the patients that come through as the pre-operative assessment. But it's only for research purposes. I don't use that in the clinical setting. I don't look at the score and decide on whether they have an operation or not. It is only for research purposes"* (P6- surgeon)

To monitor individuals with AIS outcomes. Participants perceived a strong benefit from using PROMs in this population to help them identify patient problems, monitor long-term outcomes as well as giving a good scope and understanding of patient's feelings.

> *"If a patient, for example, is being monitored for a few years, then prior to proceeding to need surgery, we could have maybe three years of data and seeing how the results have deteriorated or maybe not deteriorated, but we would see more of… able to see a larger pattern"* (P1-Nurse).

6.5.2 Personal evaluation

Whilst none of the participants reported use of any OM, they reported use a range of personal assessment tools to evaluate patients' outcomes.

Personal questions. Participants reported using personal questions to assess patients' outcomes. At every visit to the clinic, patients were asked about their life with scoliosis, how they feel about it, and how much activity they can perform.

> *"Only through questions during the history, not through a questionnaire. So, we do evaluate that, but not through a questionnaire that gives a numerical score. We would ask questions and have a more qualitative answer to that"* (P4-Surgeon)

Use patient goal and need. Physiotherapists reported using patient goals and needs as outcome measures to evaluate how their outcomes are changing.

> *"Just with specific goals, really. So, we'd use patient-specific goals to identify what they want to achieve, what they're not able to do, and then use that just as a specific measure: measure as to how much they're improving".* (P5-Physiotherapist)

Rating of outcomes. Physiotherapists reported asking patients to rate their level of pain and function, as well as to rate the percentage of improvement with regards to the return to normal function or activity.

> *"We're not using any really. ... But really, I'm probably getting them to rate their pain scale on like a one to 10 maybe. I'm also getting them, ... to rate how much better they're feeling…So out of 100%".* (P2-Physiotherapist)

6.6 PROM's Barriers and Facilitators

Four themes emerged from interviews as barriers to use PROMs for individuals with AIS while two themes emerged as facilitators, with multiple subthemes also identified. Figure 6.1 represents the themes and subthemes identified as barriers and facilitators for using PROM for individuals with AIS.

6.6.1 Barriers

6.6.1.1 Priority and support

The perceived importance of using PROMs in practice, and the lack of support was recognised as a barrier for HCPs in adopting the PROMs in practice.

Focus on providing care. Participants prioritised providing care to their patients rather than evaluating their outcomes using standardised measures.

> *"I think your kind of focused on providing physio"* (P2-Physiotherapist)

> *"Of course, as a clinician, although it's nice to be involved in research, our primary focus is patient care"*(P4-Surgeon).

Not informing practice. Participants perceived that the use of PROMs is not informing practice for AIS compared with clinical measures, which are determinant of the patient's future. Participants stated that the use of PROMs is for research purposes only and scores obtained did not inform their practice or clinical decision making. Further, they think that a PROM that evaluates quality of life is more valuable for those with AIS than radiological measures.

"They need to take formal measurements of your growth plates and your degrees of curvature. ...to decide whether it's going to be conservatively managed or surgically managed. That really dictates their future". (P2-Physiotherapist)

Difficult to implement. Participants perceived that the use of PROMs is difficult in practice owing to the lack of support, such as research team assigned to collect PROMs from patient. This in turn impacts compliance.

"Outcome measures have come in and we have certainly tried to use them in certain things…. But it is very difficult to implement that, people's compliance with it, get them trained up on it" (P2-Physiotherapist)

6.6.1.2 Practical challenges

Participants discussed a variety of practical challenges that impact the adoption of PROMs in routine clinic practice.

No time. Participants discussed time constraints as the biggest challenge to use PROMs. Clinician-time as well as patient-time and busy clinics were reported.

"I think sometimes as well it's from an environmental perspective and a perspective of just time restraints, patient-wise and clinician-wise… "I'm sorry, I can't fill that in today because I've got to go, because I've got another healthcare appointment". I think it's both ways" (P1-Nurse)

Inadequate PROMs. Participants perceived that the qualities of the currently used PROMs are inappropriate, affecting use in practice. They reported that some PROMs are lengthy, its language is outdated, and irrelevant to the current young generation, which in turn affect patient's ability to fill questionnaire.

"I mean it is quite a lengthy questionnaire. It is a number of pages and they, especially for our young adults, the AIS patients" (P7-Nurse)

"Actually, the language of the SRS study is quite difficult to interpret if you're a younger adolescent... I think some of the language in it is quite outdated in comparison to language that we may use now" (P1-Nurse)

Logistic provisions. This was reported by HCPs as barriers that enable the use of digital versions of PROMs. Further, the system that is used to collect data from patients is "not user-friendly" which limit usefulness of the data.

"The Amplitude system is not the most user friendly to get at and I don't bother to look at it because I'm too busy". (P3-Surgeon) *"I think IT and logistical provisions are the constraints".* (P6-Surgeon)

6.6.1.3 Patient-related challenges

This theme described the challenges raised by HCPs to use of PROMs regarding patient's perspective.

Potential invalidity. This refers to the concern of HCPs regarding a patient's understanding of SRS-22r questions. They reported that individuals with AIS may ask for help or clarification from their parent or care team to complete the questionnaire, which affects the validity of the collected data.

"You're having a parent talk it through and then also if you're talking it through, then you naturally may bring in your own judgement or suggestion of what something could be, which again, takes away the validity of it because one understanding, how they interpret it". (P1-Nurse)

Patient preferences. Participants discussed that some patients don't want to "talk about their health condition" or "participate" in research studies which impact the use of PROMs.

"Some of them don't want to talk about their condition. Some of them absolutely… their parents don't want them to be engaged with the research as well" (P7-Nurse)

No discharge evaluation. Physiotherapists highlighted that the last treatment session on many occasions is unknown. As patients or their parents may cancel or stop the treatment without prior knowledge of the physiotherapist. Therefore, no discharge assessment can be performed. This will limit physiotherapist's ability to provide PROMs to patients to evaluate their outcomes.

"So, there's not like this formal today's the end of your treatment so then I'm going to provide you with all these questionnaires. It's a bit more fluid and that's probably our biggest limitation" (P2-Physiotherapist)

6.6.1.4 Knowledge, education, and perceived value

This theme described the barriers recognised by HCPs with regards to the knowledge, education, and the value of using PROMs in routine practice.

Questioning importance. Participants questioned the importance of using PROMs in their clinical practice.

"I think if we were to implement some sort of outcome measure, generally, there's quite a lot of effort behind why it's important and how it will improve my life, how it will improve my patient's lives" (P3-Surgeon)

Lack of knowledge. Participants reported that they did not know about the PROMs questions, or what is the PROMs score could tell.

"Again, I don't really understand the questionnaire. Nobody has ever explained it to me, and I've been in the role for eight years. You get given a percentage at the end of it and I have no idea what that percentage means" (P7-Nurse)

Need of education. Others recognised that that they have "never reviewed their practice" with individuals with AIS. They also reported that HCPs "don't want to change" and that a "lot of education" is required to implement changes in practice.

> *"My experience of consultants, people like me and colleagues, is that you do it one way and you don't ever change…someone like me to change what they do to a different thing, can take quite a lot of education"* (P3-Surgeon)

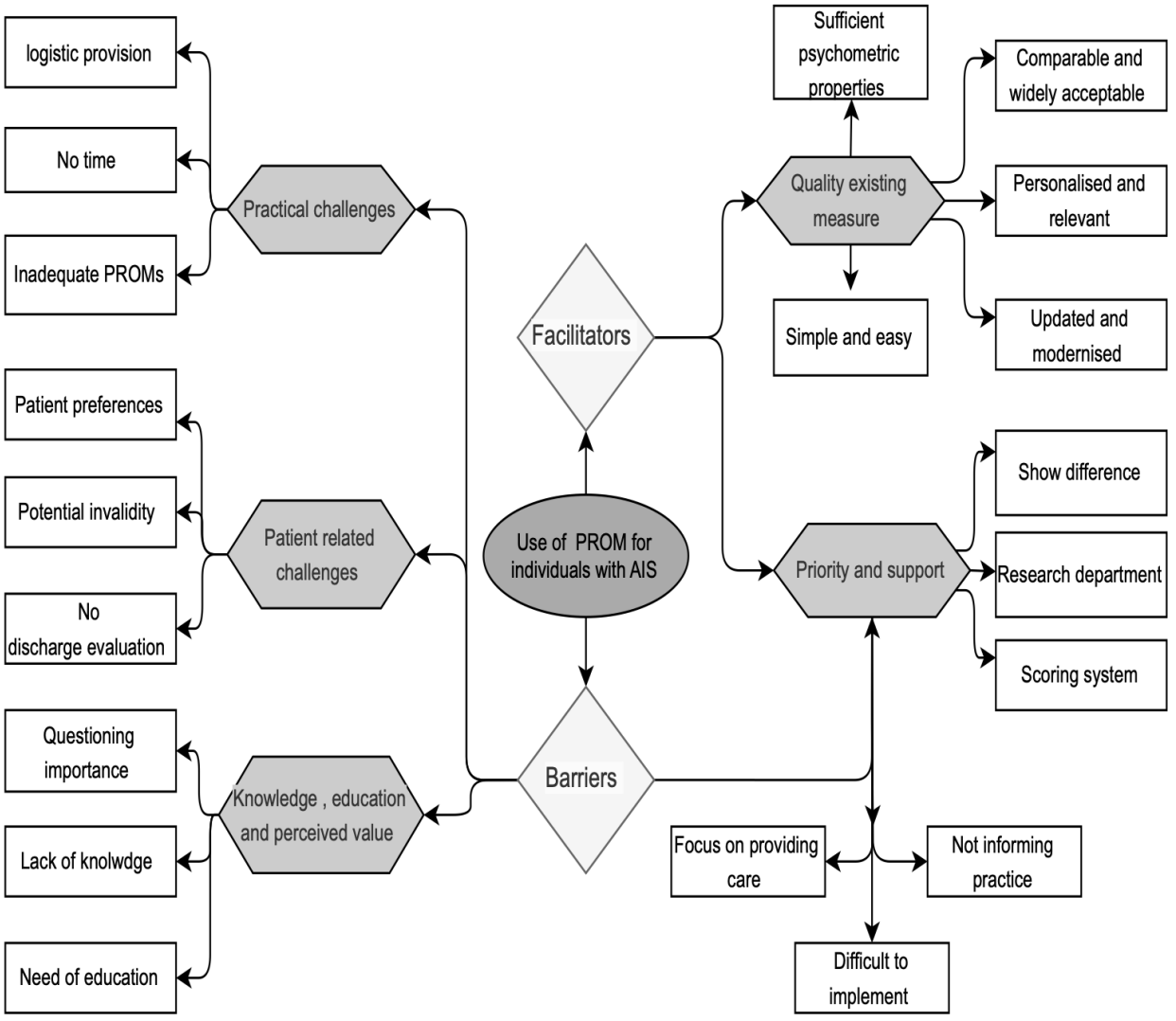


Figure 6.1 : Barriers and facilitators identified from participants data regarding use of patient reported outcome measure for individuals with AIS

6.6.2 Facilitators

Two main themes were identified as facilitators to use PROMs for individuals with AIS: quality existing measures and prioritization and support.

6.6.2.1 Quality existing measure

This theme includes the characteristics identified by HCPs that qualifies that the PROM is a "good existing measure" to enable its use for individuals with AIS.

Sufficient psychometric properties. Participants reported that the PROM should have certain characteristics to facilitate its use in practice. Such as "being repeatable", "sensitive to change", and having "minimal number of errors".

> *"I think the ideal thing would be something that means something to the patient, that changes with intervention or disease progression. So, you can show there's a change. That's easily understandable. That is easily measurable, however you measure it. It's repeatable, has minimal amount of error in it".* (P3-Surgeon)

Comparable, and widely acceptable. HCPs reported that the PROMs should be widely accepted to enable results to be compared between different treatments and among patients in different countries.

> *"If you found a questionnaire that had good psychometric properties that no-one else used, then, if you were to do a study on it, it would be very difficult to compare your results to other studies"* (P4-Surgeon)

Personalised and relevant. These features of the PROMs were highlighted by HCPs, and it was stated that it should be personalised to individuals with AIS.

> ***"****I think if we're going to measure outcome, which has to be front and centre... But it also then has to be personalised to that individual".* (P3-Surgeon)

Updated and modernised. Participants highlighted that they need a PROMs that is updated to the young generation to facilitate its use and have better engagement.

> *"I think if we had a different type of questionnaire that was more able to engage the appropriate population and updated, easy to understand terminology... we would get more accurate data… there's so much changes within practice … and the mindsets of our young people as well, that now is the time for it to kind of be modernised"* (P1-Nurse)

Simple and easy. Participants reported they need a questionnaire that has simple and easy wording for their young individuals with AIS.

> *"With the younger adolescents, you just need to keep it fairly simple wording".* (P5-Physiotherapist)

> *"I want something that is easy to use on my patients, easy to interpret for me. Something that is readily available".* (P7-Nurse)

6.6.2.2 Priority and Support

This theme described what HCPs need to support and prioritize the use of PROMs in routine clinical practice.

Research department. Participants reported the need of having a big research department to help improve the application of OM in practice, by employing staff designated to collect data from patients without interfering with patient or clinician time.

> *" Those are all things that we wish we could do, but we need a bigger research department to be able to achieve this"* (P1-Nurse)

Scoring system. Participants discussed the need of having a scoring system that could help in evaluating treatment effectiveness and identify patient problems.

".. it's good if we have a scoring system, if you were to compare either two treatment modalities or we want to look at the outcome of a particular ...That's important…the score domains are useful to identify any particular problem with the patient".. (P6-Surgeon)

Show difference in practice. Participants reported that to enable use of PROMs in practice, that OMs should show a difference in their routine clinical practice.

"I think if we had an outcome measure that could show there's a benefit to the surgery, … then go to my colleagues and say, this is important, because look, this shows what you do makes a difference. That would be worth having…" (P3-Surgeon)

6.7 Barriers and facilitators to use performance measure

6.7.1 Barriers

Two themes emerged as barriers and one theme was identified as a facilitator regarding use of performance measure in routine clinic practice among in individuals with AIS. Figure 6.2 shows the identified themes and subthemes.

6.7.1.1 Perceived value and Knowledge

PROMs are more important. Participants perceived that the performance measure is not important as a self-reported measure.

"There are some situations where performance based is important to take decisions. But in scoliosis it's a little more subjective in terms of outcomes. If a 14-year-old comes back and says, "I'm back playing football", at the six-month mark, that to me is more important than what the six-minute walking score is" (P6-Surgeon)

Performance indicators differ across individuals. Participants discussed that although the performance measures are applicable to individuals with AIS, the indictors of performance measures differed across individuals.

> *"The issue there would be what performance measure do you put most emphasis on, in an individual patient? There are so many different performance indicators that you could use, that one could potentially use….the issue, is what's important to one child will be very different to another child*" *(*P4-Surgeon*)*

Unaware about use performance measure. Study participants discussed that their knowledge and experience about using performance measures in those with AIS is limited.

> *" They do this quite a lot don't they, with shoulders, hips and knees and other elements, don't they? But I have to be honest, I haven't seen it routinely used within adolescent spinal deformity "* (P1- Nurse)

6.7.1.2 Practicality

Physical space and time. Participants reported that the main barriers to use performance measures in clinics for individuals with AIS, are the physical space and the time needed to perform it.

> *"I think if we're thinking about some sort of physical activity, then the problem we'd have with AIS, is physical space. I can do a paper form in clinic because I give it to the kid who is sitting on the chair. But if I want someone to, I don't know, walk them down the corridor 20 times. it's the practicalities of how you do that and how I do it with 30 children at the same time essentially….That would be the thing for me"* (P3-Surgeon)

6.7.2 Facilitators

6.7.2.1 Practical consideration.

This theme related to the characteristics of performance measures that may enhance its use in clinical practice per participants reports.

Relevant. Participants reported that the performance measure should be relevant to the individuals with AIS.

> *"I would group the patient. If you're looking at scoliosis, I would look at adolescent idiopathic, versus neuromuscular, versus syndromic. Then I might choose an outcome measure that would be relevant to that particular (sub-) group of patients"* (P4-Surgeon).

Simplicity and acceptability. Participants also discussed the simplicity and acceptability of performance measures, that it is acceptable to individuals with AIS and can be applied quickly in the clinic were facilitators to use performance measures.

> *"If we were just looking at, I don't know, say five key areas and what's their flexibility, what are they able to achieve in regard to, ... very simplistic system, then that could be applied fairly quickly"* (P1-Nurse).

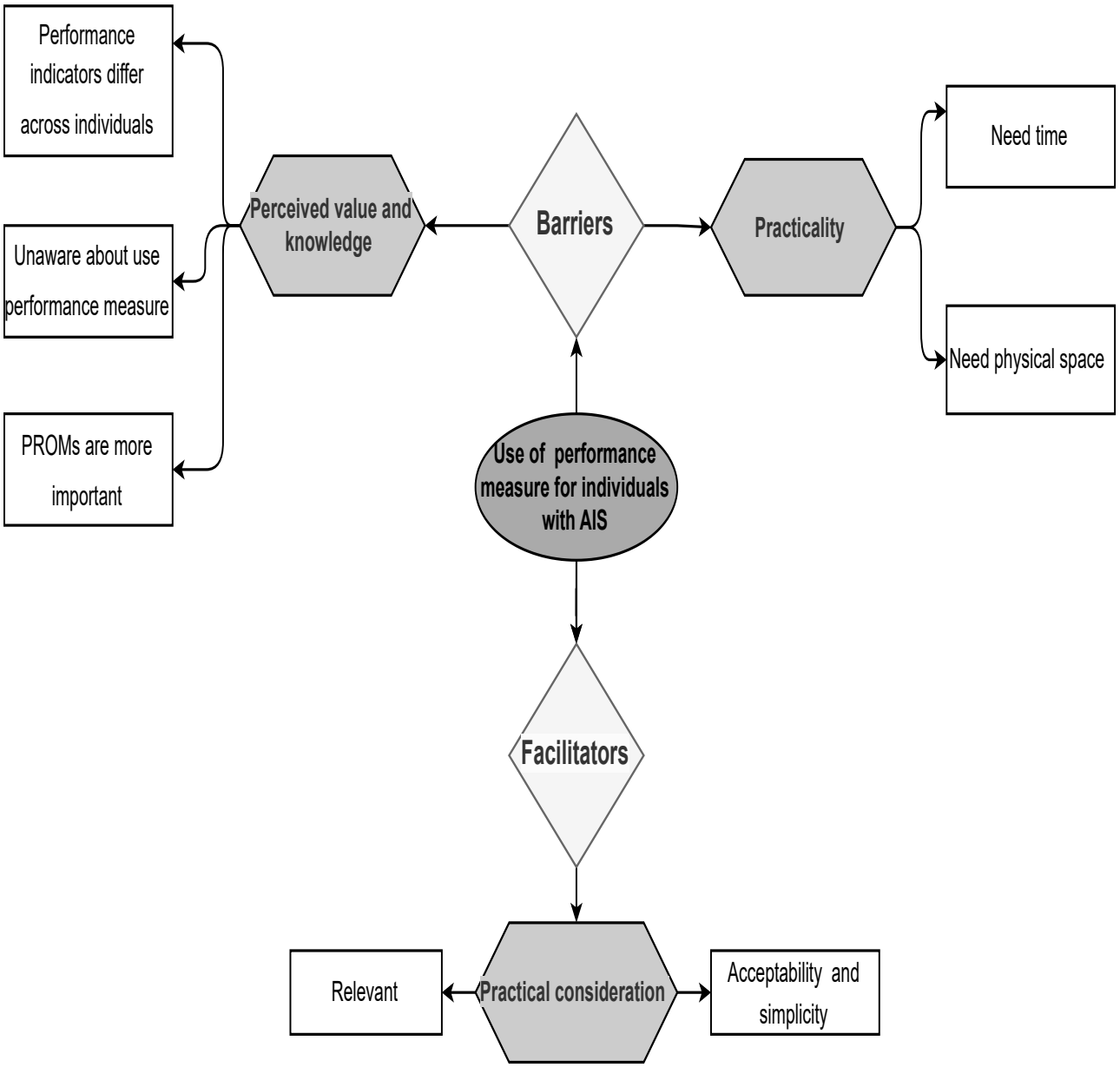


Figure 6.2 : Barriers and facilitators identified from participants data regarding use of PBOMs for individuals with AIS

6.8 Discussion

This is the first qualitative study that has evaluated the perspectives of HCPs in the use of OMs among individuals with AIS. Participants in this study were derived from different professions, all with considerable experience of AIS. Despite the perceived importance and value of using OMs, findings indicate limited use of OMs in routine clinical practice. Themes identified as barriers to the use of PROMs are knowledge, education and perceived value, priority and support, patient related, and practical challenges. Having a quality existing measure and support were identified as the main facilitators to use PROMs for individuals with AIS. Barriers to adopting PBOMs for individuals with AIS are perceived value and knowledge as well as practicality. Facilitators that promote use of PBOMs for individuals with AIS were being relevant, simple, and acceptable to individual with AIS.

6.8.1 Current practice

The British Spine Registry (BSR) was established in 2012 in UK, to monitor the outcome of patients undergoing spinal procedures, including AIS. Despite the inclusion of SRS-22 and Visual Analogue Scale, our findings suggest that the system is not fit for purpose in the AIS population. Participants reported that the system is not user-friendly, they lack understanding of the included questionnaires and what scores mean. According to a recent study, the BSR response rate dropped from 58% completed preoperatively to 25% one year afterwards (Cunningham et al., 2020). Furthermore, a low response rate to automated BSR email reminders was also observed, with 56.7% at 6 weeks dropping to 48% at a 1 year follow up (Jasani et al., 2016). As per our participants' reports, a variety of factors, including a lack of time and a burdensome questionnaire, were recognised as obstacles to compliance (Cunningham et al., 2020).

In this study, participants valued assessment of pain and function for those with AIS. They recognised the importance of using PROMs in practice to monitor individuals with AIS outcomes. However, all participants reported using informal self-reported measures, and they link it to patients' goals and needs, a finding consistent with other research in different patient populations (Briggs et al., 2020, Demers et al., 2019, Ntsiea et al., 2022).

6.8.2 Barriers and facilitators to the use of PROMs

Inadequate PROMs design and content was reported as a practical challenge and a barrier to use of PROMs in practice. The widely used SRS-22 was described by participants as being irrelevant, lengthy, and its language outdated for young individuals such as those with AIS. They were also concerned about validity of findings where patients needed assistance with understanding and completion. However, existing evidence suggests that there are no differences in SRS-22 scores if completed by the individuals with AIS or their parents (Brewer et al., 2014). Meanwhile, a study has been designed to evaluate content validity of the SRS-22r for those with AIS (Alamrani et al., 2021a). Content validity is the first and most important measurement property that should be assessed before a PROMs can be used in practice (Mokkink et al., 2018a).

Other reported barriers included non-participation in research with families not wishing to talk about their condition, restricting use of PROMs in practice. This is consistent with reports from previous studies among individuals with AIS waiting for surgery, as those with AIS tend to hide their condition from others (Essex et al., 2022, Motyer et al., 2022). For this current study, participants reported focusing on providing care to their patients, and the use of PROMs is difficult in daily practice, consistent with other reports (Briggs et al., 2020). They perceived that the use of PROMs is for research purposes only, and that they need to see change in practice. This is consistent with the

national guidance that recommended that HCPs should understand how the use of PROMs can be beneficial and lead to changes in their practice (Excellence, 2007). Participants, on the other hand, indicated that they require a large research department dedicated to collecting data from patients in order to improve its usage in practice. The wider literature indicated that 55% of HCPs require support from staff in data collection (Falavigna et al., 2017). Lack of knowledge or the perceived value of a PROMs was also reported as a barrier (Foster et al., 2018, Duncan and Murray, 2012, Fleischmann and Vaughan, 2018, Falavigna et al., 2017), however the provision of training, education and support on the value of using PROMs and how to use them were considered facilitators (Foster et al., 2018).

A quality existing PROM is a key theme that has been recognised as facilitator to PROM use in practice. Sufficient measurement properties such as validity and reliability were discussed and are widely recognised in the literature (Mokkink et al., 2010). Other PROM characteristics included being relevant, simple, easy to use and comparable with other reports (Prinsen et al., 2016). This indicates that HCPs value the use of PROMs, however they perceived that the available PROM lacks the characteristics of a quality measure. Therefore, it has been recommended that healthcare organisations should invest time and resources, in designing PROMs to facilitate use in practice (Foster et al., 2018).

6.8.3 Barriers and facilitators to the use of PBOMs

Participants reported minimal knowledge and experience about PBOM, placing greater value on PROMs in AIS. Practical issues of such measures were identified as barriers i.e., the time and physical space needed; consistent with PROM studies where time to collect data acts as barrier (Amini et al., 2021, Ntsiea et al., 2022). These findings are consistent with previous reviews, where most of

measurement properties studies identified were from a PROM compared to performance measure (Alamrani et al., 2021b). However, it is well recognised that although PROMs are important, they measure only one aspect of a condition and it should be associated with physical performance assessment (Master et al., 2020). The PBOMs are commonly used in other patient groups, such as those with upper limb (Wang et al., 2018), knee and hip (Kroman et al., 2014), and low back pain (Ferguson et al., 2005, Master et al., 2020, Gilmore et al., 2019). Up to now, little attention has been paid to the evaluation, applicability, and acceptability of such measures to those with AIS; this would be a fruitful area for further work.

Practical considerations are the main theme identified as facilitators to the use of PBOMs for individuals with AIS. Participants reported that to enhance its use, it should be acceptable, relevant, and applicable. Consistent factors such as feasibility and interpretability were identified as important to using performance tests in sport and exercise fields (Robertson et al., 2017). Ensuring that the measures are acceptable to the patient, as some patients and their families may not be willing to participate or results may not be accurate, which would make it difficult to draw valid conclusions from the data. Thus, patient and public involvement becomes an integral requirement in designing research studies and reporting results (Staniszewska et al., 2012).

6.9 Strengths and limitations

This study was designed and reported in line with published guidance of qualitative research (COREQ), and a published protocol (Alamrani et al., 2021a). All stages from design to interpretation involved experts in qualitative research, HCPs representation and a PPI representative, thus enhancing trustworthiness and credibility of findings. The outcomes of this study present the perspectives of experienced professionals with this specific group, reflecting various credentials and

professions. The main limitation of this study was designed as focus group discussion (Alamrani et al., 2021a), however, due to HCPs job commitments, and COVID restrictions in place, this was deemed impossible, and therefore virtual interviews were conducted instated (Carter et al., 2021). the sample of this study was recruited from one tertiary hospital in UK which may limit transferability of the findings. Pilot interviews were not undertaken; however, the development of topic guide was strengthened by an input from a PPI representative and the expertise of co-authors. Although, saturation was achieved in this study, it was determined retrospectively after completion of all interviews.

6.10 Implications for practice and future research

Using of OM for individuals with AIS has a variety of implications on clinical practice. It enables providing patient-centred care that may improve patient outcomes, and result in more effective use of healthcare resources (Richards et al., 2015). Understanding of barriers and facilitators identified in this study could inform the development of strategies and polices to promote the use of OMs for individuals with AIS. Future research could identify the long-term effects of using distinct types of OMs for individuals with AIS, on outcomes and HRQOL of those individuals.

6.11 Chapter summary

This chapter evaluated the use of OMs as perceived by HCPs when assessing outcomes of individuals with AIS. In-depth-semi structured interviews involved a purposive sample of HCPs. The interviews were recorded, transcribed, and then analysed. Although HCPs perceived the value of using OMs, current practice indicates its limited use among individuals with AIS. Knowledge, and support to use PROMs as well as practical and patients-related challenges work as barriers that

constrain PROMs use, while providing support and having quality PROMs facilitate its use. Barriers to adopt PBOMs are a lack of knowledge and perceived value alongside the practicality, while practical consideration as acceptability function as an enhancer to the use of PBOMs among this population

Chapter Seven

GENERAL DISCUSSION

7.1 Summary of Thesis Findings

The aim of this thesis was to explore the current use of OMs for individuals with AIS and evaluate their measurement properties. *Chapter One* provides background information around AIS including its clinical features and management approaches as well as the outcomes of theses management using the ICF as a framework. The chapter then discussed the development of PROM and emphasises the importance of assessment of measurement properties, particularly content validity. Areas of research that have not been fully documented in the literature were therefore determined to be objectives for this thesis.

In *Chapter Two,* a retrospective longitudinal observational study (Alamrani et al., 2023b) is presented. The study used the SRS-22r and analysed the functional outcomes of individuals with AIS using routinely collected data. A comparison was made between the outcomes of those who reach the SDC and those who did not. Additionally, the study aimed to identify factors that may predict achievement of these scores. The findings indicated that individuals with lower scores before surgery were more likely to achieve the SDC in pain and function following surgery, suggesting a deficit in the SRS-22r (Alamrani et al., 2023b). This highlights the importance of identifying appropriate OMs that can confidently assess function in individuals with AIS (Alamrani et al., 2023b). Therefore, a subsequent systematic review was conducted in *Chapter Three*.

Chapter Three details a systematic review designed in two stages which aimed to identify current OMs used to evaluate PF in individuals with AIS (Alamrani et al., 2020, Alamrani et al.,

2021b). The findings highlighted that the current PROMs had insufficient measurement properties based on COSMIN guidelines, particularly due to the lack of content validity assessment for any of the identified PROMs. To address this issue, studies in C*hapters Four* and *Five* were conducted to evaluate the content validity of the (SRS-22r), a commonly used PROM in the AIS literature (Alamrani et al., 2021a).

Chapter Four present a mixed method study designed to assess the content validity of the SRS-22r through cognitive debriefings with HCPs and individuals with AIS. Problems were identified in the questionnaire contents related to comprehension, comprehensiveness, and relevance. Further, refinements were suggested by participants to the, terminology and language of the questions and response options. These findings indicated a need for a concept elicitation study.

In *Chapter Five* a qualitative study was needed to evaluate the HRQOL concepts that are most important to individuals with AIS (Alamrani et al., 2023a). The findings revealed that the AIS has a broad impact on HRQOL of the affected individuals. However, when themes and codes were compared to the content of the SRS-22r, a weak association was found. The findings reported in *Chapters Four and Five* (Alamrani et al., 2023a) provide important evidence for the need to refine the current SRS-22r or develop a new PROM that is relevant to the experiences of individuals with AIS, while considering age and language appropriateness.

Additionally, the systematic review in *Chapter Three*, revealed limited studies that evaluated the measurement properties of PBOM and body structure and function measures, highlighting their limited use in practice. This was further evaluated in *Chapter Six,* by exploring the perspectives of HCPs on the use of PROMs and PBOMs for individuals with AIS through qualitative interviews. It revealed that the HCPs value the use of OMs for individuals with AIS; however, their current practice does not involve standardised use of OMs, including both PROMs and PBOMs.

The purpose of this last chapter, *Chapter Seven*, is to provide a concise overview of the primary findings of this thesis, outline conclusions based on results, discuss the strengths and limitations of the presented research, and explore the clinical implications of the findings along with recommendations for future research.

7.2 Assessment of functional outcomes

The study in *Chapter Two,* the evaluation of the functional outcomes of individuals with AIS was performed using the SDC value instead of the MIC. Additionally, factors that could predict changes in function and pain domains were assessed (Alamrani et al., 2023b). Findings revealed several issues related to the SRS questionnaire. Function showed minimal improvement over time, with a slight increase (+0.2) at 1 year and (+0.3) at 2 years compared to other domains. This was attributed to the lack of content validity or the presence of a ceiling effect. Despite revisions made to enhance the internal consistency of the SRS-22r (Bastrom et al., 2015, Asher et al., 2003b), ceiling effects persisted, limiting its ability to detect significant changes beyond its score range (Streiner et al., 2015).

At 1 year, only few individuals with AIS achieved the SDC for most SRS-22r domains, and successful individuals had low scores pre-surgery (Alamrani et al., 2023b). On the other hand, individuals with high scores prior to surgery were categorized as unsuccessful, indicating a defect in the SRS-22r (Alamrani et al., 2023b). Changes in function following surgery were predicted by older age and low function scores pre-surgery, while changes in pain outcome were predicted by an increase in age, length of hospital stays, male gender, high function scores, and low pain scores pre-surgery (Alamrani et al., 2023b). These findings are in line with previous reports (Sanders et al., 2010, Rodrigues et al., 2017, Sieberg et al., 2013), and suggest the need to revise the SRS-22r for young individuals with AIS (Parent et al., 2010, Rodrigues et al., 2017). Furthermore, pain was the only

domain in the SRS-22r that worsened at one year following surgery (Bailey et al., 2021), possibly due to pre-existing risk factors like anxiety and inadequate post-surgery pain management (Seki et al., 2018, Fletcher et al., 2015). These findings emphasise the importance of assessing factors contributing to pain in the context of surgery. In light of the high ceiling effects observed in the SRS-22r, the need to identify appropriate OMs that can confidently assess function in individuals with AIS is highlighted in *Chapter Two* (Alamrani et al., 2023b). This sets the stage for further exploration of OMs and their measurement properties in *Chapter Three* of this thesis.

7.3 Measurement properties

As discussed in *Chapters One* and *Three,* PF is considered one of the core outcome domains that should be evaluated and reported in any clinical trial (Taylor et al., 2016b, Dodd et al., 2018). Individuals with AIS commonly report a limitation in their PF (Du et al., 2016a). Therefore, it is important to measure the impact of AIS on individuals' life both in research and clinical practice. However, in *Chapter Two*, the PROM that was used to assess HRQOL for individuals with AIS showed less sensitivity in detecting changes in function due it is ceiling effects (Alamrani et al., 2023b).

Based on the findings in *Chapter Two*, a systematic review was conducted in two stages in *Chapter Three.* The first stage aimed to identify OMs used to evaluate PF for individuals with AIS, and the second stage assessed the measurement properties of these measures (Alamrani et al., 2020, Alamrani et al., 2021b). The COSMIN guidelines for systematic review of PROMs were adhered to ascertain high-quality and trustworthy results (Prinsen et al., 2018). The first search revealed a variety of OMs used in the AIS. However, nine PROMs, just one PBOM, and six measures of body structure and function were identified in the second stage for measurement properties. None of the PROMs

had evidence for sufficient content validity and internal consistency (Alamrani et al., 2021b). According to COSMIN recommendations, these PROMs have potential to be used, but further research is needed to assess their quality (Alamrani et al., 2021b, Terwee et al., 2018b).

An important finding of this review, was that the developmental studies of all identified PROMs were rated as inadequate underpinning their content validity (Alamrani et al., 2021b). For example, the SRS was developed in a population that did not represent individuals with AIS in terms of their age (Asher et al., 2000, Asher et al., 2003c, Asher et al., 2006, Haher et al., 1999), and the SQLI was tested among healthy individuals (Feise et al., 2005). Therefore, more research was required to evaluate content validity of these PROMs. Construct validity was the most frequently tested measurement property for PROMs in individuals with AIS (Glattes et al., 2007, Bouton et al., 2022, Asher et al., 2003c, Feise et al., 2005, Bastrom et al., 2015, Asher et al., 2003a). These PROMs had the potential to be recommended for use to evaluate PF, as they were rated as sufficient against COSMIN criteria, supported by moderate quality of evidence (Alamrani et al., 2021b). However, internal consistency was rated insufficient due to the lack of structural validity (Mokkink et al., 2018b, Alamrani et al., 2021b). Reliability has been rated sufficient for function of the SRS-22, CHQ-CF87, and SAQ (Glattes et al., 2007, Sarwahi et al., 2018), while measurement error for function scale of the SRS-22 and SRS-22r was rated insufficient (Alamrani et al., 2021b). Based on these findings from this chapter, two studies were conducted in *Chapters Four* and *Five,* which were designed to evaluate the content validity of the SRS-22r, the common PROM used to evaluate HRQOL including PF for individuals with AIS (Alamrani et al., 2021a).

Regarding PBOMs, only one study has been identified that assessed the construct validity of the TUG test (Gao et al., 2019). This limits the use of such measures for individuals with AIS.

Therefore, a subsequent study in *Chapter Six* was conducted to understand HCPs perspectives on using PBOMs for individuals with AIS including barriers and facilitators.

As stated in *Chapter One*, PF is also assessed using body structure and function measures such as MST and FTF tests. However, a limited number of studies assessed the measurement properties of these measures, and the literature is focused on the radiographs, the gold standard measure of the curve angle using Cobb method (Alamrani et al., 2021b). The identified measures in this review are inexpensive, easy to implement, and quick to use and could be surrogate to the radiographs, if their measurement properties are established for individuals with AIS (Alamrani et al., 2021b). In summary, more studies on measurement properties were still needed to support recommendation of these measures for research and clinical practice among individuals with AIS.

7.4 Content validity of the SRS-22r

Based on findings from *Chapter Two* and *Chapter Three*, a study was conducted to evaluate the content validity of the SRS-22r using two approaches: cognitive debriefings (*Chapter Four*) and concept elicitation (Alamrani et al., 2023a) (*Chapter Five*). The study was conducted with the same participants during the same interview session. Results were presented and discussed in two separate chapters. This approach was chosen to provide a more detailed presentation and analysis of the results, and to enhance clarity and comprehension for the reader.

As has been discussed in *Chapter One*, section 1.6, the development of a PROM involves several steps, which is an iterative rather than linear process requiring re-evaluations and adaptations of the PROM, before it can be used in its final version (Patrick et al., 2011a). (*See Figure* 1.7). In this thesis the development of the SRS-22r was assessed and qualitative assessment were conducted to evaluate the content validity of the SRS-22r. The findings shows that further assessment of the

SRS-22r is still needed before it can be used, and its psychometric properties assessed in its definitive version (*See Figure* 7.1). The current version of the SRS-22r still required assessment as it not adequate for this population.

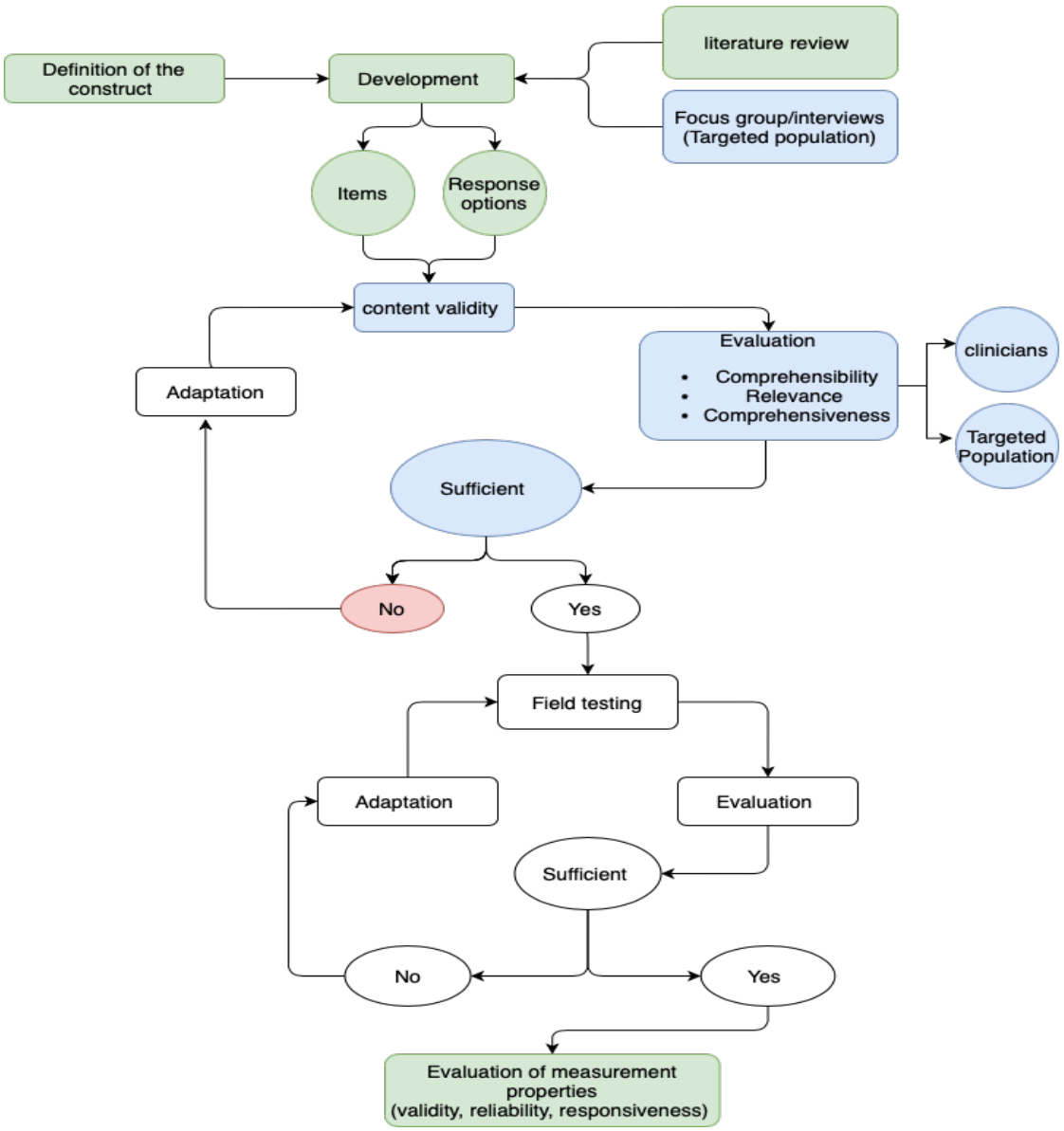


Figure 7.1: Overview of the steps in the development and evaluation of a PROM.

Figure from (De Vet et al., 2011). The blue coloured shapes are the steps completed in this thesis

Content validity is considered the most important measurement property and the first to be considered when selecting a PROM (Terwee et al., 2018b). It ensures that the contents of the PROM are comprehensive, comprehensible, and relevant to the population of interest, measuring what is intended to measure (Terwee et al., 2018b, Brod et al., 2009). To ensure that the PROM has sufficient content validity, the perspective of the targeted population should be considered and qualitative interviews are recommended (Terwee et al., 2018c, De Vet et al., 2011). Ideally, cognitive debriefing interviews should be conducted following the concept elicitation interviews, to evaluate whether the PROM developed from these interviews is suitable to the population of interest (Terwee et al., 2018b). In this thesis, however, the cognitive interview study was presented prior to the concept elicitation study because the SRS-22r had not been assessed by cognitive debriefings evaluation before.

Chapter Four and *Chapter Five* present findings on the content validity of the SRS-22r obtained through in depth semi-structured interviews with individuals with AIS and HCPs following COSMIN recommendations (Terwee et al., 2018b). The findings form *Chapter Three* (Alamrani et al., 2021b), highlighted that the previous research studies focused on evaluating the validity of the PROM using quantitative approaches (Asher et al., 2003c, Asher et al., 2000, Asher et al., 2006, Asher et al., 2003d, Glattes et al., 2007, Asher et al., 2003e), without attention being given to qualitative methods (Alamrani et al., 2021b). Errors within the questionnaire such as ambiguous or confusing questions, required more attention and continued research, as any small modification in the wording, formatting and design of the questionnaire makes a significant change in the responses of the respondents (Willis and Artino, 2013, Willis, 2017). In *Chapter Four,* the cognitive debriefings approaches was used to evaluate content validity of the SRS-22r in accordance with COSMIN recommendations (Terwee et al., 2018b). Despite the SRS-22r has undergone cultural adaptations into several languages and has been in use for many years (Alamrani et al., 2021b), it has not been systematically evaluated using cognitive debriefings.

Relevance assessment ensures that the PROM sufficiently assesses all key areas of the health condition and treatment (Matza et al., 2013, Terwee et al., 2018b). For the HRQOL questionnaires for an adolescent it should be tailored to the experiences, activities, and environments that are specific to the target population's age (Matza et al., 2004, Lohr and Zebrack, 2009, Matza et al., 2013). Two self-image, two mental health and financial difficulties questions were reported as irrelevant by both groups. Suggestions were made by participants to delete the financial difficulties question as it is less relevant to children living in the UK who are treated by the publicly funded NHS system and add unnecessary worry to children (Rullander et al., 2017, Essex et al., 2022, Motyer et al., 2022). Attractiveness and clothes questions were also reported to be irrelevant to the adolescent's experience with scoliosis. Further, mental health question was reported as irrelevant as it lacked necessary reference to the back by being too generic and not specific enough to AIS. This was explained by the fact the questions were derived from the SF-36 questionnaire, which was primarily designed for adults rather than adolescents with AIS (Asher et al., 2006, Asher et al., 2000).

The use of age-appropriate language is emphasised by the recommended practice to help respondents understand the questions and response options (Matza et al., 2013, Matza et al., 2004). This can be achieved by using the same language that individuals with AIS use during concept elicitation interviews (Matza et al., 2013), which are presented in *Chapter Five* (Alamrani et al., 2023a). Both groups of participants reported problems in the comprehension of questions as being negative, emphasising as particular aspect, sensitive, or vague, that affect the overall understanding. Additionally, some responses options used language inappropriate for adolescents such as the use of "narcotic" to describe the pain medications. Therefore, it has been recommended to perform pilot testing and cognitive debriefings with the intended population at different age groups, when developing a questionnaire to determine the age appropriateness (Matza et al., 2004).

Participants who had surgery reported that the six-month recall period is inappropriate, consistent with HCPs notes. The common recall period for PROMs that assess HRQOL of children is the past four weeks or the past month (Landraf et al., 2005). A long recall period are likely to undermine content validity, as the response is likely to be influenced by the patient's state at the time of recall (Health et al., 2006). Thus, items with short recall periods are usually preferable (Matza et al., 2013).

The SRS-22r included questions about HRQOL of individuals with AIS (Asher et al., 2006, Solans et al., 2008). However, participants suggested including questions about sport and school related activities, which were identified as important to individuals with AIS and has been revealed in the concept elicitation study (Kakar et al., 2017, Fabricant et al., 2012, Alamrani et al., 2023a). It also shows that the knowledge of scoliosis and the treatment options available, is also an important aspect to this population consistent with the previous qualitative studies (Motyer et al., 2021a, Motyer et al., 2022, Alamrani et al., 2023a). Including such questions would provide valuable insights into the patient's level of understanding of their condition, which could have implications for their adherence to treatment plans, and overall management of their health condition (Motyer et al., 2021a). Study participants provided examples of potential changes to the wording of questions and response options that could improve the content of the SRS-22r questionnaire in *Chapter Four*.

The concept elicitation interviews in *Chapter Five* used words and phrases from participants with AIS in the generation of concepts, to ensure that the data are a true reflection of participants' perspectives (Alamrani et al., 2023a). When the emerged qualitative data were compared to the contents of the SRS-22r, a poor match was found indicating a lack of content validity of the SRS-22r (Alamrani et al., 2023a). Furthermore, the terminology used in the SRS-22r does not reflect the language used by participants with AIS (Alamrani et al., 2023a). Findings from this study provided

strong evidence for the need to revise the SRS-22r, or development of a new PROM using data similar to that generated from this study that represents the voice of adolescents with AIS, that is language and age relevant (Alamrani et al., 2023a). Concepts elicited form interviews were physical, activity, psychological and social effects. Although, it seems consistent with the main domains of the SRS-22r, i.e., function, pian, mental health, self-image, and satisfaction. The matching analysis between the content (items) of the SRS-22r and the codes elicited from interviews revealed a weak association (Alamrani et al., 2023a).

Pain was the main and most reported symptom reported by participants, consistent with all previous reviews (Théroux et al., 2017, Bastrom et al., 2013, Perry-Eaddy, 2018), associated with preforming exercise (Thompson et al., 2019, Monticone et al., 2014), or surgery which is a major concern for individuals with AIS and their parent prior to surgery (Motyer et al., 2021a, Motyer et al., 2022). An important finding that indicates lack of content validity of the SRS-22r, was the terminology used by participants as they used "hurt" rather than "pain", to describe their pain experience (Alamrani et al., 2023a). One of the important aspects of content validity is that the PROM should be age and language relevant (Matza et al., 2013, Haynes et al., 1995). Different symptoms were also reported and were not included within the SRS-22r, such as difficulty in breathing, stiffness, hip, and shoulder pain as well as body asymmetry consistent with previous studies (Motyer et al., 2022, Weinstein et al., 2008, Altaf et al., 2013, Aroeira et al., 2016).

The impact of AIS on PF is particularly evident during performing daily life activities, especially when engaged in prolonged periods of sitting, standing, bending walking and balance (Motyer et al., 2022, Alamrani et al., 2023a, Nault et al., 2002, Yang et al., 2013, Danielsson et al., 2006, Nokariya et al., 2022). Symptoms are aggravated when carrying backpacks and going upstairs (Schmid et al., 2020, Motyer et al., 2022, Lima et al., 2018). Another important finding was the effect

of AIS on school performance including difficulties in maintaining focus during lesson time and lower school attendance rate (Groenewald et al., 2020, Willimon et al., 2019).

Individuals with AIS experience significant negative psychological feelings including; frustration, irritation, disappointment, and annoyance as well sadness (Toye et al., 2016). They also express worries and fear about the need for surgery and the effects of living with scoliosis in the future (Rullander et al., 2017, Essex et al., 2022, Motyer et al., 2022). These individuals feel insecure to change in front of others and they conceal their back from others (Essex et al., 2022, Motyer et al., 2022, Alamrani et al., 2023a). The impact of AIS extends to sleep quality both before and after surgery (Yakut et al., 2022, Rullander et al., 2013) and overall dissatisfaction with body image (Bertuccelli et al., 2022). It is worth noting that the SRS-22r which evaluates mental health in adolescents, was found to use language that is more suited for adults, as discussed in *Chapter Four*. Individuals with AIS tend to reduce their participation in social and sporting activities (Kakar et al., 2017), despite the importance for their physical and psychological development (Huus et al., 2021). Providing support for those individuals is crucial to help in reducing stress and improve pain coping (LaMontagne et al., 2004, Rullander et al., 2016, Bull and Grogan, 2010, Rullander et al., 2013). This support should include providing comprehensive information about their scoliosis and management options (Essex et al., 2022, LaMontagne et al., 2004).

Findings from studies in *Chapter Four* and *Chapter Five*, suggest that the SRS-22r questionnaire requires updates and modifications to better reflect the experiences of individuals with AIS. The concept elicitation study highlighted the importance of assessing impact of AIS on physical, activity, psychological, and social aspects (Alamrani et al., 2023a). Additionally, the cognitive interviewing study provided further evidence that the current terminology used in the SRS-22r is difficult for younger individuals to comprehend and irrelevant to their experiences, as it was not

developed based on qualitative input from adolescents with AIS. Therefore, revisions and modifications are necessary to ensure that the SRS-22r is more relevant and comprehensible for this population (See figure 7.1).

7.5 Use of Outcome Measures for individuals with AIS

The aim of this thesis was to explore the current use of OMs for individuals with AIS and evaluate their measurement properties. A qualitative study was conducted with HCPs to address three specific objectives. The first objective focused on the current use of OMs, revealing that their use was limited according to interviews with HCPs. While recognising the importance of using OMs, HCPs expressed that their primary focus was providing patient care, leading to limited incorporating PROMs in their daily practice. PROMs were primarily used for research purposes, consistent with other reports (Briggs et al., 2020). The findings indicated that the HCPs highly valued the assessment of pain and function for individuals with AIS, which is consistent with findings from the study in *Chapter Five* (Alamrani et al., 2023a). As individuals with AIS reported that the AIS has an impact on their pain and function level (Alamrani et al., 2023a).

While there is an existing system known as the British Spine Registry for evaluating outcomes in individuals with AIS, the findings from *Chapter Six* indicate that registry is not fit for purpose. The system was found to be non-user-friendly, and HCPs lacked a clear understanding of the included PROMs and the significance of the scores provided. This lack of understanding and usability likely contributed to the low response rate to the PROMs, as demonstrated in *Chapter Two*, where more than 60% of the data were missing at the 1-year follow-up (Alamrani et al., 2023b). This issue is consistent with previous studies that also used the same system (Jasani et al., 2016, Cunningham et al., 2020). Therefore, there is a need for a more user-friendly system and a system that enables data completeness to effectively evaluate outcomes in individuals with AIS across specialist centres.

The second objective of the study in *Chapter Six* was to identify barriers and facilitators to use PROMs for individuals with AIS. Findings suggested that the currently used PROM i.e., SRS-22r lacks the proper design and contents for AIS population. This was consistent with findings from *Chapters Two, Three, Four,* and *Five* (Alamrani et al., 2023b, Alamrani et al., 2021b, Alamrani et al., 2023a). Participants reported that the SRS-22r was irrelevant, lengthy, and using outdated language for young individuals such as those with AIS, as has been shown in *Chapter Four* and *Chapter Five* (Alamrani et al., 2023a). Non-participation in research and the reluctance of families to discuss their condition were identified as barriers to using PROMs in practice, aligning with the findings from *Chapter Five* and previous studies (Alamrani et al., 2023a, Essex et al., 2022, Motyer et al., 2022). Lack of knowledge, support and the perceived value among HCPs were additional barriers to PROM utilisation (Foster et al., 2018, Duncan and Murray, 2012, Fleischmann and Vaughan, 2018, Falavigna et al., 2017). Whereas, having a high-quality existing PROM which possess sufficient measurement properties, relevance, simplicity, ease of use and comparable with other reports, was recognised as facilitator (Mokkink et al., 2010, Prinsen et al., 2016). Further, the provision of training, education and support on the value and usage of PROMs, were considered as facilitators (Foster et al., 2018).

The third objective of the study in *Chapter Six* was to assess barriers and facilitators to use PBOMs for individuals with AIS. Findings from the study in *Chapter Three* shows that only one study has assessed the measurement properties of PBOMs for individuals with AIS (Alamrani et al., 2021b), indicating limited use of PBOM in practice. Results from the study in *Chapter Six* shows that having minimal knowledge and experience about PBOMs and placing greater value on the use of PROMs function as barriers to use PBOM in clinical practice. Additionally, practical challenges such as the need for time and physical space to administer PBOMs were identified, mirroring findings from previous studies (Amini et al., 2021, Ntsiea et al., 2022). Therefore, the feasibility and

interpretability of using OMs was recognised as important factor that should be considered and reported when recommending OMs for use (Mokkink et al., 2018b).

While PROMs are considered important, they measure only one aspect of a condition and should be complemented by PBOM assessment (Master et al., 2020, Taylor et al., 2016b). PBOMs are commonly used in other patient groups, such as those with upper limb (Wang et al., 2018), knee and hip (Kroman et al., 2014), and low back pain (Ferguson et al., 2005, Master et al., 2020, Gilmore et al., 2019). Up to now, little attention has been paid to the evaluation, applicability, and acceptability of PBOMs for those with AIS indicating a fruitful area for further research.

7.6 Heterogeneity of individuals with AIS

The heterogeneity of individuals with AIS poses challenges when selecting and utilizing OMs for assessing their condition and treatment outcomes, since each individual with scoliosis can vary in terms of gender, curve severity, age, symptoms, functional limitations, and psychosocial impact and treatment options and response (de Baat et al., 2012). To consider these factors the following approaches could be helpful when using OMs for this specific population.

1. Engagement of the targeted population in the development and selection of the OMs. This will ensure that the OMs used are relevant, meaningful, and reflective of the individual's experience (Haynes et al., 1995, Terwee et al., 2018b, Terwee et al., 2018c). In this thesis, the targeted population engaged in assessment of the content validity of the SRS-22r *Chapter Four,* and *Chapter Five.*

2. The OMs should be age-appropriate and suitable for the developmental stage of the adolescent (Matza et al., 2004, Matza et al., 2013). In this thesis, the PIS and consent forms as well as topic guide were language and age-appropriate for adolescents. For example, those aged 10-12 years

old had a PIS that is appropriate to their age and reading level and different from those 16-18 years old. Therefore, when evaluating the measurement properties of OMs for adolescents it should involve all age groups.

3. The curve characteristics such as severity and pattern should be considered when selecting OMs for individuals with AIS. In *Chapter Five*, interviews with individuals with AIS who had severe curves indicate that their symptoms and the impact of the AIS on their life, are different from those presenting with mild curves (Alamrani et al., 2023a). Therefore, the OMs should be able to identify changes in both severe and mild curves, as long as the changes are relevant and meaningful to the individual's wellbeing and functional outcomes.

4. Designing of the PROMs to the individual's unique circumstances, goals, needs and concerns. HCPs in *Chapter Six* have suggested this as using specific personal questions. For example, include a question about individuals' feelings and then ask patient to select their feelings and then the reason behind that *(See Chapter Six, section 4.6.4)*. This provides the opportunity for the individual to report on aspects that are personally relevant to them, enabling a more patient-centred assessment.

7.7 Quality of evidence for the conclusions of this thesis

Different risk of bias and reporting tools have been used to ensure the quality reporting of studies in this thesis and they have been mentioned in each study. The STROBE statement was used for designing and reporting the study in *Chapter Two* (von Elm et al., 2014) (See *Appendix* 6). The rigorous COSMIN methodology was followed for conducting the review in *Chapter Three* (Mokkink et al., 2018b). The risk of bias for the review was assessed by the critical appraisal tool; A Measurement Tool to Assess Systematic Reviews 2 (AMSTAR-2) (Shea et al., 2017) (*See Appendix*

7). The majority of the requirements were met, including having a comprehensive list of inclusion and exclusion criteria, a published and registered PROSPERO protocol, an extensive search strategy, and the use of two independent reviewers for research selection and data extraction. All of these aspects ensure that no bias was introduced into the review at any stage (Shea et al., 2017). Although there was a partial reporting of the inclusion of a complete list of justifications for exclusion for each individual full-text article that was excluded, reasons for exclusion were documented in accordance with PRISMA standards at both stages of the review (Moher et al., 2009). The COSMIN risk of bias tool was used to assess methodological quality of individuals studies and modified GRADE criteria were utilised to assess the overall quality of the evidence for OMs across studies (Mokkink et al., 2018a, Balshem et al., 2011). Therefore, it is unlikely that any relevant articles have been excluded or bias was introduced in this review, and we may be confidence in the general results and conclusions of this study.

The COREQ guidelines for reporting qualitative studies were used to design and report the qualitative studies in *Chapter Four, Chapter Five* (Alamrani et al., 2023a) *and Chapter Six (See Appendices 9 and 10*). Rigour was maintained in the qualitative studies in this thesis to enhance trustworthiness and minimize researcher bias. This was achieved through clear research design with clear objectives, appropriate qualitative approach, and detailed methodology (Johnson et al., 2020). The use of purposive sampling strategy was deemed appropriate for the research objectives and allowed for the recruitment of a diverse and representative sample of participants (Mays and Pope, 1995). In-depth semi-structured interviews were used to gather rich and comprehensive data until saturation was achieved (Johnson et al., 2020). A systematic approach was followed in the data analysis to ensure transparency in the analysis process and to arrive at the final themes (Maher et al., 2018). Triangulation was used by involvement of multiple researchers in the analysis, reducing the impact of researcher bias (Mays and Pope, 1995). To enhance the credibility of the results and ensure

that participants' perspectives were accurately represented, the PPI representative (ER) checked the final findings. In this thesis efforts have been made to reduce risk of bias and results are reported clearly.

7.8 Strengths and limitations

This thesis assessed and discussed the use of OMs using different methodologies including qualitative and qualitative approaches. It emphasises the significance of gaining feedback and understanding the perspective of the individuals with AIS regarding their health condition, and the PROM that used to evaluate their HRQOL. Based on the findings on *Chapter Four* and *Chapter Five*, several issues were identified in the SRS-22r, a PROM that has been widely used for several years to evaluate HRQOL for individuals with AIS (Alamrani et al., 2021b).

The study in *Chapter Two* evaluated the clinically relevant changes in surgical outcomes for individuals with AIS, comparing those who achieved SDC in pain and function at 1-year post-surgery and those who did not. The SDC was used since there is no established cut-off point in the SRS-22r that can effectively differentiate between patients (Alamrani et al., 2023b). The findings from this study highlighted many problems within the SRS-22r such as ceiling effects, low response rate (Alamrani et al., 2023b), which has been confirmed, and assessed more in the subsequent studies in *Chapter Three, Chapter Four, Chapter Five, and Chapter Six.*

The two-stage search strategy, in *Chapter Three*, was a strength of the systematic review (Alamrani et al., 2021b). The first stage allowed better insight into the literature around tools used to assess PF for individuals with AIS. This list provides an important inventory of OMs that would be fruitful area for future research by evaluating their measurement properties for individuals with AIS (Alamrani et al., 2020, Alamrani et al., 2021b). An additional strength of this review is that it comprehensively evaluated all types of tools used for assessing PF for individuals with AIS. This

included PROMs, PBOMs as well as body structure and function measures (Alamrani et al., 2020, Alamrani et al., 2021b). The use of COSMIN methodology was another strength as it the preferred approach for the systematic review of measurement properties (Mokkink et al., 2018b).

The content validity of the SRS-22r was evaluated through concept elicitation (Alamrani et al., 2023a), and cognitive debriefings (*Chapter Four* and *Chapter Five*), using rigorous methodology following COSMIN recommendations (Terwee et al., 2018a) and a published protocol (Alamrani et al., 2021a). The concept elicitation study in *Chapter Five* involved a heterogeneous sample from individuals with AIS, representing the population of interest with various ages, gender, curve sizes and management approaches (Alamrani et al., 2023a). The cognitive debriefings study involved HCPs, alongside individuals with AIS. They were part from their health care team with different qualification, specialisms, and experiences. Furthermore, the patient perspective was central in all of these studies, through involvement of PPI representative (ER) in the study design, interpretation of the results and revising the final versions of the manuscript prior publications.

ER has shared her personal experience as individuals with AIS and described how AIS has a significant impact on her mental health (See Appendix 11). This was consistent with the thesis findings as many participants suffered from emotional symptoms that affect their life and they require support from family, friends, and HCPs. ER was diagnosed with scoliosis at the age of 13 and went through a period of insecurity and self-consciousness due to her condition. After undergoing successful surgery, she experienced new symptoms that were eventually diagnosed as somatisation, highlighting the importance of addressing the potential impacts on mental health and providing support within orthopaedic clinics for individuals with AIS.

The main strength of cognitive debriefings study in *Chapter Four,* is that the majority of participants had no previous experience with the SRS-22r which allowed for more accurate responses of their experience and opinions, without any influence from prior learning. The data from interviews

with HCPs in *Chapter Six*, presents perspectives of experienced professionals with this specific group, reflecting various credentials and professions.

However, limitations of this body of work do exist. Although a rigorous systematic review was performed, as highlighted in *Chapter Three*, only studies in English were included, this may introduce language bias. Furthermore, ratings of studies were determined using the lowest score principle, which may underestimate a study's final methodological quality score (Mokkink et al., 2018a).

The main limitation in *Chapter Two,* was the low follow up rate of only 41%, which may introduce some bias into the results (Dettori, 2011). However, previous studies that utilised the BSR reported consistent results, indicating a problem in the PROM that has been used to collect data, this has been confirmed by findings in *Chapter Four, Chapter Five* and *Chapter Six* (Cunningham et al., 2020, Alamrani et al., 2023a).

The participants in studies for *Chapter Four, Chapter Five* and *Chapter Six* were heterogenous. However, they were recruited from a single centre in UK, which may limit transferability of the findings. Recruitment of both groups was challenging for the researcher and led to amendment to the planned protocol of collecting data from HCPs for studies in *Chapter Four* and *Chapter Six* (Alamrani et al., 2021a). Only seven participants out of the thirteen invited agreed to participate. The recruitment of individuals with AIS was also challenging, as a number of participants or their parent withdrew their consent to participate for several reasons such as school or their parents' commitments which was as far as the authors knows was not associated with the participation in the study. Participants with AIS were also not conversational, tended to give short answers and were reluctant despite prompting to elaborate on their responses and give further details. This led to the change in the planned analysis of the data in *Chapter Four*, as interviews with individuals with AIS were quantitively summarised. Ideally the concept elicitation should be performed prior of conducting

cognitive debriefings, to confirm that the concepts identified by the participants are adequately represented in the questionnaire. However, the SRS-22r, has been culturally adapted to various languages and has been used without prior input from the relevant population. Because of the challenges that the researcher faced because of the impact of COVID-19 pandemic, and the recruitment of participants, it was not possible to conduct interview to assess concept elicitations and cognitive debriefings separately.

7.9 Clinical Implications

Using OMs for individuals with AIS has a variety of implications on clinical practice. It highlights the importance of using a valid OMs as they contribute to providing patient-centred care, leading to potential improvement in patient outcomes, and facilitating more effective use of healthcare resources (Richards et al., 2015).

Although findings from study in *Chapter Two* are based on retrospective data, they reveal that the successful group had lower pre-surgery scores compared to the unsuccessful group. Consequently, individuals with lower pre-surgery scores may have little probability of obtaining a meaningful clinical improvement following surgery (Alamrani et al., 2023b). As a result, it becomes important for surgeons to have discussion with their patients about the purpose of the surgery and to manage any misconceptions and unrealistic expectations. This discussion may lead to different treatment strategies e.g. provide rehabilitation prior to surgery or alternative treatment approaches like exercises.

The review in *Chapter Three* has highlighted a substantial gap in the content validity of frequently utilised PROMs for individuals with AIS. This raises concerns about the appropriateness of these measures for the target population. Until additional evidence is established, clinicians should approach the use of PROMs with caution, recognizing potential influences from factors such as pain

and psychological distress commonly reported by individuals with AIS (Bastrom et al., 2013). To ensure a more comprehensive evaluation of PF in individuals with AIS, it is recommended that clinicians consider incorporating a combination of measures. This may include not only PROMs but also alternative functional assessments.

Findings from studies in *Chapters Four,* and *Five,* serve as an important reminder that the widespread use of PROMs does not automatically guarantee their content validity. Testing the content validity of PROM would improve the quality of the data collected from patients and enhance the understanding of patient experience and outcomes. The OMs with good content validity ensure that the assessment tool captures the relevant aspects of a patient's condition. This will enable HCPs to obtain a comprehensive and accurate understanding of the patient's health status and treatment outcomes. Understanding the barriers and facilitators identified in *Chapter Six,* could inform the development of strategies, and polices to promote the use of PROMs and PBOMs for individuals with AIS. This will enable a comprehensive and patient-centred care and facilitate communications between HCPs and their patients.

7.10 Future research

Based on the findings reported in *Chapter Two,* future studies could evaluate the effectiveness of specific rehabilitation programs or pain management strategy on pain and function recovery post-surgery. Further, if the measurement properties of the revised SRS-22r established for individuals with AIS, future research could focus on investigating the cut-off score that can be reliably classify patients into successful and unsuccessful based on their scores (Alamrani et al., 2023b).

The systematic review in *Chapter Three* offers an inventory of OMs that can be used in future research to evaluate their measurement properties in individuals with AIS (Alamrani et al., 2021b).

While the literature is focusing on the use of PROMs to evaluates functional ability of individuals with AIS, future studies may consider use physical performance assessment. This could provide other perspectives on the functional ability of these individuals.

Finally, findings reported in *Chapter Six* can be used to conduct studies evaluating the effects of educational programs and training for supporting HCPs in using PROMs and PBOMs in clinical practice. Longitudinal studies can also be conducted to evaluate the impact of using PROMs and PBOMs on clinical decision making and treatment outcomes for this group of individuals.

7.11 Future research to develop a PROM for individuals with AIS

The findings from *Chapter Four* and *Chapter Five* (Alamrani et al., 2023a) underscore the imperative to develop a novel PROM for individuals with AIS that is informed by qualitative insights (Alamrani et al., 2023a). To achieve this goal, several research steps need to be undertaken.

First, a qualitative concept elicitation study with a purposive sample from individuals with AIS from other centres in the UK. This will allow more comprehensive understanding about the impact of scoliosis at the HRQOL of adolescent.

Second, based on the results from the concept elicitation studies, a draft of the potential new PROM would be developed (De Vet et al., 2011). It should consist of pool of items that is relevant to the experience of individuals with AIS, using their words and phrases that comes from the data revealed from the qualitative interviews. The contents of the PROM should be easily understood by the individuals with AIS regardless of their educational level; by using simple language and avoiding

complex or ambiguous terminology. As well as having clear instructions and short- recall periods (Streiner et al., 2015, De Vet et al., 2011).

Third, a pilot testing using a cognitive debriefing needs to be undertaken to examine the relevance, comprehensibility and comprehensiveness of the PROM items (Terwee et al., 2018b). This could be performed in a focus groups or individuals semi-structured interviews that involve a new sample from individuals with AIS and HCPs (De Vet et al., 2011). The cognitive interviews are conducted in iterative manner. If the first set of interviews reveal any problems, this should be revised in the questionnaire and further cognitive interviews need to be undertaken (Rothman et al., 2009). If no problems are detected, then measurement properties of the new PROM can be tested (De Vet et al., 2011).

Fourth, the developed PROM should be tested to determine its dimensionality using methods like factor analysis and item response theory (De Vet et al., 2011). In this step, a larger sample of individuals with AIS needs to be recruited e.g. n=100 (De Vet et al., 2011). If any problems are detected then adaptation is needed to refine the PROM and retested in a field testing study (De Vet et al., 2011). If no problems are revealed, then the PROM can be used with confidence among individuals with AIS.

7.12 Conclusion

Accurate and reliable assessment of HRQOL using appropriate PROMs is crucial for guiding treatment plans, monitoring patient progress, and improving the HRQOL in individuals with AIS. According to the findings of this thesis, the existing PROMs used in this group lack adequate measurement properties, particularly content validity, which may impair the accuracy of the data obtained. This thesis highlights the importance of involving individuals with AIS in the development

and validation of new PROMs, to ensure that the measures are relevant, meaningful, and reflective of this unique group of individuals. Furthermore, the research provided in this thesis offers compelling evidence to the need to establish a new PROM that are relevant to the age, level of language and experience of individuals with AIS. The limited application of PBOM in clinical practice emphasises the need for additional studies in this field. This can enhance the use of PBOMs and lead to more comprehensive understandings of the impact of AIS on HRQOL of those individuals.

LIST OF REFERENCES

ABMA, I. L., ROVERS, M. & VAN DER WEES, P. J. 2016. Appraising convergent validity of patient-reported outcome measures in systematic reviews: constructing hypotheses and interpreting outcomes. *BMC Research Notes,* 9**,** 226.

ADOBOR, R. D., RIMESLATTEN, S., KELLER, A. & BROX, J. I. 2010. Repeatability, reliability, and concurrent validity of the scoliosis research society-22 questionnaire and EuroQol in patients with adolescent idiopathic scoliosis. *Spine (Phila Pa 1976),* 35**,** 206-9.

ADOGWA, O., ELSAMADICY, A. A., HAN, J. L., CHENG, J., KARIKARI, I. & BAGLEY, C. A. 2016. Do measures of surgical effectiveness at 1 year after lumbar spine surgery accurately predict 2-year outcomes? *J Neurosurg Spine,* 25**,** 689-696.

AGHDASI, B., BACHMANN, K. R., CLARK, D., KOLDENHOVEN, R., SULTAN, M., GEORGE, J., SINGLA, A. & ABEL, M. F. 2020. Patient-reported outcomes following surgical intervention for adolescent idiopathic scoliosis: a systematic review and meta-analysis. *Clinical Spine Surgery,* 33**,** 24-34.

AHN, U. M., AHN, N. U., NALLAMSHETTY, L., BUCHOWSKI, J. M., ROSE, P. S., MILLER, N. H., KOSTUIK, J. P. & SPONSELLER, P. D. 2002. The etiology of adolescent idiopathic scoliosis. *Am J Orthop (Belle Mead NJ),* 31**,** 387-95.

ALAMRANI, S., GARDNER, A., FALLA, D., RUSSELL, E., RUSHTON, A. B. & HENEGHAN, N. R. 2021a. Content validity of Scoliosis Research Society questionnaire-22 revised (SRS-22r) for adolescents with idiopathic scoliosis: protocol for a qualitative study exploring patient's and practitioner's perspectives. *BMJ Open,* 11**,** e053911.

ALAMRANI, S., GARDNER, A., FALLA, D., RUSSELL, E., RUSHTON, A. B. & HENEGHAN, N. R. 2023a. Content validity of the Scoliosis Research Society questionnaire (SRS-22r): A qualitative concept elicitation study. *PLOS ONE,* 18**,** e0285538.

ALAMRANI, S., GARDNER, A., RUSHTON, A. B., FALLA, D. & HENEGHAN, N. R. 2023b. Predictors of Relevant Changes in Pain and Function for Adolescents with Idiopathic Scoliosis following Surgery. *Spine (Phila Pa 1976)*.

ALAMRANI, S., RUSHTON, A., GARDNER, A., FALLA, D. & HENEGHAN, N. R. 2020. Outcome measures evaluating physical functioning and their measurement properties in adolescent idiopathic scoliosis: a protocol for a systematic review. *BMJ Open,* 10**,** e034286.

ALAMRANI, S., RUSHTON, A. B., GARDNER, A., BINI, E., FALLA, D. & HENEGHAN, N. R. 2021b. Physical Functioning in Adolescents With Idiopathic Scoliosis: A Systematic Review of Outcome Measures and Their Measurement Properties. *Spine*

ALANAY, A., CIL, A., BERK, H., ACAROGLU, R. E., YAZICI, M., AKCALI, O., KOSAY, C., GENC, Y. & SURAT, A. 2005. Reliability and validity of adapted Turkish Version of Scoliosis Research Society-22 (SRS-22) questionnaire. *Spine (Phila Pa 1976),* 30**,** 2464-8.

ALASSIRI, S. S., ALEISSA, S. I., ALHANDI, A. A., KONBAZ, F. M., ALHELAL, F., ABAALKHAIL, M., AL-ANNAIM, M. M., ALHABEEB, A. & ALSHEHRI, K. M. 2022. Prevalence and Predictors of Scoliosis and Back Pain in 591 Adolescents: A Randomized, Stratified, Cross-Sectional Study in Riyadh, Saudi Arabia. *Cureus,* 14**,** e26478.

ALMOAJIL, H., TOYE, F., DAWES, H., PIERCE, J., MEANEY, A., BAKLOUTI, A., POVERINI, L., HOPEWELL, S. & THEOLOGIS, T. 2022. Outcomes of importance to children and young adults with cerebral palsy, their parents and health professionals following lower limb orthopaedic surgery: A qualitative study to inform a Core Outcome Set. *Health Expect,* 25**,** 925-935.

ALTAF, F., GIBSON, A., DANNAWI, Z. & NOORDEEN, H. 2013. Adolescent idiopathic scoliosis. *The BMJ,* 346**,** f2508.

ALVES, V. L. D. S., ROBERTO, S. & OSMAR, A. 2015. Long-term impact of pre-operative physical rehabilitation protocol on the 6-min walk test of patients with adolescent idiopathic scoliosis: A randomized clinical trial. *Revista Portuguesa De Pneumologia*.

AMĂRICĂI, E., SUCIU, O., ONOFREI, R. R., MICLĂUȘ, R. S., IACOB, R. E., CAŢAN, L., POPOIU, C. M., CERBU, S. & BOIA, E. 2020. Respiratory function, functional capacity, and physical activity behaviours in children and adolescents with scoliosis. *J Int Med Res,* 48**,** 300060519895093.

AMINI, M., OEMRAWSINGH, A., VERWEIJ, L. M., LINGSMA, H. F., HAZELZET, J. A., EIJKENAAR, F. & VAN LEEUWEN, N. 2021. Facilitators and barriers for implementing patient-reported outcome measures in clinical care: An academic center's initial experience. *Health Policy,* 125**,** 1247-1255.

AN, J. K., BERMAN, D. & SCHULZ, J. 2023. Back pain in adolescent idiopathic scoliosis: A comprehensive review. *J Child Orthop,* 17**,** 126-140.

ANDERSEN, M., ANDERSEN, G. R., THOMSEN, K. & CHRISTENSEN, S. B. 2002. Early weaning might reduce the psychological strain of Boston bracing: a study of 136 patients with adolescent idiopathic scoliosis at 3.5 years after termination of brace treatment. *J Pediatr Orthop B,* 11**,** 96-9.

ANEKAR, A. A. & CASCELLA, M. 2021. WHO analgesic ladder. *StatPearls [Internet].* StatPearls Publishing.

ANTONARAKOS, P. D., KATRANITSA, L., ANGELIS, L., PAGANAS, A., KOEN, E. M., CHRISTODOULOU, E. A. & CHRISTODOULOU, A. G. 2009. Reliability and validity of the adapted Greek version of scoliosis research society - 22 (SRS-22) questionnaire. *Scoliosis,* 4**,** 14.

ANTUNES, B., RODRIGUES, P. P., HIGGINSON, I. J. & FERREIRA, P. L. 2018. Outcome measurement-a scoping review of the literature and future developments in palliative care clinical practice. *Ann Palliat Med,* 7**,** S196-s206.

ARBUCKLE, R., MARSHALL, C., GRANT, L., BURROWS, K., ALBRECHT, H. H. & SHEA, T. 2022. Development and content validity testing of patient-reported outcome (PRO) items to assess chest congestion associated with the common cold for use in children and adolescents. *J Patient Rep Outcomes,* 6**,** 56.

ARCHIBALD, M. M., AMBAGTSHEER, R. C., CASEY, M. G. & LAWLESS, M. 2019. Using Zoom Videoconferencing for Qualitative Data Collection: Perceptions and Experiences of Researchers and Participants. *International Journal of Qualitative Methods,* 18**,** 1609406919874596.

AROEIRA, R. M. C., DE LAS CASAS, E. B., PERTENCE, A. E. M., GRECO, M. & TAVARES, J. M. R. S. 2016. Non-invasive methods of computer vision in the posture evaluation of adolescent idiopathic scoliosis. *Journal of Bodywork and Movement Therapies,* 20**,** 832-843.

ASHER, M., LAI, S. M., BURTON, D. & MANNA, B. 2003a. The reliability and concurrent validity of the scoliosis research society-22 patient questionnaire for idiopathic scoliosis. *Spine,* 28**,** 63-69.

ASHER, M., LAI, S. M., BURTON, D. & MANNA, B. 2003b. Scoliosis research society-22 patient questionnaire: responsiveness to change associated with surgical treatment. *Spine,* 28**,** 70-73.

ASHER, M., LAI, S. M., BURTON, D. & MANNA, B. 2004. The Influence of Spine and Trunk Deformity on Preoperative Idiopathic Scoliosis Patients' Health-related Quality of Life Questionnaire Responses. *Spine,* 29**,** 861-868.

ASHER, M., MIN LAI, S., BURTON, D. & MANNA, B. 2003c. Discrimination Validity of the Scoliosis Research Society-22 Patient Questionnaire: Relationship to Idiopathic Scoliosis Curve Pattern and Curve Size. *Spine,* 28.

ASHER, M., MIN LAI, S., BURTON, D. & MANNA, B. 2003d. The reliability and concurrent validity of the scoliosis research society-22 patient questionnaire for idiopathic scoliosis. *Spine* 28**,** 63-9.

ASHER, M., MIN LAI, S., BURTON, D. & MANNA, B. 2003e. Scoliosis research society-22 patient questionnaire: responsiveness to change associated with surgical treatment. *Spine (Phila Pa 1976),* 28**,** 70-3.

ASHER, M. A., LAI, S. M., GLATTES, R. C., BURTON, D. C., ALANAY, A. & BAGO, J. 2006. Refinement of the SRS-22 Health-Related Quality of Life questionnaire Function domain. *Spine* 31**,** 593-7.

ASHER, M. A., MIN LAI, S. & BURTON, D. C. 2000. Further development and validation of the Scoliosis Research Society (SRS) outcomes instrument. *Spine (Phila Pa 1976),* 25**,** 2381-6.

BAGO, J., CLIMENT, J. M., EY, A., PEREZ-GRUESO, F. J. & IZQUIERDO, E. 2004. The Spanish version of the SRS-22 patient questionnaire for idiopathic scoliosis: transcultural adaptation and reliability analysis. *Spine,* 29**,** 1676-1680.

BAGÓ, J., CLIMENT, J. M., PÉREZ-GRUESO, F. J. & PELLISÉ, F. 2013. Outcome instruments to assess scoliosis surgery. *European Spine Journal,* 22**,** 195-202.

BAGO, J., PEREZ-GRUESO, F. J. S., PELLISE, F. & LES, E. 2012. How do idiopathic scoliosis patients who improve after surgery differ from those who do not exceed a minimum detectable change? *European Spine Journal,* 21**,** 50-56.

BAILEY, K. M., HOWARD, J. J., EL-HAWARY, R., CHORNEY, J. & GROUP, P. S. 2021. Pain trajectories following adolescent idiopathic scoliosis correction: analysis of predictors and functional outcomes. *JBJS Open Access,* 6**,** e20.

BAIRD, C. & GARDNER, A. 2021. A report of the number of adolescents screened as warranting further investigation for depression and social anxiety in a pre-operative cohort with idiopathic scoliosis. *Surgeon,* 19**,** 263-267.

BALDUS, C., BRIDWELL, K., HARRAST, J., SHAFFREY, C., ONDRA, S., LENKE, L., SCHWAB, F., MARDJETKO, S., GLASSMAN, S., EDWARDS, C., 2ND, LOWE, T., HORTON, W. & POLLY, D., JR. 2011. The Scoliosis Research Society Health-Related Quality of Life (SRS-30) age-gender normative data: an analysis of 1346 adult subjects unaffected by scoliosis. *Spine* 36**,** 1154-62.

BALSHEM, H., HELFAND, M., SCHÜNEMANN, H. J., OXMAN, A. D., KUNZ, R., BROZEK, J., VIST, G. E., FALCK-YTTER, Y., MEERPOHL, J., NORRIS, S. & GUYATT, G. H. 2011. GRADE guidelines: 3. Rating the quality of evidence. *J Clin Epidemiol,* 64**,** 401-6.

BARADARAN MAHDAVI, S., RIAHI, R., VAHDATPOUR, B. & KELISHADI, R. 2021. Association between sedentary behavior and low back pain; A systematic review and meta-analysis. *Health Promot Perspect,* 11**,** 393-410.

BASQUES, B. A., BOHL, D. D., GOLINVAUX, N. S., SMITH, B. G. & GRAUER, J. N. 2015. Patient factors are associated with poor short-term outcomes after posterior fusion for adolescent idiopathic scoliosis. *Clinical Orthopaedics & Related Research,* 473**,** 286-294.

BASTROM, T. P., BARTLEY, C., MARKS, M. C., YASZAY, B. & NEWTON, P. O. 2015. Postoperative Perfection: Ceiling Effects and Lack of Discrimination With Both SRS-22 and -24 Outcomes Instruments in Patients With Adolescent Idiopathic Scoliosis. *Spine (Phila Pa 1976),* 40**,** E1323-E1329.

BASTROM, T. P., MARKS, M. C., YASZAY, B., NEWTON, P. O. & GROUP, H. S. 2013. Prevalence of postoperative pain in adolescent idiopathic scoliosis and the association with preoperative pain. *Spine,* 38**,** 1848-1852.

BEAN, J. F., OLVECZKY, D. D., KIELY, D. K., LAROSE, S. I. & JETTE, A. M. 2011. Performance-based versus patient-reported physical function: what are the underlying predictors? *Phys Ther,* 91**,** 1804-11.

BEAUDART, C., ROLLAND, Y., CRUZ-JENTOFT, A. J., BAUER, J. M., SIEBER, C., COOPER, C., AL-DAGHRI, N., ARAUJO DE CARVALHO, I., BAUTMANS, I. & BERNABEI, R. 2019. Assessment of muscle function and physical performance in daily clinical practice. *Calcified tissue international,* 105**,** 1-14.

BEAUSEJOUR, M., JONCAS, J., GOULET, L., ROY-BEAUDRY, M., PARENT, S., GRIMARD, G., FORCIER, M., LAURIAULT, S. & LABELLE, H. 2009. Reliability and validity of adapted French Canadian version of Scoliosis Research Society Outcomes Questionnaire (SRS-22) in Quebec. *Spine,* 34**,** 623-628.

BEIGHLEY, A., ZHANG, A., HUANG, B., CARR, C., MATHKOUR, M., WERNER, C., SCULLEN, T., KILGORE, M. D., MAULUCCI, C. M., DALLAPIAZZA, R. F. & KALYVAS, J. 2022. Patient-reported outcome measures in spine surgery: A systematic review. *J Craniovertebr Junction Spine,* 13**,** 378-389.

BELUR, J., TOMPSON, L., THORNTON, A. & SIMON, M. 2018. Interrater Reliability in Systematic Review Methodology: Exploring Variation in Coder Decision-Making. *Sociological Methods & Research,* 50**,** 837-865.

BERDISHEVSKY, H., LEBEL, V. A., BETTANY-SALTIKOV, J., RIGO, M., LEBEL, A., HENNES, A., ROMANO, M., BIAŁEK, M., M'HANGO, A., BETTS, T., DE MAUROY, J. C. & DURMALA, J. 2016. Physiotherapy scoliosis-specific exercises – a comprehensive review of seven major schools. *Scoliosis and Spinal Disorders,* 11**,** 20.

BERLINER, J. L., VERMA, K., LONNER, B. S., PENN, P. U. & BHARUCHA, N. J. 2013. Discriminative validity of the Scoliosis Research Society 22 questionnaire among five curve-severity subgroups of adolescents with idiopathic scoliosis. *Spine Journal: Official Journal of the North American Spine Society,* 13**,** 127-133.

BERTUCCELLI, M., CANTELE, F. & MASIERO, S. 2022. Body Image and Body Schema in Adolescents with Idiopathic Scoliosis: A Scoping Review. *Adolescent Research Review*.

BETTANY-SALTIKOV, J., WEISS, H.-R., CHOCKALINGAM, N., KANDASAMY, G. & ARNELL, T. 2016. A comparison of patient-reported outcome measures following different treatment approaches for adolescents with severe idiopathic scoliosis: A systematic review. *Asian Spine Journal,* 10**,** 1170.

BOUTON, D., FEDORAK, G., OEFFINGER, D. J., GUPTA, P., LUHMANN, S., STASIKELIS, P., SZCZODRY, M., TALWALKAR, V. & HUNG, M. 2022. Baseline patient reported outcome measurement information system (PROMIS) scores in children with idiopathic scoliosis and their relation to the SRS-22. *Spine Deform,* 10**,** 63-68.

BRAUN, V. & CLARKE, V. 2006. Using thematic analysis in psychology. *Qual Res Psychol,* 3**,** 77-101.

BRAZIER, J. E., HARPER, R., JONES, N. M., O'CATHAIN, A., THOMAS, K. J., USHERWOOD, T. & WESTLAKE, L. 1992. Validating the SF-36 health survey questionnaire: new outcome measure for primary care. *Bmj,* 305**,** 160-4.

BRÉDART, A., MARREL, A., ABETZ-WEBB, L., LASCH, K. & ACQUADRO, C. 2014. Interviewing to develop Patient-Reported Outcome (PRO) measures for clinical research: eliciting patients' experience. *Health Qual Life Outcomes,* 12**,** 15.

BREWER, P., BERRYMAN, F., BAKER, D., PYNSENT, P. & GARDNER, A. 2014. Analysis of the Scoliosis Research Society-22 Questionnaire Scores: Is There a Difference Between a Child and Parent and Does Physician Review Change That? *Spine Deform,* 2**,** 34-39.

BRIGGS, M. S., RETHMAN, K. K., CROOKES, J., CHEEK, F., POTTKOTTER, K., MCGRATH, S., DEWITT, J., HARMON-MATTHEWS, L. E. & QUATMAN-YATES, C. C. 2020. Implementing Patient-Reported Outcome Measures in Outpatient Rehabilitation Settings: A

Systematic Review of Facilitators and Barriers Using the Consolidated Framework for Implementation Research. *Archives of Physical Medicine and Rehabilitation,* 101**,** 1796-1812.

BROD, M., TESLER, L. E. & CHRISTENSEN, T. L. 2009. Qualitative research and content validity: developing best practices based on science and experience. *Qual Life Res,* 18**,** 1263-78.

BULL, J. & GROGAN, S. 2010. Children having spinal surgery to correct scoliosis: a qualitative study of parents' experiences. *Journal of Health Psychology,* 15**,** 299-309.

BURWELL, R. G. 2003. Aetiology of idiopathic scoliosis: current concepts. *Pediatric Rehabilitation,* 6**,** 137-170.

CAMARINI, P. M., ROSANOVA, G. C., GABRIEL, B. S., GIANINI, P. E. & OLIVEIRA, A. S. 2013. The Brazilian version of the SRS-22r questionnaire for idiopathic scoliosis. *Brazilian Journal of Physical Therapy,* 17**,** 494-505.

CANTELE, F., MAGHINI, I., TONELLATO, M., MENEGUZZO, P., FAVARO, A. & MASIERO, S. 2021. An Analysis of Eating Disorders in Adolescent Idiopathic Scoliosis: A Prospective Cross-sectional Study in a Female Population. *Spine,* 46.

CARRASCO, M. I. B. & RUIZ, M. C. S. 2014. Perceived self-image in adolescent idiopathic scoliosis: an integrative review of the literature. *Revista da Escola de Enfermagem da USP,* 48**,** 748-757.

CARRASCO, M. I. B. & RUIZ, M. C. S. 2016. Experiences of young diagnosed with idiopathic scoliosis. *Enfermeria Global,* 15**,** 37-50.

CARREON, L. Y., SANDERS, J. O., DIAB, M., SUCATO, D. J., STURM, P. F. & GLASSMAN, S. D. 2010. The minimum clinically important difference in Scoliosis Research Society-22 Appearance, Activity, And Pain domains after surgical correction of adolescent idiopathic scoliosis. *Spine (Phila Pa 1976),* 35**,** 2079-83.

CARRICO, G., MEVES, R. & AVANZI, O. 2012. Cross-cultural adaptation and validity of an adapted Brazilian Portuguese version of Scoliosis Research Society-30 questionnaire. *Spine (Phila Pa 1976),* 37**,** E60-3.

CARTER, S. M., SHIH, P., WILLIAMS, J., DEGELING, C. & MOONEY-SOMERS, J. 2021. Conducting Qualitative Research Online: Challenges and Solutions. *The Patient - Patient-Centered Outcomes Research,* 14**,** 711-718.

CHALMERS, I. & GLASZIOU, P. 2009. Avoidable waste in the production and reporting of research evidence. *Lancet,* 374**,** 86-9.

CHEN, A. F., BI, W., SINGHABAHU, D., LONDINO, J., HOHL, J., WARD, M. & WARD, W. T. 2013. Converting Scoliosis Research Society-24 to Scoliosis Research Society-22r in a Surgical-Range, Medical/Interventional Adolescent Idiopathic Scoliosis Patient Cohort. *Spine Deform,* 1**,** 108-114.

CHEUNG, K. M., SENKOYLU, A., ALANAY, A., GENC, Y., LAU, S. & LUK, K. D. 2007. Reliability and concurrent validity of the adapted Chinese version of Scoliosis Research Society-22 (SRS-22) questionnaire. *Spine,* 32**,** 1141-1145.

CHIAROTTO, A., DEYO, R. A., TERWEE, C. B., BOERS, M., BUCHBINDER, R., CORBIN, T. P., COSTA, L. O., FOSTER, N. E., GROTLE, M., KOES, B. W., KOVACS, F. M., LIN, C. W., MAHER, C. G., PEARSON, A. M., PEUL, W. C., SCHOENE, M. L., TURK, D. C.,

VAN TULDER, M. W. & OSTELO, R. W. 2015. Core outcome domains for clinical trials in non-specific low back pain. *Eur Spine J,* 24**,** 1127-42.

CLIMENT, J. M., BAGO, J., EY, A., PEREZ-GRUESO, F. J. S. & IZQUIERDO, E. 2005. Validity of the Spanish version of the Scoliosis Research Society-22 (SRS-22) Patient Questionnaire. *Spine (03622436),* 30**,** 705-709.

COBB, J. 1948. Outline for the study of scoliosis. *Instr Course Lect AAOS,* 5**,** 261-275.

COHEN, J. 1960. A Coefficient of Agreement for Nominal Scales. *Educational and Psychological Measurement,* 20**,** 37-46.

COOMBES, L., BRISTOWE, K., ELLIS-SMITH, C., AWORINDE, J., FRASER, L. K., DOWNING, J., BLUEBOND-LANGNER, M., CHAMBERS, L., MURTAGH, F. E. M. & HARDING, R. 2021. Enhancing validity, reliability and participation in self-reported health outcome measurement for children and young people: a systematic review of recall period, response scale format, and administration modality. *Quality of Life Research,* 30**,** 1803-1832.

CORTES PAREDES, S., BARRIOS PITARQUE, C., ESCRIVA PEIRO, M. D., BENET COBO DEL PRADO, I., BURGOS FLORES, J., HEVIA SIERRA, E., PIZA VALLESPIR, G. & DOMENECH FERNANDEZ, P. 2011. Self-image, self-esteem, and eating behavior in adolescents with idiopathic scoliosis: Correlation with the SRS-22 clinical and functional domains. *European Spine Journal,* 20 (11)**,** 2092-2092.

CRESWELL, J. W. & POTH, C. N. 2016. *Qualitative inquiry and research design: Choosing among five approaches*, Sage publications.

CUNNINGHAM, G., WRIGHT, D., NNADI, C. & KIESER, D. 2020. Patient Outcome Questionnaires in the British Spine Registry: Why are Response Rates Low and Which Patients Groups are responding. *J Spine Neurosurg 9,* 1.

D'ANDREA, L. P., BETZ, R. R., LENKE, L. G., CLEMENTS, D. H., LOWE, T. G., MEROLA, A., HAHER, T., HARMS, J., HUSS, G. K., BLANKE, K. & MCGLOTHLEN, S. 2000. Do Radiographic Parameters Correlate With Clinical Outcomes in Adolescent Idiopathic Scoliosis? *Spine,* 25**,** 1795-1802.

DANIELSSON, A. J. & NACHEMSON, A. L. 2003. Back pain and function 23 years after fusion for adolescent idiopathic scoliosis: a case-control study-part II. *Spine (Phila Pa 1976),* 28**,** E373-83.

DANIELSSON, A. J. & ROMBERG, K. 2013. Reliability and Validity of the Swedish Version of the Scoliosis Research Society–22 (SRS-22r) Patient Questionnaire for Idiopathic Scoliosis. *Spine,* 38**,** 1875-1884.

DANIELSSON, A. J., ROMBERG, K. & NACHEMSON, A. L. 2006. Spinal Range of Motion, Muscle Endurance, and Back Pain and Function at Least 20 Years After Fusion or Brace Treatment for Adolescent Idiopathic Scoliosis: A Case-Control Study. *Spine,* 31**,** 275-283.

DANIELSSON, A. J., WIKLUND, I., PEHRSSON, K. & NACHEMSON, A. L. 2001. Health-related quality of life in patients with adolescent idiopathic scoliosis: a matched follow-up at least 20 years after treatment with brace or surgery. *European Spine Journal,* 10**,** 278-288.

DAVID, L. 2017. Evidence-Based Treatment of Adolescent Idiopathic Scoliosis.

DE BAAT, P., VAN BIEZEN, F. C. & DE BAAT, C. 2012. [Scoliosis: review of types, aetiology, diagnostics, and treatment 2]. *Nederlands Tijdschrift voor Tandheelkunde,* 119**,** 531-535.

DE KLEUVER, M., FARAJ, S. S. A., HOLEWIJN, R. M., GERMSCHEID, N. M., ADOBOR, R. D., ANDERSEN, M., TROPP, H., DAHL, B., KESKINEN, H., OLAI, A., POLLY, D. W., VAN HOOFF, M. L. & HAANSTRA, T. M. 2017. Defining a core outcome set for adolescent and young adult patients with a spinal deformity. *Acta Orthop,* 88**,** 612-618.

DE VET, H. C., TERWEE, C. B., MOKKINK, L. B. & KNOL, D. L. 2011. *Measurement in medicine: a practical guide*, Cambridge university press.

DEMERS, M., BLANCHETTE, A. K., MULLICK, A. A., SHAH, A., WOO, K., SOLOMON, J. & LEVIN, M. F. 2019. Facilitators and barriers to using neurological outcome measures in developed and developing countries. *Physiother Res Int,* 24**,** e1756.

DETTORI, J. R. 2011. Loss to follow-up. *Evid Based Spine Care J,* 2**,** 7-10.

DJURASOVIC, M., GLASSMAN, S. D., SUCATO, D. J., LENKE, L. G. & CARREON, L. Y. 2016. Improvement in SRS-22R pain scores after surgery for adolescent idiopathic scoliosis. *The Spine Journal,* 16**,** S343-S344.

DODD, S., CLARKE, M., BECKER, L., MAVERGAMES, C., FISH, R. & WILLIAMSON, P. R. 2018. A taxonomy has been developed for outcomes in medical research to help improve knowledge discovery. *J Clin Epidemiol,* 96**,** 84-92.

DONNELLY, M. J., DOLAN, L. A., GRANDE, L. & WEINSTEIN, S. L. 2004. Patient and parent perspectives on treatment for adolescent idiopathic scoliosis. *Iowa Orthopaedic Journal,* 24**,** 76-83.

DU, C., YU, J., ZHANG, J., JIANG, J., LAI, H., LIU, W., LIU, Y., LI, H. & WANG, P. 2016a. Relevant areas of functioning in patients with adolescent idiopathic scoliosis on the International Classification of Functioning, Disability and Health: The patients' perspective. *J Rehabil Med,* 48**,** 806-814.

DU, Q., ZHOU, X., NEGRINI, S., CHEN, N., YANG, X., LIANG, J. & SUN, K. 2016b. Scoliosis epidemiology is not similar all over the world: a study from a scoliosis school screening on Chongming Island (China). *BMC Musculoskeletal Disorders,* 17**,** 303.

DUNCAN, E. A. S. & MURRAY, J. 2012. The barriers and facilitators to routine outcome measurement by allied health professionals in practice: a systematic review. *BMC Health Services Research,* 12**,** 96.

EMANUELSON, U. & EGENVALL, A. 2014. The data–Sources and validation. *Preventive veterinary medicine,* 113**,** 298-303.

EPSTEIN, R. M. & STREET, R. L., JR. 2011. The values and value of patient-centered care. *Ann Fam Med,* 9**,** 100-3.

ESSEX, R., BRUCE, G., DIBLEY, M., NEWTON, P., THOMPSON, T., SWAINE, I. & DIBLEY, L. 2022. A systematic scoping review and textual narrative synthesis of the qualitative evidence related to adolescent idiopathic scoliosis. *International Journal of Orthopaedic and Trauma Nursing,* 45**,** 100921.

ETIKAN, I. 2016. Comparison of Convenience Sampling and Purposive Sampling. *American Journal of Theoretical and Applied Statistics,* 5**,** 1.

EXCELLENCE, N. C. F. H. 2007. How to change practice.

EYVAZOV, K., SAMARTZIS, D. & CHEUNG, J. P. Y. 2017. The association of lumbar curve magnitude and spinal range of motion in adolescent idiopathic scoliosis: a cross-sectional study. *BMC musculoskeletal disorders,* 18**,** 51-51.

FABRICANT, P. D., ADMONI, S.-H., GREEN, D. W., IPP, L. S. & WIDMANN, R. F. 2012. Return to athletic activity after posterior spinal fusion for adolescent idiopathic scoliosis: analysis of independent predictors. *Journal of Pediatric Orthopaedics,* 32**,** 259-265.

FALAVIGNA, A., DOZZA, D. C., TELES, A. R., WONG, C. C., BARBAGALLO, G., BRODKE, D., AL-MUTAIR, A., GHOGAWALA, Z. & RIEW, K. D. 2017. Current Status of Worldwide Use of Patient-Reported Outcome Measures (PROMs) in Spine Care. *World Neurosurg,* 108**,** 328-335.

FEDORAK, G. T., LARKIN, K., HEFLIN, J. A., XU, J. & HUNG, M. 2019. Pediatric Patient-Reported Outcomes Measurement Information System is Equivalent to Scoliosis Research Society-22 in Assessing Health Status in Adolescent Idiopathic Scoliosis. *Spine,* 44**,** E1206-E1210.

FEISE, R. J., DONALDSON, S., CROWTHER, E. R., MENKE, J. M. & WRIGHT, J. G. 2005. Construction and validation of the scoliosis quality of life index in adolescent idiopathic scoliosis. *Spine (Phila Pa 1976),* 30**,** 1310-5.

FERGUSON, S. A., MARRAS, W. S. & BURR, D. L. 2005. Differences Among Outcome Measures in Occupational Low Back Pain. *Journal of Occupational Rehabilitation,* 15**,** 329-341.

FERNANDES, P., DO BRITO, J. S., FLORES, I. & MONTEIRO, J. 2019. Impact of surgery on the quality of life of adolescent idiopathic scoliosis. *The Iowa Orthopaedic Journal,* 39**,** 66.

FLEISCHMANN, M. & VAUGHAN, B. 2018. The challenges and opportunities of using patient reported outcome measures (PROMs) in clinical practice. *International Journal of Osteopathic Medicine,* 28**,** 56-61.

FLETCHER, D., STAMER, U. M., POGATZKI-ZAHN, E., ZASLANSKY, R., TANASE, N. V., PERRUCHOUD, C., KRANKE, P., KOMANN, M., LEHMAN, T. & MEISSNER, W. 2015. Chronic postsurgical pain in Europe: An observational study. *Eur J Anaesthesiol,* 32**,** 725-34.

FONG, D. Y., LEE, C. F., CHEUNG, K. M., CHENG, J. C., NG, B. K., LAM, T. P., MAK, K. H., YIP, P. S. & LUK, K. D. 2010. A meta-analysis of the clinical effectiveness of school scoliosis screening. *Spine (Phila Pa 1976),* 35**,** 1061-71.

FOSTER, A., CROOT, L., BRAZIER, J., HARRIS, J. & O'CATHAIN, A. 2018. The facilitators and barriers to implementing patient reported outcome measures in organisations delivering health related services: a systematic review of reviews. *J Patient Rep Outcomes,* 2**,** 46.

GALLANT, J. N., MORGAN, C. D., STOKLOSA, J. B., GANNON, S. R., SHANNON, C. N. & BONFIELD, C. M. 2018. Psychosocial Difficulties in Adolescent Idiopathic Scoliosis: Body Image, Eating Behaviors, and Mood Disorders. *World Neurosurgery,* 116**,** 421-432.e1.

GAO, C.-C., CHERN, J.-S., CHANG, C.-J., LAI, P.-L. & LUNG, C.-W. 2019. Center of pressure progression patterns during level walking in adolescents with idiopathic scoliosis. *PLOS ONE,* 14**,** e0212161.

GILMORE, S. J., HAHNE, A. J., DAVIDSON, M. & MCCLELLAND, J. A. 2019. Predictors of substantial improvement in physical function six months after lumbar surgery: is early post-

operative walking important? A prospective cohort study. *BMC Musculoskeletal Disorders,* 20**,** 418.

GLATTES, R. C., BURTON, D. C., LAI, S. M., FRASIER, E. & ASHER, M. A. 2007. The reliability and concurrent validity of the Scoliosis Research Society-22r patient questionnaire compared with the Child Health Questionnaire-CF87 patient questionnaire for adolescent spinal deformity. *Spine,* 32**,** 1778-84.

GLOWACKI, M., MISTERSKA, E., LAURENTOWSKA, M. & MANKOWSKI, P. 2009. Polish adaptation of scoliosis research society-22 questionnaire. *Spine (Phila Pa 1976),* 34**,** 1060-5.

GRIVAS, T. B., NEGRINI, S., AUBIN, C. E., AULISA, A. G., DE MAUROY, J. C., DONZELLI, S., HRESKO, M. T., KOTWICKI, T., LOU, E., MARUYAMA, T., PARENT, E. C., RIGO, M., THOMETZ, J. G., WONG, M. S. & ZAINA, F. 2022. Nonoperative management of adolescent idiopathic scoliosis (AIS) using braces. *Prosthet Orthot Int,* 46**,** 383-391.

GRIVAS, T. B., VASILIADIS, E., MOUZAKIS, V., MIHAS, C. & KOUFOPOULOS, G. 2006. Association between adolescent idiopathic scoliosis prevalence and age at menarche in different geographic latitudes. *Scoliosis,* 1**,** 9.

GROENEWALD, C. B., THAM, S. W. & PALERMO, T. M. 2020. Impaired School Functioning in Children With Chronic Pain: A National Perspective. *Clinical Journal of Pain,* 36**,** 693-699.

HAHER, T. R., GORUP, J. M., SHIN, T. M., HOMEL, P., MEROLA, A. A., GROGAN, D. P., PUGH, L., LOWE, T. G. & MURRAY, M. 1999. Results of the Scoliosis Research Society instrument for evaluation of surgical outcome in adolescent idiopathic scoliosis: a multicenter study of 244 patients. *Spine,* 24**,** 1435.

HAIDAR, R. K., KASSAK, K., MASROUHA, K., IBRAHIM, K. & MHAIDLI, H. 2015. Reliability and validity of an adapted Arabic version of the Scoliosis Research Society-22r Questionnaire. *Spine (Phila Pa 1976),* 40**,** E971-7.

HARVEY, P. D., VELLIGAN, D. I. & BELLACK, A. S. 2007. Performance-based measures of functional skills: usefulness in clinical treatment studies. *Schizophr Bull,* 33**,** 1138-48.

HASHIMOTO, H., SASE, T., ARAI, Y., MARUYAMA, T., ISOBE, K. & SHOUNO, Y. 2007. Validation of a Japanese version of the Scoliosis Research Society-22 Patient Questionnaire among idiopathic scoliosis patients in Japan. *Spine (Phila Pa 1976),* 32**,** E141-6.

HAYNES, S. N., RICHARD, D. & KUBANY, E. S. 1995. Content validity in psychological assessment: A functional approach to concepts and methods. *Psychol Assess,* 7**,** 238.

HAYS, R. D. & WOOLLEY, J. M. 2000. The concept of clinically meaningful difference in health-related quality-of-life research. *Pharmacoeconomics,* 18**,** 419-423.

HE, C. & WONG, M. S. 2018. Spinal Flexibility Assessment on the Patients With Adolescent Idiopathic Scoliosis: A Literature Review. *Spine (Phila Pa 1976),* 43**,** E250-e258.

HEALTH, U. D. O., EVALUATION, H. S. F. C. F. D., GOV, R. L. B. F. H., HEALTH, U. D. O., EVALUATION, H. S. F. C. F. B., GOV, R. T. S. F. H., HEALTH, U. D. O., DEVICES, H. S. F. C. F. & GOV, R. H. S. C. F. 2006. Guidance for industry: patient-reported outcome measures: use in medical product development to support labeling claims: draft guidance. *Health and Quality of Life Outcomes,* 4**,** 79.

HELENIUS, L., DIARBAKERLI, E., GRAUERS, A., LASTIKKA, M., OKSANEN, H., PAJULO, O., LÖYTTYNIEMI, E., MANNER, T., GERDHEM, P. & HELENIUS, I. 2019. Back pain

and quality of life after surgical treatment for adolescent idiopathic scoliosis at 5-year follow-up: comparison with healthy controls and patients with untreated idiopathic scoliosis. *JBJS,* 101**,** 1460-1466.

HOLMAN, H. R. 1993. Qualitative inquiry in medical research. *Journal of Clinical Epidemiology,* 46**,** 29-36.

HOLROYD, A. 2007. Interpretive Hermeneutic Phenomenology: Clarifying Understanding. *Indo-Pacific Journal of Phenomenology,* 7.

HONEYMAN, C. & DAVISON, J. 2016. Patients' experience of adolescent idiopathic scoliosis surgery: a phenomenological analysis. *Nurs Child Young People,* 28**,** 29-36.

HORNE, J. P., FLANNERY, R. & USMAN, S. 2014. Adolescent idiopathic scoliosis: diagnosis and management. *Am Fam Physician,* 89**,** 193-8.

HORNG, M. H., KUOK, C. P., FU, M. J., LIN, C. J. & SUN, Y. N. 2019. Cobb Angle Measurement of Spine from X-Ray Images Using Convolutional Neural Network. *Comput Math Methods Med,* 2019**,** 6357171.

HOWE, C. J., COLE, S. R., LAU, B., NAPRAVNIK, S. & ERON, J. J., JR. 2016a. Selection Bias Due to Loss to Follow Up in Cohort Studies. *Epidemiology (Cambridge, Mass.),* 27**,** 91-97.

HOWE, C. J., COLE, S. R., LAU, B., NAPRAVNIK, S. & ERON JR, J. J. 2016b. Selection bias due to loss to follow up in cohort studies. *Epidemiology (Cambridge, Mass.),* 27**,** 91.

HRESKO, M. T., MESIHA, M., RICHARDS, K. & ZURAKOWSKI, D. 2006. A comparison of methods for measuring spinal motion in female patients with adolescent idiopathic scoliosis. *J Pediatr Orthop,* 26**,** 758-63.

HUANG, N. Q., GUO, H. S., LIU, J., HUANG, G. X., YANG, X. H., CHEN, J. & SU, P. Q. 2011. [A survey on adolescent scoliosis in Guangzhou]. *Zhonghua Liu Xing Bing Xue Za Zhi,* 32**,** 138-41.

HUGHES, J., YASZAY, B., BASTROM, T. P., BARTLEY, C. E., PARENT, S., CAHILL, P. J., LONNER, B., SHAH, S. A., SAMDANI, A. & NEWTON, P. O. 2021. Long-term Patient Perception Following Surgery for Adolescent Idiopathic Scoliosis if Dissatisfied at 2-year Follow-up. *Spine,* 46**,** 507-511.

HUUS, K., SCHLEBUSCH, L., RAMAAHLO, M., SAMUELS, A., BERGLUND, I. G. & DADA, S. 2021. Barriers and facilitators to participation for children and adolescents with disabilities in low- and middle-income countries - A scoping review. *Afr J Disabil,* 10**,** 771.

ILLEEZ, O. G., AKPINAR, P., BAHADIR ULGER, F. E., OZKAN, F. U. & AKTAS, I. 2020. Low back pain in children and adolescents: Real life experience of 106 patients. *North Clin Istanb,* 7**,** 603-608.

JADA, A., MACKEL, C. E., HWANG, S. W., SAMDANI, A. F., STEPHEN, J. H., BENNETT, J. T. & BAAJ, A. A. 2017. Evaluation and management of adolescent idiopathic scoliosis: a review. *Neurosurgical focus,* 43**,** E2.

JAMES, W., IAN, S. & CHRIS, G. 2018. Core Outcome Sets. *Archives of disease in childhood - Education & practice edition,* 103**,** 163.

JANICKI, J. A. & ALMAN, B. 2007. Scoliosis: Review of diagnosis and treatment. *Paediatr Child Health,* 12**,** 771-6.

JASANI, V., BALIGA, S. & AHMAD, S. 2016. Non-compliance for email responses on the British Spine Registry (BSR). *The Spine Journal,* 16**,** S80.

JAYADEVAPPA, R. & CHHATRE, S. 2011. Patient centered care-a conceptual model and review of the state of the art. *The Open Health Services and Policy Journal,* 4.

JOARDER, I., TANIGUCHI, S., MENDOZA, A. & SNOW, M. E. 2023. Defining "successful" treatment outcomes in adolescent idiopathic scoliosis: a scoping review. *Eur Spine J,* 32**,** 1204-1244.

JOHNSON, J. L., ADKINS, D. & CHAUVIN, S. 2020. A Review of the Quality Indicators of Rigor in Qualitative Research. *Am J Pharm Educ,* 84**,** 7120.

KAELIN, A. J. 2020. Adolescent idiopathic scoliosis: indications for bracing and conservative treatments. *Ann Transl Med,* 8**,** 28.

KAKAR, R. S., SIMPSON, K. J., DAS, B. M. & BROWN, C. N. 2017. Review of physical activity benefits and potential considerations for individuals with surgical fusion of spine for scoliosis. *International journal of exercise science,* 10**,** 166.

KARIN, R. & KARIN, R. 2019. Spinal mobility, muscle strength and function in patients with idiopathic scoliosis. Different aspects on long term outcome.

KAUSHIK, V. & WALSH, C. A. 2019. Pragmatism as a Research Paradigm and Its Implications for Social Work Research. *Social Sciences,* 8**,** 255.

KELLY, M. P., KIM, H. J., AMES, C. P., BURTON, D. C., CARREON, L. Y., POLLY JR, D. W., HOSTIN, R., JAIN, A., GUM, J. L. & LAFAGE, V. 2018. Minimum detectable measurement difference for health-related quality of life measures varies with age and disability in adult spinal deformity: implications for calculating minimal clinically important difference. *Spine,* 43**,** E790-E795.

KELLY, M. P., LENKE, L. G., SPONSELLER, P. D., PAHYS, J. M., BASTROM, T. P., LONNER, B. S. & ABEL, M. F. 2019. The minimum detectable measurement difference for the Scoliosis Research Society-22r in adolescent idiopathic scoliosis: a comparison with the minimum clinically important difference. *The Spine Journal,* 19**,** 1319-1323.

KERR, C., NIXON, A. & WILD, D. 2010. Assessing and demonstrating data saturation in qualitative inquiry supporting patient-reported outcomes research. *Expert Rev Pharmacoecon Outcomes Res,* 10**,** 269-281.

KIESER, D., CUNNINGHAM, G., WRIGHT, D. & NNADI, C. 2020. Patient outcome questionnaires in the British Spine Registry: Why are response rates low and which patient groups are responding. *Journal of Spine & Neurosurgery,* 9.

KIESER, D. C., YUKSEL, S., BOISSIERE, L., YILGOR, C., CAWLEY, D. T., HAYASHI, K., ALANAY, A., KLEINSTUECK, F. S., PELLISE, F., PEREZ-GRUESO, F. J. S., JEAN-MARC, V., BOURGHLI, A., ACAROGLU, E. R., OBEID, I. & THE EUROPEAN SPINE STUDY, G. 2022. Impact of radiologic variables on item responses of ODI, SRS22 and SF-36. in adult spinal deformity patients: differential item functioning (DIF) analysis results from a multi-center database. *European Spine Journal,* 31**,** 1166-1173.

KIM, H. Y. 2019. Statistical notes for clinical researchers: the independent samples t-test. *Restor Dent Endod,* 44**,** e26.

KLEINBERG, S. 1922. THE OPERATIVE TREATMENT OF SCOLIOSIS. *Archives of Surgery,* 5**,** 631-645.

KNAFL, K., DEATRICK, J., GALLO, A., HOLCOMBE, G., BAKITAS, M., DIXON, J. & GREY, M. 2007. The analysis and interpretation of cognitive interviews for instrument development. *Research in Nursing & Health,* 30**,** 224-234.

KONIECZNY, M. R., SENYURT, H. & KRAUSPE, R. 2013. Epidemiology of adolescent idiopathic scoliosis. *J Child Orthop,* 7**,** 3-9.

KORDI, R. & ROSTAMI, M. 2011. Low back pain in children and adolescents: an algorithmic clinical approach. *Iran J Pediatr,* 21**,** 259-70.

KOUWENHOVEN, J. W. & CASTELEIN, R. M. 2008. The pathogenesis of adolescent idiopathic scoliosis: review of the literature. *Spine (Phila Pa 1976),* 33**,** 2898-908.

KROMAN, S. L., ROOS, E. M., BENNELL, K. L., HINMAN, R. S. & DOBSON, F. 2014. Measurement properties of performance-based outcome measures to assess physical function in young and middle-aged people known to be at high risk of hip and/or knee osteoarthritis: a systematic review. *Osteoarthritis and cartilage,* 22**,** 26-39.

KUHN, J. E. 2016. Why Measure Outcomes? *Instr Course Lect,* 65**,** 583-6.

KYROLA, K., JARVENPAA, S., YLINEN, J., MECKLIN, J. P., REPO, J. P. & HAKKINEN, A. 2017. Reliability and Validity Study of the Finnish Adaptation of Scoliosis Research Society Questionnaire Version SRS-30. *Spine (Phila Pa 1976),* 42**,** 943-949.

LAMONTAGNE, L., HEPWORTH, J. T., SALISBURY, M. H. & COHEN, F. 2003. Effects of coping instruction in reducing young adolescents' pain after major spinal surgery. *Orthopaedic Nursing,* 22**,** 398-403.

LAMONTAGNE, L. L., HEPWORTH, J. T., COHEN, F. & SALISBURY, M. H. 2004. Adolescent scoliosis: Effects of corrective surgery, cognitive-behavioral interventions, and age on activity outcomes. *Applied Nursing Research,* 17**,** 168-177.

LANDRAF, J., FAYERS, P. & HAYS, R. 2005. Practical considerations in the measurement of HRQoL in child/adolescent clinical trials. *Assessing quality of life in clinical trials: methods and practice,* 339**,** 367.

LANGENSIEPEN, S., SEMLER, O., SOBOTTKE, R., FRICKE, O., FRANKLIN, J., SCHONAU, E. & EYSEL, P. 2013. Measuring procedures to determine the Cobb angle in idiopathic scoliosis: a systematic review. *Eur Spine J,* 22**,** 2360-71.

LASCH, K., MARQUIS, P., VIGNEUX, M., ABETZ-WEBB, L., ARNOULD, B., BAYLISS, M., CRAWFORD, B. & ROSA, K. 2010. PRO development: Rigorous qualitative research as the crucial foundation. *Quality of life research : an international journal of quality of life aspects of treatment, care and rehabilitation,* 19**,** 1087-96.

LEE, A. C., FEGER, M. A., SINGLA, A. & ABEL, M. F. 2016. Effect of surgical approach on pulmonary function in adolescent idiopathic scoliosis patients: a systemic review and meta-analysis. *Spine,* 41**,** E1343-E1355.

LEE, H., CHOI, J., HWANG, J. H. & PARK, J. H. 2017. Psychometric evaluation of the Scoliosis Research Society-22 Revised questionnaire among adolescents with idiopathic scoliosis. *Journal of Pediatric Orthopaedics, Part B,* 26**,** 59-63.

LEE, J. S., LEE, D. H., SUH, K. T., KIM, J. I., LIM, J. M. & GOH, T. S. 2011. Validation of the Korean version of the Scoliosis Research Society-22 questionnaire. *European Spine Journal,* 20**,** 1751-1756.

LEELAPATTANA, P., KEOROCHANA, G., JOHNSON, J., WAJANAVISIT, W. & LAOHACHAROENSOMBAT, W. 2011. Reliability and validity of an adapted Thai version of the Scoliosis Research Society-22 questionnaire. *J Child Orthop,* 5**,** 35-40.

LEITE, M. N., KAMPER, S. J., BRODERICK, C. & YAMATO, T. P. 2022. What Works When Treating Children and Adolescents With Low Back Pain? *Journal of Orthopaedic & Sports Physical Therapy,* 52**,** 419-424.

LENKE, L. G., BETZ, R. R., HARMS, J., BRIDWELL, K. H., CLEMENTS, D. H., LOWE, T. G. & BLANKE, K. 2001. Adolescent idiopathic scoliosis: a new classification to determine extent of spinal arthrodesis. *JBJS,* 83**,** 1169-1181.

LENSSINCK, M.-L., FRIJLINK, A. C., BERGER, M. Y., BIERMA - ZEINSTRA, S., VERKERK, K. & VERHAGEN, A. 2005. Effect of Bracing and Other Conservative Interventions in the Treatment of Idiopathic Scoliosis in Adolescents: A Systematic Review of Clinical Trials. *Phys Ther,* 85**,** 1329-1339.

LERMAN, J. A., SULLIVAN, E. & HAYNES, R. J. 2002. The Pediatric Outcomes Data Collection Instrument (PODCI) and functional assessment in patients with adolescent or juvenile idiopathic scoliosis and congenital scoliosis or kyphosis. *Spine (Phila Pa 1976),* 27**,** 2052-7; discussion 2057-8.

LESZCZEWSKA, J., CZAPROWSKI, D., PAWLOWSKA, P., KOLWICZ, A. & KOTWICKI, T. 2012. Evaluation of the stress level of children with idiopathic scoliosis in relation to the method of treatment and parameters of the deformity. *Scientific World Journal,* 2012**,** 538409.

LI, M., WANG, C. F., GU, S. X., HE, S. S., ZHU, X. D., ZHAO, Y. C. & ZHANG, J. T. 2009. Adapted simplified Chinese (mainland) version of Scoliosis Research Society-22 questionnaire. *Spine (Phila Pa 1976),* 34**,** 1321-4.

LIMA, M., FERREIRA, A. S., REIS, F. J. J., PAES, V. & MEZIAT-FILHO, N. 2018. Chronic low back pain and back muscle activity during functional tasks. *Gait & Posture,* 61**,** 250-256.

LITTLE, R. J. 1988. A test of missing completely at random for multivariate data with missing values. *Journal of the American statistical Association,* 83**,** 1198-1202.

LOHR, K. N. & ZEBRACK, B. J. 2009. Using patient-reported outcomes in clinical practice: challenges and opportunities. *Quality of Life Research,* 18**,** 99-107.

LONJON, G., ILHARREBORDE, B., ODENT, T., MOREAU, S., GLORION, C. & MAZDA, K. 2014. Reliability and Validity of the French-Canadian Version of the Scoliosis Research Society 22 Questionnaire in France. *Spine,* 39**,** E26-E34.

LONSTEIN, J. E. 1994. Adolescent idiopathic scoliosis. *Lancet,* 344**,** 1407-12.

LOWE, T. G., EDGAR, M., MARGULIES, J. Y., MILLER, N. H., RASO, V. J., REINKER, K. A. & RIVARD, C. H. 2000. Etiology of idiopathic scoliosis: current trends in research. *J Bone Joint Surg Am,* 82**,** 1157-68.

LUAN, F. J., WAN, Y., MAK, K. C., MA, C. J. & WANG, H. Q. 2020. Cancer and mortality risks of patients with scoliosis from radiation exposure: a systematic review and meta-analysis. *Eur Spine J,* 29**,** 3123-3134.

LUBICKY, J. P., HANSON, J. E. & RILEY, E. H. 2011. Instrumentation constructs in pediatric patients undergoing deformity correction correlated with Scoliosis Research Society scores. *Spine (Phila Pa 1976),* 36**,** 1692-700.

MADERA, M., BRADY, J., DEILY, S., MCGINTY, T., MOROZ, L., SINGH, D., TIPTON, G. & TRUUMEES, E. 2017. The role of physical therapy and rehabilitation after lumbar fusion surgery for degenerative disease: a systematic review. *Journal of Neurosurgery: Spine,* 26**,** 694-704.

MAGASI, S., RYAN, G., REVICKI, D., LENDERKING, W., HAYS, R. D., BROD, M., SNYDER, C., BOERS, M. & CELLA, D. 2012. Content validity of patient-reported outcome measures: perspectives from a PROMIS meeting. *Qual Life Res,* 21**,** 739-46.

MAHER, C., HADFIELD, M., HUTCHINGS, M. & DE EYTO, A. 2018. Ensuring Rigor in Qualitative Data Analysis:A Design Research Approach to Coding Combining NVivo With Traditional Material Methods. *International Journal of Qualitative Methods,* 17**,** 1609406918786362.

MAKINO, T., KAITO, T., KASHII, M., IWASAKI, M. & YOSHIKAWA, H. 2015. Low back pain and patient-reported QOL outcomes in patients with adolescent idiopathic scoliosis without corrective surgery. *Springerplus,* 4**,** 397.

MANNION, A. F., ELFERING, A., FEKETE, T. F., HARDING, I. J., MONTICONE, M., OBID, P., NIEMEYER, T., LILJENQVIST, U., BOSS, A., ZIMMERMANN, L., VILA-CASADEMUNT, A., SÁNCHEZ PÉREZ-GRUESO, F. J., PIZONES, J., PELLISÉ, F., RICHNER-WUNDERLIN, S., KLEINSTÜCK, F. S., OBEID, I., BOISSIERE, L., ALANAY, A. & BAGÓ, J. 2022. Shorter and sweeter: the 16-item version of the SRS questionnaire shows better structural validity than the 20-item version in young patients with spinal deformity. *Spine Deformity,* 10**,** 1055-1062.

MASTER, H., PENNINGS, J. S., CORONADO, R. A., HENRY, A. L., O'BRIEN, M. T., HAUG, C. M., SKOLASKY, R. L., RILEY, L. H. I., NEUMAN, B. J., CHENG, J. S., AARONSON, O. S., DEVIN, C. J., WEGENER, S. T. & ARCHER, K. R. 2020. Physical Performance Tests Provide Distinct Information in Both Predicting and Assessing Patient-Reported Outcomes Following Lumbar Spine Surgery. *Spine,* 45**,** E1556-E1563.

MATZA, L. S., PATRICK, D. L., RILEY, A. W., ALEXANDER, J. J., RAJMIL, L., PLEIL, A. M. & BULLINGER, M. 2013. Pediatric Patient-Reported Outcome Instruments for Research to Support Medical Product Labeling: Report of the ISPOR PRO Good Research Practices for the Assessment of Children and Adolescents Task Force. *Value Health,* 16**,** 461-479.

MATZA, L. S., SWENSEN, A. R., FLOOD, E. M., SECNIK, K. & LEIDY, N. K. 2004. Assessment of health-related quality of life in children: a review of conceptual, methodological, and regulatory issues. *Value Health,* 7**,** 79-92.

MAYS, N. & POPE, C. 1995. Qualitative Research: Rigour and qualitative research. *BMJ : British Medical Journal,* 311**,** 109.

MAYS, N. & POPE, C. 2000. Qualitative research in health care. Assessing quality in qualitative research. *Bmj,* 320**,** 50-2.

MCNEILL, M., NOYEK, S., ENGEDA, E. & FAYED, N. 2021. Assessing the engagement of children and families in selecting patient-reported outcomes (PROs) and developing their measures: a systematic review. *Quality of Life Research,* 30**,** 983-995.

MENS, R. H., BISSELING, P., DE KLEUVER, M. & VAN HOOFF, M. L. 2022. Relevant impact of surgery on quality of life for adolescent idiopathic scoliosis: a registry-based two-year follow-up cohort study. *The Bone & Joint Journal,* 104**,** 265-273.

MERENDA, L., COSTELLO, K., SANTANGELO, A. M. & MULCAHEY, M. J. 2011. Perceptions of self-image and physical appearance: Conversations with typically developing youth and youth with idiopathic scoliosis. *Orthopaedic Nursing,* 30**,** 383-390.

MILLER, K., CHEPP, V., WILLSON, S. & PADILLA, J.-L. 2014. *Cognitive interviewing methodology*, John Wiley & Sons.

MISTERSKA, E., GŁOWACKI, J., OKRĘT, A., LAURENTOWSKA, M. & GŁOWACKI, M. 2017. Back and neck pain and function in females with adolescent idiopathic scoliosis: A follow-up at least 23 years after conservative treatment with a Milwaukee brace. *PLoS One,* 12**,** e0189358.

MISTOVICH, R. J., LUCAS, A. B. & GLOTZBECKER, M. 2023. Surgical Level Selection in Adolescent Idiopathic Scoliosis: An Evidence-Based Approach. *Journal of the American Academy of Orthopaedic Surgeons*.

MITCHELL, S. L., MCLAUGHLIN, K. H., BACHMANN, K. R., SPONSELLER, P. D. & REIDER, L. M. 2022. Construct Validity of Pediatric PROMIS Computerized Adaptive Testing Measures in Children With Adolescent Idiopathic Scoliosis. *J Pediatr Orthop,* 42**,** e720-e726.

MITSIAKI, I., THIRIOS, A., PANAGOULI, E., BACOPOULOU, F., PASPARAKIS, D., PSALTOPOULOU, T., SERGENTANIS, T. N. & TSITSIKA, A. 2022. Adolescent Idiopathic Scoliosis and Mental Health Disorders: A Narrative Review of the Literature. *Children (Basel),* 9.

MOHTADI, N. G. 2016. Outcome Measure Development. *Instr Course Lect,* 65**,** 577-82.

MOKKINK, L. B., DE VET, H. C. W., PRINSEN, C. A. C., PATRICK, D. L., ALONSO, J., BOUTER, L. M. & TERWEE, C. B. 2018a. COSMIN Risk of Bias checklist for systematic reviews of Patient-Reported Outcome Measures. *Qual Life Res,* 27**,** 1171-1179.

MOKKINK, L. B., PRINSEN, C., PATRICK, D. L., ALONSO, J., BOUTER, L. M., DE VET, H., TERWEE, C. B. & MOKKINK, L. 2018b. COSMIN methodology for systematic reviews of Patient-Reported outcome measures (PROMs). *User manual*.

MOKKINK, L. B., TERWEE, C. B., PATRICK, D. L., ALONSO, J., STRATFORD, P. W., KNOL, D. L., BOUTER, L. M. & DE VET, H. C. 2010. The COSMIN study reached international consensus on taxonomy, terminology, and definitions of measurement properties for health-related patient-reported outcomes. *J Clin Epidemiol,* 63**,** 737-45.

MONTICONE, M., AMBROSINI, E., CAZZANIGA, D., ROCCA, B. & FERRANTE, S. 2014. Active self-correction and task-oriented exercises reduce spinal deformity and improve quality of life in subjects with mild adolescent idiopathic scoliosis. Results of a randomised controlled trial. *European Spine Journal,* 23**,** 1204-1214.

MONTICONE, M., BAIARDI, P., CALABRO, D., CALABRO, F. & FOTI, C. 2010. Development of the Italian version of the revised Scoliosis Research Society-22 Patient Questionnaire, SRS-22r-I: cross-cultural adaptation, factor analysis, reliability, and validity. *Spine (Phila Pa 1976),* 35**,** E1412-7.

MONTICONE, M., CARABALONA, R. & NEGRINI, S. 2004. Reliability of the Scoliosis Research Society-22 Patient Questionnaire (Italian version) in mild adolescent vertebral deformities. *Eura Medicophys,* 40**,** 191-7.

MOTYER, G., DOOLEY, B., KIELY, P. & FITZGERALD, A. 2021a. Parents' information needs, treatment concerns, and psychological well-being when their child is diagnosed with adolescent idiopathic scoliosis: A systematic review. *Patient Educ Couns,* 104**,** 1347-1355.

MOTYER, G. S., KIELY, P. J. & FITZGERALD, A. 2021b. Adolescents' Experiences of Idiopathic Scoliosis in the Presurgical Period: A Qualitative Study. *Journal of Pediatric Psychology,* 47**,** 225-235.

MOTYER, G. S., KIELY, P. J. & FITZGERALD, A. 2022. Adolescents' Experiences of Idiopathic Scoliosis in the Presurgical Period: A Qualitative Study. *Journal of Pediatric Psychology,* 47**,** 225-235.

MOUSAVI, S. J., MOBINI, B., MEHDIAN, H., AKBARNIA, B., BOUZARI, B., ASKARY-ASHTIANI, A., MONTAZERI, A. & PARNIANPOUR, M. 2010. Reliability and validity of the persian version of the scoliosis research society-22r questionnaire. *Spine,* 35**,** 784-789.

MUKAKA, M. M. 2012. Statistics corner: A guide to appropriate use of correlation coefficient in medical research. *Malawi Med J,* 24**,** 69-71.

MUNGUÍA-IZQUIERDO, D., PULIDO-MARTOS, M., ACOSTA, F. M., ACOSTA-MANZANO, P., GAVILÁN-CARRERA, B., RODRIGUEZ-AYLLON, M., GEENEN, R., DELGADO-FERNÁNDEZ, M., ÁLVAREZ-GALLARDO, I. C., SEGURA-JIMÉNEZ, V., WALITT, B. & ESTÉVEZ-LÓPEZ, F. 2021. Objective and subjective measures of physical functioning in women with fibromyalgia: what type of measure is associated most clearly with subjective well-being? *Disabil Rehabil,* 43**,** 1649-1656.

NAULT, M. L., ALLARD, P., HINSE, S., LE BLANC, R., CARON, O., LABELLE, H. & SADEGHI, H. 2002. Relations between standing stability and body posture parameters in adolescent idiopathic scoliosis. *Spine,* 27**,** 1911-1917.

NAVARRO, I., ROSA, B. N. D. & CANDOTTI, C. T. 2019. Anatomical reference marks, evaluation parameters and reproducibility of surface topography for evaluating the adolescent idiopathic scoliosis: a systematic review with meta-analysis. *Gait Posture,* 69**,** 112-120.

NEGRINI, S., AULISA, A. G., AULISA, L., CIRCO, A. B., DE MAUROY, J. C., DURMALA, J., GRIVAS, T. B., KNOTT, P., KOTWICKI, T., MARUYAMA, T., MINOZZI, S., O'BRIEN, J. P., PAPADOPOULOS, D., RIGO, M., RIVARD, C. H., ROMANO, M., WYNNE, J. H., VILLAGRASA, M., WEISS, H. R. & ZAINA, F. 2012. 2011 SOSORT guidelines: Orthopaedic and Rehabilitation treatment of idiopathic scoliosis during growth. *Scoliosis,* 7**,** 3.

NEGRINI, S., DONZELLI, S., AULISA, A., CZAPROWSKI, D., SCHREIBER, S., MAUROY, J., DIERS, H., GRIVAS, T., KNOTT, P., KOTWICKI, T., LEBEL, A., MARTI, C., MARUYAMA, T., O'BRIEN, J., PRICE, N., PARENT, E., RIGO, M., ROMANO, M., STIKELEATHER, L. & ZAINA, F. 2018. 2016 SOSORT guidelines: Orthopaedic and rehabilitation treatment of idiopathic scoliosis during growth. *Scoliosis and Spinal Disorders,* 13.

NEWTON, P. O. 1996. Moe's Textbook of Scoliosis and Other Spinal Deformities (Third Edition). *Journal of Pediatric Orthopaedics,* 16.

NIEMEYER, T., SCHUBERT, C., HALM, H. F., HERBERTS, T., LEICHTLE, C. & GESICKI, M. 2009. Validity and reliability of an adapted german version of scoliosis research society-22 questionnaire. *Spine (Phila Pa 1976),* 34**,** 818-21.

NOKARIYA, S., KOTANI, T., SAKUMA, T., IIJIMA, Y., OKUMURA, T., KATOGI, T., OKUWAKI, S., MIYAGI, M., INOUE, G., AKAZAWA, T., SHIGA, Y., MINAMI, S., OHTORI, S. & TAKASO, M. 2022. Trunk flexibility using a sit-and-reach test after surgery for adolescent idiopathic scoliosis. *Spine deformity*.

NOWELL, L. S., NORRIS, J. M., WHITE, D. E. & MOULES, N. J. 2017. Thematic Analysis:Striving to Meet the Trustworthiness Criteria. *International Journal of Qualitative Methods,* 16**,** 1609406917733847.

NTSIEA, V., MUDZI, W., MALEKA, D., COMLEY-WHITE, N. & PILUSA, S. 2022. Barriers and facilitators of using outcome measures in stroke rehabilitation in South Africa. *International Journal of Therapy and Rehabilitation,* 29**,** 1-15.

PAGE, M. J., MCKENZIE, J. E., BOSSUYT, P. M., BOUTRON, I., HOFFMANN, T. C., MULROW, C. D., SHAMSEER, L., TETZLAFF, J. M., AKL, E. A., BRENNAN, S. E., CHOU, R., GLANVILLE, J., GRIMSHAW, J. M., HRÓBJARTSSON, A., LALU, M. M., LI, T., LODER, E. W., MAYO-WILSON, E., MCDONALD, S., MCGUINNESS, L. A., STEWART, L. A., THOMAS, J., TRICCO, A. C., WELCH, V. A., WHITING, P. & MOHER, D. 2021. The PRISMA 2020 statement: an updated guideline for reporting systematic reviews. *BMJ,* 372**,** n71.

PAINTER, P., STEWART, A. L. & CAREY, S. 1999. Physical Functioning: Definitions, Measurement, and Expectations. *Advances in Renal Replacement Therapy,* 6**,** 110-123.

PANAGIOTIS, K., VASILEIOS, V., VASILEIOS, S. & VASILEIOS, T. 2019. Evolution observation of coronal and sagittal spinal curvatures in school children with non-invasive, non-radiating methods: Scoliometer and Debrunner Kyphometer. *Trends in Medicine*.

PARENT, E. C., DANG, R., HILL, D., MAHOOD, J., MOREAU, M., RASO, J. & LOU, E. 2010. Score distribution of the scoliosis research society-22 questionnaire in subgroups of patients of all ages with idiopathic scoliosis. *Spine,* 35**,** 568-577.

PARENT, E. C., HILL, D., MAHOOD, J., MOREAU, M., RASO, J. & LOU, E. 2009. Discriminative and predictive validity of the scoliosis research society-22 questionnaire in management and curve-severity subgroups of adolescents with idiopathic scoliosis. *Spine,* 34**,** 2450-2457.

PARENT, E. C., HILL, D., MOREAU, M., MAHOOD, J., RASO, J. & LOU, E. 2007. Score distribution of the Scoliosis Quality of Life Index questionnaire in different subgroups of patients with adolescent idiopathic scoliosis. *Spine (Phila Pa 1976),* 32**,** 1767-77.

PARENT, S., NEWTON, P. O. & WENGER, D. R. 2005. Adolescent idiopathic scoliosis: etiology, anatomy, natural history, and bracing. *Instr Course Lect,* 54**,** 529-36.

PARIA, N. & WISE, C. A. 2015. Genetics of adolescent idiopathic scoliosis. *Seminars in Spine Surgery,* 27**,** 9-15.

PATRICK, D. L., BURKE, L. B., GWALTNEY, C. J., LEIDY, N. K., MARTIN, M. L., MOLSEN, E. & RING, L. 2011a. Content validity--establishing and reporting the evidence in newly developed patient-reported outcomes (PRO) instruments for medical product evaluation: ISPOR PRO good research practices task force report: part 1--eliciting concepts for a new PRO instrument. *Value Health,* 14**,** 967-77.

PATRICK, D. L., BURKE, L. B., GWALTNEY, C. J., LEIDY, N. K., MARTIN, M. L., MOLSEN, E. & RING, L. 2011b. Content validity--establishing and reporting the evidence in newly developed patient-reported outcomes (PRO) instruments for medical product evaluation: ISPOR PRO Good Research Practices Task Force report: part 2--assessing respondent understanding. *Value Health,* 14**,** 978-88.

PAYNE, W. K., III, OGILVIE, J. W., RESNICK, M. D., KANE, R. L., TRANSFELDT, E. E. & BLUM, R. W. 1997. Does Scoliosis Have a Psychological Impact and Does Gender Make a Difference? *Spine,* 22.

PERRY, M., STARKWEATHER, A., BAUMBAUER, K. & YOUNG, E. 2018. Factors Leading to Persistent Postsurgical Pain in Adolescents Undergoing Spinal Fusion: An Integrative Literature Review. *Journal of Pediatric Nursing,* 38**,** 74-80.

PERRY-EADDY, M. 2018. "I thought I was going to die": A meta-synthesis exploring pediatric pain after scoliosis surgery. *International Journal of Orthopaedic and Trauma Nursing,* 31.

PORTER, M. E. 2010. What is value in health care? *N Engl J Med,* 363**,** 2477-81.

PRINSEN, C. A. C., MOKKINK, L. B., BOUTER, L. M., ALONSO, J., PATRICK, D. L., DE VET, H. C. W. & TERWEE, C. B. 2018. COSMIN guideline for systematic reviews of patient-reported outcome measures. *Qual Life Res,* 27**,** 1147-1157.

PRINSEN, C. A. C., VOHRA, S., ROSE, M. R., BOERS, M., TUGWELL, P., CLARKE, M., WILLIAMSON, P. R. & TERWEE, C. B. 2016. How to select outcome measurement instruments for outcomes included in a "Core Outcome Set" – a practical guideline. *Trials,* 17**,** 449.

PROWSE, A., POPE, R., GERDHEM, P. & ABBOTT, A. 2016. Reliability and validity of inexpensive and easily administered anthropometric clinical evaluation methods of postural asymmetry measurement in adolescent idiopathic scoliosis: a systematic review. *European spine journal,* 25**,** 450-466.

RAAD, M., NEUMAN, B. J., JAIN, A., HASSANZADEH, H., PASSIAS, P. G., KLINEBERG, E., MUNDIS, G. M., PROTOPSALTIS, T. S., MILLER, E. K. & SMITH, J. S. 2018. The use of patient-reported preoperative activity levels as a stratification tool for short-term and long-term outcomes in patients with adult spinal deformity. *Journal of Neurosurgery: Spine,* 29**,** 68-74.

RAF, H. M., PEPIJN, B., MARINUS DE, K. & MIRANDA, L. V. H. 2022. Relevant impact of surgery on quality of life for adolescent idiopathic scoliosis : a registry-based two-year follow-up cohort study. *The bone & joint journal*.

RAMIREZ, N., JOHNSTON, C. E. & BROWNE, R. H. 1997. The prevalence of back pain in children who have idiopathic scoliosis. *J Bone Joint Surg Am,* 79**,** 364-8.

RAZALI, N. M. & WAH, Y. B. 2011. Power comparisons of shapiro-wilk, kolmogorov-smirnov, lilliefors and anderson-darling tests. *Journal of statistical modeling and analytics,* 2**,** 21-33.

REAMY, B. V. & SLAKEY, J. B. 2001. Adolescent idiopathic scoliosis: review and current concepts. *Am Fam Physician,* 64**,** 111-6.

REIMAN, M. P. & MANSKE, R. C. 2011. The assessment of function: How is it measured? A clinical perspective. *J. Man. Manip. Ther.,* 19**,** 91-99.

RICHARDS, T., COULTER, A. & WICKS, P. 2015. Time to deliver patient centred care. British Medical Journal Publishing Group.

RIGO, M. 2011. Patient evaluation in idiopathic scoliosis: Radiographic assessment, trunk deformity and back asymmetry. *Physiotherapy Theory & Practice,* 27**,** 7-25.

RITCHIE, J., LEWIS, J., MCNAUGHTON NICHOLLS, C. & ORMSTON, R. 2014. Qualitative research practice : a guide for social science students and researchers. Second edition / edited by Jane Ritchie, Jane Lewis, Carol McNaughton Nicholls, Rachel Ormston. ed.: Los Angeles : SAGE, 2014.

ROBERTS, D. W., SAVAGE, J. W., SCHWARTZ, D. G., CARREON, L. Y., SUCATO, D. J., SANDERS, J. O., RICHARDS, B. S., LENKE, L. G., EMANS, J. B., PARENT, S. & SARWARK, J. F. 2011. Male-female differences in Scoliosis Research Society-30 scores in adolescent idiopathic scoliosis. *Spine (Phila Pa 1976),* 36**,** E53-9.

ROBERTSON, S., KREMER, P., AISBETT, B., TRAN, J. & CERIN, E. 2017. Consensus on measurement properties and feasibility of performance tests for the exercise and sport sciences: a Delphi study. *Sports Med Open,* 3**,** 2.

RODRIGUES, L. M. R., GOTFRYD, A. O., MACHADO, A. N., DEFINO, M. & ASANO, L. Y. J. 2017. ADOLESCENT IDIOPATHIC SCOLIOSIS: SURGICAL TREATMENT AND QUALITY OF LIFE. *Acta Ortopedica Brasileira,* 25**,** 85-89.

ROSE, L. D., WILLIAMS, R., AJAYI, B., ABDALLA, M., BERNARD, J., BISHOP, T., PAPADAKOS, N. & LUI, D. F. 2023. Reducing radiation exposure and cancer risk for children with scoliosis: EOS the new gold standard. *Spine Deform,* 11**,** 847-851.

ROTHMAN, M., BURKE, L., ERICKSON, P., LEIDY, N. K., PATRICK, D. L. & PETRIE, C. D. 2009. Use of existing patient-reported outcome (PRO) instruments and their modification: the ISPOR Good Research Practices for Evaluating and Documenting Content Validity for the Use of Existing Instruments and Their Modification PRO Task Force Report. *Value Health,* 12**,** 1075-83.

RULLANDER, A.-C., ISBERG, S., KARLING, M., JONSSON, H. & LINDH, V. 2013. Adolescents' experience with scoliosis surgery: a qualitative study. *Pain Management Nursing,* 14**,** 50-59.

RULLANDER, A.-C., LUNDSTRÖM, M., ÖSTLUND, U. & LINDH, V. 2017. Adolescents' Experiences of Scoliosis Surgery and the Trajectory of Self-Reported Pain. *Orthopaedic Nursing,* 36**,** 414-423.

RULLANDER, A. C., LUNDSTRÖM, M., LINDKVIST, M., HÄGGLÖF, B. & LINDH, V. 2016. Stress symptoms among adolescents before and after scoliosis surgery: correlations with postoperative pain. *Journal of Clinical Nursing,* 25**,** 1086-1094.

RUSHTON, P. R. & GREVITT, M. P. 2013. What is the effect of surgery on the quality of life of the adolescent with adolescent idiopathic scoliosis? A review and statistical analysis of the literature. *Spine,* 38**,** 786-794.

RYAN, K., GANNON-SLATER, N. & CULBERTSON, M. J. 2012. Improving Survey Methods With Cognitive Interviews in Small- and Medium-Scale Evaluations. *American Journal of Evaluation,* 33**,** 414-430.

SAINANI, K. L. 2015. Dealing With Missing Data. *Pm r,* 7**,** 990-994.

SANDERS, A. E., ANDRAS, L. M., IANTORNO, S. E., HAMILTON, A., CHOI, P. D. & SKAGGS, D. L. 2018. Clinically Significant Psychological and Emotional Distress in 32% of Adolescent Idiopathic Scoliosis Patients. *Spine Deform,* 6**,** 435-440.

SANDERS, J. O., CARREON, L. Y., SUCATO, D. J., STURM, P. F., DIAB, M. & GROUP, S. D. S. 2010. Preoperative and perioperative factors effect on adolescent idiopathic scoliosis surgical outcomes. *Spine,* 35**,** 1867-1871.

SAPOUNTZI-KREPIA, D., PSYCHOGIOU, M., PETERSON, D., ZAFIRI, V., IORDANOPOULOU, E., MICHAILIDOU, F. & CHRISTODOULOU, A. 2006. The experience of brace treatment in children/adolescents with scoliosis. *Scoliosis,* 1**,** 8.

SARWAHI, V., WENDOLOWSKI, S., GECELTER, R., MAGUIRE, K., GAMBASSI, M., ORLANDO, D., LO, Y. & AMARAL, T. 2018. When Do Patients Return to Physical Activities and Athletics After Scoliosis Surgery?: A Validated Patient Questionnaire Based Study. *Spine (Phila Pa 1976),* 43**,** 167-171.

SATHIRA-ANGKURA, V., PITHANKUAKUL, K., SAKULPIPATANA, S., PIYASKULKAEW, C. & KUNAKORNSAWAT, S. 2012. Validity and reliability of an adapted Thai version of Scoliosis Research Society-22 questionnaire for adolescent idiopathic scoliosis. *Spine,* 37**,** 783-787.

SCHLOSSER, T. P., STADHOUDER, A., SCHIMMEL, J. J., LEHR, A. M., VAN DER HEIJDEN, G. J. & CASTELEIN, R. M. 2014. Reliability and validity of the adapted Dutch version of the revised Scoliosis Research Society 22-item questionnaire. *Spine J,* 14**,** 1663-72.

SCHMID, S., BURKHART, K. A., ALLAIRE, B. T., GRINDLE, D., BASSANI, T., GALBUSERA, F. & ANDERSON, D. E. 2020. Spinal Compressive Forces in Adolescent Idiopathic Scoliosis With and Without Carrying Loads: A Musculoskeletal Modeling Study. *Frontiers in Bioengineering and Biotechnology,* 8.

SEID, M., VARNI, J. W. & JACOBS, J. R. 2004. Pediatric Health-Related Quality-of-Life Measurement Technology: Intersections between Science, Managed Care, and Clinical Care. *Journal of Clinical Psychology in Medical Settings,* 7**,** 17-27.

SEKI, H., IDENO, S., ISHIHARA, T., WATANABE, K., MATSUMOTO, M. & MORISAKI, H. 2018. Postoperative pain management in patients undergoing posterior spinal fusion for adolescent idiopathic scoliosis: a narrative review. *Scoliosis Spinal Disord.,* 13**,** 17.

SENKOYLU, A., ILHAN, M. N., ALTUN, N., SAMARTZIS, D. & LUK, K. D. K. 2021. A simple method for assessing rotational flexibility in adolescent idiopathic scoliosis: modified Adam's forward bending test. *Spine Deform,* 9**,** 333-339.

SEOKWON, H., LUCY YOUGMIN, E., NAM KYUN, K., NAM KYUN, K., JO WON, J., JAE YOUNG, C., JAE YOUNG, C., JAE YOUNG, C. & HAK SUN, K. 2015. Cardiopulmonary function and scoliosis severity in idiopathic scoliosis children. *Korean Journal of Pediatrics*.

SHAN, G. & GERSTENBERGER, S. 2017. Fisher's exact approach for post hoc analysis of a chi-squared test. *PloS one,* 12**,** e0188709.

SHEA, B. J., REEVES, B. C., WELLS, G., THUKU, M., HAMEL, C., MORAN, J., MOHER, D., TUGWELL, P., WELCH, V., KRISTJANSSON, E. & HENRY, D. A. 2017. AMSTAR 2: a critical appraisal tool for systematic reviews that include randomised or non-randomised studies of healthcare interventions, or both. *BMJ,* 358**,** j4008.

SHOREY, S. & NG, E. D. 2022. Examining characteristics of descriptive phenomenological nursing studies: A scoping review. *Journal of Advanced Nursing,* 78**,** 1968-1979.

SIEBERG, C. B., SIMONS, L. E., EDELSTEIN, M. R., DEANGELIS, M. R., PIELECH, M., SETHNA, N. & HRESKO, M. T. 2013. Pain prevalence and trajectories following pediatric spinal fusion surgery. *Journal of Pain,* 14**,** 1694-1702.

SIMONY, A., CARREON, L. Y. & ANDERSEN, M. O. 2016. Reliability and Validity Testing of a Danish Translated Version of the Scoliosis Research Society Instrument-22 Revised (SRS-22R). *Spine Deformity,* 4**,** 16-21.

SINGH, H., SHIPRA, SHARMA, V., SHARMA, I., SHARMA, A., MODEEL, S., GUPTA, N., GUPTA, G., PANDITA, A. K., BUTT, M. F., SHARMA, R., PANDITA, S., SINGH, V., RAI, E., IKEGAWA, S. & SHARMA, S. 2022. The first study of epidemiology of adolescent idiopathic scoliosis shows lower prevalence in females of Jammu and Kashmir, India. *Am J Transl Res,* 14**,** 1100-1106.

SNYDER, C. F., WATSON, M. E., JACKSON, J. D., CELLA, D. & HALYARD, M. Y. 2007. Patient-reported outcome instrument selection: designing a measurement strategy. *Value Health,* 10 Suppl 2**,** S76-85.

SOLANS, M., PANE, S., ESTRADA, M. D., SERRA-SUTTON, V., BERRA, S., HERDMAN, M., ALONSO, J. & RAJMIL, L. 2008. Health-related quality of life measurement in children and adolescents: a systematic review of generic and disease-specific instruments. *Value Health,* 11**,** 742-64.

SPERANDEI, S. 2014. Understanding logistic regression analysis. *Biochemia medica,* 24**,** 12-18.

SPRATT, K. F. 2009. Minimal clinically important difference based on clinical judgment and minimally detectable measurement difference: a rationale for the SF-36 Physical Function scale in the SPORT Intervertebral disc herniation cohort. *Spine,* 34**,** 1722.

STANISZEWSKA, S., HAYWOOD, K. L., BRETT, J. & TUTTON, L. 2012. Patient and public involvement in patient-reported outcome measures: evolution not revolution. *Patient,* 5**,** 79-87.

STĘPIEŃ, A., GUZEK, K., PAŁDYNA, B. & REKOWSKI, W. 2018. The Trunk-Pelvis-Hip Angle Test is a Reliable Measurement of the Range of the Lower Trunk-Pelvis Rotation in Adolescents. *J Orthop Ther: JORT-1124. DOI,* 10**,** 2575-8241.

STREINER, D. L., NORMAN, G. R. & CAIRNEY, J. 2015. *Health Measurement Scales : A Practical Guide to Their Development and Use,* Oxford, UNITED KINGDOM, Oxford University Press, Incorporated.

STUDENSKI, S., PERERA, S., WALLACE, D., CHANDLER, J. M., DUNCAN, P. W., ROONEY, E., FOX, M. & GURALNIK, J. M. 2003. Physical performance measures in the clinical setting. *Journal of the American Geriatrics Society,* 51**,** 314-322.

SWIERKOSZ, S., NOWAK, Z., WIERKOSZ, S. & NOWAK, Z. 2015. Low back pain in adolescents. An assessment of the quality of life in terms of qualitative and quantitative pain variables. *Journal of Back and Musculoskeletal Rehabilitation,* 28**,** 25-34.

TARRANT, R. C., O'LOUGHLIN, P. F., LYNCH, S., QUEALLY, J. M., SHEERAN, P., MOORE, D. P. & KIELY, P. J. 2014. Timing and predictors of return to short-term functional activity

in adolescent idiopathic scoliosis after posterior spinal fusion: a prospective study. *Spine,* 39**,** 1471-1478.

TAYLOR, A. M., PHILLIPS, K., PATEL, K. V., TURK, D. C., DWORKIN, R. H., BEATON, D., CLAUW, D. J., GIGNAC, M. A., MARKMAN, J. D. & WILLIAMS, D. A. 2016a. Assessment of physical function and participation in chronic pain clinical trials: IMMPACT/OMERACT recommendations. *Pain,* 157**,** 1836-1850.

TAYLOR, A. M., PHILLIPS, K., PATEL, K. V., TURK, D. C., DWORKIN, R. H., BEATON, D., CLAUW, D. J., GIGNAC, M. A., MARKMAN, J. D., WILLIAMS, D. A., BUJANOVER, S., BURKE, L. B., CARR, D. B., CHOY, E. H., CONAGHAN, P. G., COWAN, P., FARRAR, J. T., FREEMAN, R., GEWANDTER, J., GILRON, I., GOLI, V., GOVER, T. D., HADDOX, J. D., KERNS, R. D., KOPECKY, E. A., LEE, D. A., MALAMUT, R., MEASE, P., RAPPAPORT, B. A., SIMON, L. S., SINGH, J. A., SMITH, S. M., STRAND, V., TUGWELL, P., VANHOVE, G. F., VEASLEY, C., WALCO, G. A., WASAN, A. D. & WITTER, J. 2016b. Assessment of physical function and participation in chronic pain clinical trials: IMMPACT/OMERACT recommendations. *Pain,* 157**,** 1836-50.

TERRY, G., HAYFIELD, N., CLARKE, V. & BRAUN, V. 2017. Thematic analysis. *The SAGE handbook of qualitative research in psychology,* 2**,** 17-37.

TERWEE, C., PRINSEN, C., CHIAROTTO, A., WESTERMAN, M., PATRICK, D., ALONSO, J., BOUTER, L., DE VET, H. & MOKKINK, L. 2018a. COSMIN methodology for evaluating the content validity of patient-reported outcome measures: a Delphi study. *Quality of Life Research,* 27.

TERWEE, C. B., JANSMA, E. P., RIPHAGEN, I. I. & DE VET, H. C. W. 2009. Development of a methodological PubMed search filter for finding studies on measurement properties of measurement instruments. *Quality of Life Research,* 18**,** 1115-1123.

TERWEE, C. B., PRINSEN, C., CHIAROTTO, A., DE VET, H., BOUTER, L. M., ALONSO, J., WESTERMAN, M. J., PATRICK, D. L. & MOKKINK, L. B. 2018b. COSMIN methodology for assessing the content validity of PROMs–user manual. *Amsterdam: VU University Medical Center*.

TERWEE, C. B., PRINSEN, C. A. C., CHIAROTTO, A., WESTERMAN, M. J., PATRICK, D. L., ALONSO, J., BOUTER, L. M., DE VET, H. C. W. & MOKKINK, L. B. 2018c. COSMIN methodology for evaluating the content validity of patient-reported outcome measures: a Delphi study. *Qual Life Res,* 27**,** 1159-1170.

THÉROUX, J., STOMSKI, N., HODGETTS, C. J., BALLARD, A., KHADRA, C., LE MAY, S. & LABELLE, H. 2017. Prevalence of low back pain in adolescents with idiopathic scoliosis: a systematic review. *Chiropractic & Manual Therapies,* 25**,** 10.

THOMPSON, J. Y., WILLIAMSON, E. M., WILLIAMS, M. A., HEINE, P. J. & LAMB, S. E. 2019. Effectiveness of scoliosis-specific exercises for adolescent idiopathic scoliosis compared with other non-surgical interventions: a systematic review and meta-analysis. *Physiotherapy,* 105**,** 214-234.

TOMEY, K. M. & SOWERS, M. R. 2009. Assessment of physical functioning: a conceptual model encompassing environmental factors and individual compensation strategies. *Phys Ther,* 89**,** 705-14.

TONG, A., SAINSBURY, P. & CRAIG, J. 2007. Consolidated criteria for reporting qualitative research (COREQ): a 32-item checklist for interviews and focus groups. *Int J Qual Health Care,* 19**,** 349-357.

TOYE, F., WILLIAMSON, E., WILLIAMS, M. A., FAIRBANK, J. & LAMB, S. E. 2016. What value can qualitative research add to quantitative research design? An example from an adolescent idiopathic scoliosis trial feasibility study. *Qualitative Health Research,* 26**,** 1838-1850.

TRUUMEES, D., DUNCAN, A., MAYER, E. K., GECK, M., SINGH, D. & TRUUMEES, E. 2021. Social media as a new source of medical information and support: analysis of scoliosis-specific information. *Spine Deform,* 9**,** 1241-1245.

TSIRIKOS, A. I., ROBERTS, S. B. & BHATTI, E. 2020. Incidence of spinal deformity surgery in a national health service from 2005 to 2018: an analysis of 2,205 children and adolescents. *Bone & Joint Open,* 1**,** 19-28.

VAN KAMPEN, D. A., WILLEMS, W. J., VAN BEERS, L. W., CASTELEIN, R. M., SCHOLTES, V. A. & TERWEE, C. B. 2013. Determination and comparison of the smallest detectable change (SDC) and the minimal important change (MIC) of four-shoulder patient-reported outcome measures (PROMs). *J Orthop Surg Res,* 8**,** 40.

VAN NIEKERK, M., RICHEY, A., VORHIES, J., WONG, C. & TILESTON, K. 2023. Effectiveness of psychosocial interventions for pediatric patients with scoliosis: a systematic review. *World J Pediatr Surg,* 6**,** e000513.

VERMA, K. 2014. International Utilization of the SRS-22 Instrument to Assess Outcomes in Adolescent Idiopathic Scoliosis: What Can We Learn From a Medical Outreach Group in Ghana? *Journal of Pediatric Orthopaedics,* 34**,** 503-509.

VERMA, K., LONNER, B., TOOMBS, C. S., FERRISE, P., WRIGHT, B., KING, A. B. & BOACHIE-ADJEI, O. 2014. International utilization of the SRS-22 instrument to assess outcomes in adolescent idiopathic scoliosis: what can we learn from a medical outreach group in Ghana? *J Pediatr Orthop,* 34**,** 503-8.

VIGNESWARAN, H. T., GRABEL, Z. J., EBERSON, C. P., PALUMBO, M. A. & DANIELS, A. H. 2015. Surgical treatment of adolescent idiopathic scoliosis in the United States from 1997 to 2012: an analysis of 20, 346 patients. *Journal of Neurosurgery: Pediatrics,* 16**,** 322-328.

VOGT, D., KING, D. & KING, L. 2004. Focus Groups in Psychological Assessment: Enhancing Content Validity by Consulting Members of the Target Population. *Psychological assessment,* 16**,** 231-43.

VON ELM, E., ALTMAN, D. G., EGGER, M., POCOCK, S. J., GØTZSCHE, P. C. & VANDENBROUCKE, J. P. 2014. The Strengthening the Reporting of Observational Studies in Epidemiology (STROBE) Statement: guidelines for reporting observational studies. *Int J Surg,* 12**,** 1495-9.

VON HEIDEKEN, J., IVERSEN, M. D. & GERDHEM, P. 2018. Rapidly increasing incidence in scoliosis surgery over 14 years in a nationwide sample. *European Spine Journal,* 27**,** 286-292.

WADE, R., YANG, H., MCKENNA, C., FARIA, R., GUMMERSON, N. & WOOLACOTT, N. 2013. A systematic review of the clinical effectiveness of EOS 2D/3D X-ray imaging system. *Eur Spine J,* 22**,** 296-304.

WANG, K., EFTANG, C. N., JAKOBSEN, R. B. & ÅRØEN, A. 2020. Review of response rates over time in registry-based studies using patient-reported outcome measures. *BMJ Open,* 10**,** e030808.

WANG, S., HSU, C. J., TRENT, L., RYAN, T., KEARNS, N. T., CIVILLICO, E. F. & KONTSON, K. L. 2018. Evaluation of performance-based outcome measures for the upper limb: a comprehensive narrative review. *PM&R,* 10**,** 951-962. e3.

WATANABE, K., HASEGAWA, K., HIRANO, T., UCHIYAMA, S. & ENDO, N. 2005. Use of the Scoliosis Research Society Outcomes Instrument to Evaluate Patient Outcome in Untreated Idiopathic Scoliosis Patients in Japan: Part II: Relation Between Spinal Deformity and Patient Outcomes. *Spine,* 30**,** 1202-1205.

WEINSTEIN, S. L., DOLAN, L. A., CHENG, J. C., DANIELSSON, A. & MORCUENDE, J. A. 2008. Adolescent idiopathic scoliosis. *Lancet,* 371**,** 1527-37.

WEISS, H.-R., BESS, S., WONG, M. S., PATEL, V., GOODALL, D. & BURGER, E. 2008. Adolescent idiopathic scoliosis – to operate or not? A debate article. *Patient Safety in Surgery,* 2**,** 25.

WEISS, H.-R. & GOODALL, D. 2008. Rate of complications in scoliosis surgery–a systematic review of the Pub Med literature. *Scoliosis,* 3**,** 1-18.

WEISS, H. R., ÇOLAK, T. K., LAY, M. & BORYSOV, M. 2021. Brace treatment for patients with scoliosis: State of the art. *S Afr J Physiother,* 77**,** 1573.

WHITE, S. F., ASHER, M. A., LAI, S. M. & BURTON, D. C. 1999. Patients' perceptions of overall function, pain, and appearance after primary posterior instrumentation and fusion for idiopathic scoliosis. *Spine (Phila Pa 1976),* 24**,** 1693-9; discussion 1699-700.

WHO 2001. International classification of functioning, disability and health : ICF. Geneva: World Health Organization.

WHO 2007. *International Classification of Functioning, Disability, and Health: Children & Youth Version: ICF-CY*, World Health Organization.

WILK, B., KAROL, L. A., JOHNSTON, C. E., 2ND, COLBY, S. & HAIDERI, N. 2006. The effect of scoliosis fusion on spinal motion: a comparison of fused and nonfused patients with idiopathic scoliosis. *Spine (Phila Pa 1976),* 31**,** 309-14.

WILLIAMSON, P. R., ALTMAN, D. G., BLAZEBY, J. M., CLARKE, M., DEVANE, D., GARGON, E. & TUGWELL, P. 2012. Developing core outcome sets for clinical trials: issues to consider. *Trials,* 13**,** 132.

WILLIMON, S. C., JOHNSON, M. M., HERZOG, M. M. & BUSCH, M. T. 2019. Time to Return to School After 10 Common Orthopaedic Surgeries Among Children and Adolescents. *J Pediatr Orthop,* 39**,** 322-327.

WILLIS, D. G., SULLIVAN-BOLYAI, S., KNAFL, K. & COHEN, M. Z. 2016. Distinguishing Features and Similarities Between Descriptive Phenomenological and Qualitative Description Research. *Western Journal of Nursing Research,* 38**,** 1185-1204.

WILLIS, G. 2017. Cognitive Evaluation of Survey Instruments.

WILLIS, G. B. & ARTINO, A. R., JR. 2013. What Do Our Respondents Think We're Asking? Using Cognitive Interviewing to Improve Medical Education Surveys. *J Grad Med Educ,* 5**,** 353-6.

WONG, A. Y. L., SAMARTZIS, D., CHEUNG, P. W. H. & CHEUNG, J. P. Y. 2019. How Common Is Back Pain and What Biopsychosocial Factors Are Associated With Back Pain in Patients With Adolescent Idiopathic Scoliosis? *Clin Orthop Relat Res,* 477**,** 676-686.

WONG, C. K. H., CHEUNG, P. W. H., SAMARTZIS, D., LUK, K. D. K., CHEUNG, K. M. C., LAM, C. L. K. & CHEUNG, J. P. Y. 2017. Mapping the SRS-22r questionnaire onto the EQ-5D-5L utility score in patients with adolescent idiopathic scoliosis. *PLoS ONE [Electronic Resource],* 12**,** e0175847-e0175847.

WONG, G., YUEN, V., CHOW, B. & IRWIN, M. 2007. Persistent pain in patients following scoliosis surgery. *European Spine Journal,* 16**,** 1551-1556.

WONG, L. P. K., CHEUNG, P. W. H. & CHEUNG, J. P. Y. 2022. Curve type, flexibility, correction, and rotation are predictors of curve progression in patients with adolescent idiopathic scoliosis undergoing conservative treatment. *The Bone & Joint Journal,* 104-B**,** 424-432.

WU, H. D., LIU, W. & WONG, M. S. 2020. Reliability and validity of lateral curvature assessments using clinical ultrasound for the patients with scoliosis: a systematic review. *Eur Spine J,* 29**,** 717-725.

WU, W., DU, Y., LIANG, J., CHEN, Y., TAN, X., XIANG, X., WANG, W. & RU, N. 2014. Score distribution of the scoliosis research society health-related quality of life in different subgroups of adolescent subjects unaffected by scoliosis in China. *Spine,* 39**,** 256-262.

YAGCI, G., KARATEL, M. & YAKUT, Y. 2020. Body Awareness and its Relation to Quality of Life in Individuals with Idiopathic Scoliosis. *Perceptual and Motor Skills,* 127**,** 841-857.

YAKUT, Y., PELIN, Z. & YAGCI, G. 2022. An investigation of sleep profiles in individuals with idiopathic scoliosis. *Sleep Science,* 15**,** 172-178.

YANG, J. H., SUH, S.-W., SUNG, P. S. & PARK, W.-H. 2013. Asymmetrical gait in adolescents with idiopathic scoliosis. *European Spine Journal,* 22**,** 2407-2413.

YAU, A., HEATH, M. R. & FABRICANT, P. D. 2020. Discrimination Ability of Patient Reported Outcome Measurement Information System Pediatric Domains Compared With Scoliosis Research Society-22r and Legacy Patient Reported Outcome Measures in Juvenile and Adolescent Idiopathic Scoliosis. *Spine (Phila Pa 1976),* 45**,** 1713-1719.

Appendices

Appendix 1. Alamrani et al. (2023b)

Appendix 2. Alamrani et al. (2023a)

Appendix 3. Alamrani et al. (2021b)

Appendix 4. Alamrani et al. (2021a)

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Appendix 6. STROBE Checklist and Ethical Review for *Chapter Two*

Appendix 7. Terms and keywords used in search one and search two, Risk of bias results, Interpretability and feasibility results, AMSTAR checklist for *Chapter Three*

Appendix 8. Scoliosis Research Society -22revised patient questionnaire

Appendix 9. Ethical Review, Consent Forms. Patient Information Sheets, COREQ checklist, Topic guide, and Saturation table for *Chapter Four* and *Chapter Five*

Appendix 10. Ethical Review, Ethical amendment, Consent Forms, Patient Information Sheets, COREQ checklist, and Topic guide, for *Chapter Six.*

Appendix 11. Emily Russel personal experience

Appendix 1. Alamrani et al. (2023 b)

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Predictors of relevant changes in pain and function for Adolescents with Idiopathic Scoliosis following Surgery

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ABSTRACT

Study Design. Retrospective analysis of longitudinal data.

Objective To evaluate clinically relevant change in surgical outcomes for Adolescents with Idiopathic Scoliosis (AIS), comparing those who achieved smallest detectable change (SDC) in pain and function at 1-year post-surgery and those who did not, and to evaluate the influencing factors.

Summary of Background Data. The SDC is recommended to evaluate the surgical outcomes of AIS. However, little is known about the use of SDC in AIS and its influencing factors.

Methods. This was a retrospective analysis of longitudinal data from patients who underwent surgical correction at a tertiary spinal centre from to 2009-2019. Surgical outcomes were assessed at short-term (6 weeks, 6 months) and long-term (1- and 2-years) post-surgery using the Scoliosis Research Society (SRS-22r). The difference between 'successful' (≥ SDC) and 'unsuccessful' (< SDC) groups was assessed using an independent t-test. Univariate and logistic regression analyses enabled the assessment of influencing factors.

Results All SRS-22r domains decreased in the short term, except for self-image and satisfaction. In the long term, self-image increased by 1.21 and function increased by 0.2, and pain decreased by 0.1. In all SRS-22r domains 'successful' group had low pre-surgery scores and were statistically different to the 'unsuccessful group'. The difference remained statistically significant at 1-year for most SRS-22r domains. Being older and having low pre-surgery SRS-22r scores increased the chances of achieving SDC function at 1-year. Achieving SDC in the pain domain was significantly associated with age, sex, length of hospital stay, and pre-surgical scores.

Conclusion. Notably, the self-image domain showed the largest change compared to other SRS-22r domains. A low preoperative score increases the likelihood of clinical benefit from surgery. These findings demonstrate the utility of SDC for assessing the benefits and factors that may underpin surgical benefit in AIS.

Data Access Statement: Research data supporting this publication are available from the NN repository at located at www.NNN.org/download/.

Key points

1. The smallest detectable change (SDC) can be utilized to assess clinical benefit from surgery
2. Low preoperative scores increase the likelihood of clinical benefit from surgery.
3. The largest change in the Scoliosis Research Society (SRS-22r) scores is at the self-image domain while the least change is at function domain.

Introduction

Approximately 10% of adolescents with idiopathic scoliosis (AIS) require some type of management intervention including surgery [1]. Currently, the incidence of AIS surgery increased worldwide [2-4], requiring an urgent need to further evaluate surgical outcomes.

The aim of surgery is to prevent curve progression, to improve self-image and respiratory function [1,5-7]. However, reported postoperative complaints include severe pain [8,9], new onset of pain [10,11], 20-60% decrease in spinal mobility and flexibility [12,13]. and reduced participation in sport [13] . It is important to evaluate not only absolute effect of surgery on pain and function, which are important domains in health-related quality of life (HRQOL) [14], but also to determine whether these changes are relevant and important from a patient's perspective.

Smallest change in outcome score that patient perceives as important is the Minimal Clinically Important Difference (MCID) [15]. For a clinical change to be detected, MCID value should be larger than Smallest Detectable Change (SDC), which is the smallest change that exceeds measurement error within instrument [16]. The MCID is a clinical concept that determines whether the change is meaningful to the patient, whereas the SDC is a statistical concept that ascertains that the change is real [16,17]. Any change in the score below the SDC was attributed to a measurement error [16]. Therefore, it is important to consider both values as statistical significance may not be reflecting clinical significance. Both values refer to the interpretability of patient reported outcome measures (PROM), which is an important aspect of an instrument's measurement properties, as reflected in its inclusion in COnsensus-based Standards for the selection of health Measurement INstrument (COSMIN) group's COSMIN taxonomy [18].

The Scoliosis Research Society-22 revised (SRS-22r) is a frequently used PROM to evaluate HRQOL for AIS, consisting of five domains, including pain and function [19]. The MCID values

for pain and function domains of SRS-22r are smaller than the calculated SDC values; MCID values are 0.2 and 0.08, while SDC values are 0.3 and 0.24, respectively [20,21]. Therefore, it has been suggested that SDC be used as a threshold for clinically relevant change as an alternative to MCID [20,22]. The SRS-22r has been reported to have a high ceiling effect in its function domain, which affects its ability to discriminate change [23,24]. Therefore, it is important to evaluate whether any changes are real and not due to measurement error [16]. A 30% rule was suggested by Spratt et al. for measuring clinical change rather than MCID, by accommodating PROM baseline scores [17]. However, their method is theoretical, and therefore validity of 20% and 50% rule is questionable [17].

Previous studies that used the MCID showed an improvement in pain and function scores [25-27], while others revealed no change [28]. No previous study has applied SDC values to evaluate postoperative changes and factors that influence achieving it are poorly understood.

Objectives

1] To evaluate clinically relevant change in outcomes for AIS, in the short and long-term using SDC.

2] To compare those who achieved SDC in pain and function at 1-year post-surgery and those who did not, and to evaluate influencing factors.

METHODS

Study design

Retrospective longitudinal observational study design

Setting

Data were collected at a tertiary spinal center in United Kingdom between 2009 and 2019.

Participants

Adolescents (10 to 18 years old) diagnosed with idiopathic scoliosis and completed SRS-22r questionnaire before or after surgery. Patients with other spine pathologies (e.g., neuromuscular scoliosis) were excluded. The SRS-22r domain score was excluded from analyses when less than three of possible five questions that contributed to total domain score were answered.

Ethics

Institutional Review Board at Royal Orthopaedic Hospital, Birmingham, United Kingdom (DP/ROH20ORTH01). Data were previously routinely collected and anonymized, where there was no possibility of affecting care of individuals concerned. Therefore, informed consent was not required.

Data Sources

The SRS-22r comprising domains for function, pain, self-image, mental health, and satisfaction, all but satisfaction have five items. Higher scores from 5-points Likert scale indicate better outcomes [23]. It demonstrates reliability, concurrent validity [24], and responsiveness to change post-surgically [19,29]. Demographic and clinical characteristics of participants were extracted from their medical records. Data were checked for accuracy and consistency before conducting data analyses [30].

Statistical methods

Data were checked for normality using Shapiro-wilk test [31]. Categorical variables were presented as frequencies and percentages. Demographic and clinical characteristics were descriptively summarized using mean and standard deviation (SD) or median and interquartile range (IQR), as appropriate. To check for potential selection bias, a comparison between study cohort and lost at follow-up group was performed [32], with an independent-samples t-test or Mann-Whitney U test for continuous data [33] and chi-squared test for ordinal data [34].

Outcomes for AIS were assessed using SRS-22r scores at 6 weeks, 6 months, 1 year, and 2 years. Pre-surgery scores were used as baseline scores to determine changes at 1 and 2 years. An increase beyond SDC is considered positive for improvement in outcome and reverse for a decrease below SDC.

Participants' data were then classified into successful (≥ SDC) and unsuccessful (< SDC) groups and compared using an independent-sample t-test. The SDC values for pain (0.3), function (0.24), self-image (0.3), mental health (0.27), and subtotal score (0.23) have been previously determined [20].

A univariate analysis was performed to evaluate factors associated with reaching SDC in function and pain domains at 1 year, (frequently used time point for assessment of HRQOL for spinal fusion surgery) [35]. To ascertain effects of these factors, logistic regression analysis was performed [36]. Final prediction models with odds ratios and 95% confidence intervals were calculated for each predictive factor [36]. Statistical significance was set at $p<0.05$. Statistical analysis was performed using SPSS for Mac package (IBM SPSS Statistics V.28).

RESULTS

Participants

Of 304 participants, 124 had complete follow-up data at 1-year and were included in the analysis. The cohort was compared with those who were lost to follow-up with respect to their baseline characteristics (Table 1). Only pre-surgery function scores differed (p=.037), with the study cohort exhibiting a higher functional median score (4: IQR 1.2) (Table 1)

Relevant changes

At 6 weeks post-surgery, self-image and satisfaction scores increased, while function, pain, and mental health scores decreased compared to baseline scores. All domain scores increased at 2 years, with pain scores lower at 1 year, but improved at 2 years. (Table 2)

Overall, most domains reached or exceeded SDC at 1-year, except for function and pain. Function scores increased (+ 0.2), while pain scores decreased (− 0.1) and did not exceed the SDC values. At 2 years, the pain and function had increased 0.5 by 0.3, respectively. Self-image showed the greatest improvement (+1.3) and satisfaction (+1.0), whereas the least improvement was in function domain (+0.3).

Comparison analysis

Using SDC values for each domain, fewer participants were assigned to successful group than to unsuccessful group, except for self-image and sub-total domains (Table 3). The groups were comparable with respect to general demographic and clinical characteristics, except for age (p=.02) (Supplementary File 1, Supplemental Digital Content 1, http://links.lww.com/BRS/C96).

For all domains, successful group had a low pre-surgery score, which was statistically different from that of unsuccessful group. The difference between groups remained statistically significant at 1-year post-surgery for all but function and subtotal domains. Successful group achieved and exceeded SDC value for all domains. In contrast, unsuccessful group did not achieve SDC values for any domain, except for mental health domain (+0.4). Furthermore, difference in achieving SDC values between two groups was significant in all domains (Table 3).

Predictors of relevant changes

Univariate analysis revealed a significant but weak association between functional scores, age, and length of hospital stay. Furthermore, a statistically significant negative association was found between function scores at 1 year and preoperative scores. For pain, there was a statistically significant positive association between pain at one year and sex and age. Furthermore, significant negative associations were found between pain and pre-surgery scores for all the SRS-22r domains (Supplementary File 2, Supplemental Digital Content 2, http://links.lww.com/BRS/C97).

Logistic regression analysis was performed to ascertain effects of these factors on likelihood of AIS reaching SDC value at 1-year. The model explained 44.7% (Nagelkerke R^2) (p=.001) of the variance in function scores and correctly classified 83% of cases. The increase in age increased the chance of achieving SDC in function at 1-year by 1.6 (OR: 1.6, 95% CI [1.1, 2.2]), while the chance of reaching the SDC is decreased by 0.11 with increase in function scores pre-surgery (OR: 0.11, 95% CI [0.1, 0.2]) (Table 4).

The logistic regression model for pain explained 52.6% (Nagelkerke R^2) of variance in pain scores and correctly classified 82% of cases. Sex, age, length of hospital stay, and pre-surgery scores for function and pain were significantly associated with achieving the SDC values. Males were 4.5 times more likely to achieve the SDC compared to females (OR= 4.5, 95% CI: [1.1, 18.5]). Similar to the function model, older ages were 2.7 times more likely to achieve SDC than younger patients (OR=2.7, 95% CI: [1.7, 4.5]). Also, those who spent more days at hospital were 1.1 more likely to achieve SDC post-surgery (OR=1.1, 95% CI: [1.1, 1.3]). The chance of reaching the SDC for pain decreased by 0.12 with each increase in pain scores pre-surgery (OR=0.12, 95% CI: [0.1, 0.3]), while the chance is increased by 3.5 with every increase in function score pre-surgery (OR= 3.5, 95% CI: [1.3, 9.4]) (Table 5).

DISCUSSION

Findings indicated that function, pain, and mental health scores deteriorated in short-term following surgery, while self-image and satisfaction improved. In long term, all SRS-22r domains improved for up to two years, with the highest improvement in self-image and the lowest in function scores. Using the SDC values, a small number of AIS were assigned to the successful group and had poor scores in all SRS-22r domains preoperatively compared to unsuccessful group. Predictors to attain SDC values for pain and function at 1-year were pre-surgery scores, age, length of hospital stay, and sex.

Relevant changes

Spinal fusion surgery requires muscle dissection and spinal immobilization through rigid internal fixation [37]. Thus, the decrease in functional scores seen immediately post-surgery could be reasonably attributable to post-surgery pain, healing, and reduced spinal mobility [13,38]. The SRS-22r is a widely used PROM for evaluating HRQOL in AIS as well as in adult populations [23]. Our previous review of outcome measures for AIS showed that functional domain of this questionnaire had a high ceiling effect that may limit its ability to detect changes [23]. In this study, the function domain was the farthest domain from achieving the SDC, with increases (+0.2) only at 1 year and (+0.3) at 2 years compared to the other domains. The function was also the least sensitive domain to change in previous studies, attributed to the fact that AIS surgery is mainly cosmetic rather than functional impairment [20]. In contrast to this study, the function domain had the highest improvement (+ 1.2) at the years follow up [25], in which the pre-surgery score was lower than that in this study cohort (median 3.8) compared to (median 3.2) [25]. Although the functional domain of the SRS-22 has been refined to enhance its internal consistency [23], true changes may not be detected, because of the lack of content validity or ceiling effect [16]. Ceiling

effects can make a PROM insensitive to change, as it cannot detect improvements beyond a questionnaire width [39]. Therefore, a PROM that is sensitive to change near the ceiling effects is needed.

Pain was the only domain that worsened at 1 year compared with the other SRS-22r domains. This can be potentially explained by baseline risk factors such as anxiety and pain, which may lead to persistent pain post-surgery [27]. Surgery has also been reported as the leading cause of pain [11], as well as inadequate postoperative pain management [40,41]. The literature suggests that some adolescents develop new-onset pain post-surgery, [12,45] or experience moderate pain 1-year post-surgery [29]. This further supports the need for a better understanding of factors that influence pain.

Self-image showed the highest change at 1-year following surgery. These findings are consistent with those of other studies, where perceived self-image improved immediately following surgery [42], indicating that AIS is principally focused on back shape and spinal deformity [43]. As a result, it is important to manage any misconceptions and unrealistic expectations about the surgery, with those who at risk of post-surgery dissatisfaction, because of their pre-surgery self-image. In this case, they may need additional support to cope with negative emotions associated with self-image.

Poor mental health scores following surgery might be explained through symptoms of depression and isolation and the impact of spine deformity on their mental health domain [6]. A study showed that patients with AIS (n=685) had more suicidal thoughts, concerns about body image and development, and relationships compared to their peers [44]. Therefore, early intervention (pre-16 years) has shown an improvement in psychological well-being of adolescents, supporting the recommendation that surgery should not be delayed, especially in those with poor mental health scores [45].

Participants were satisfied with their management at all time points, with this domain having the highest score (5). With a high ceiling effect in surgically managed participants and among young patients [46], this domain likely needs some revisions for those managed with other approaches.

Comparison analysis

Successful group had poor pre-surgery scores compared to the unsuccessful group in most SRS-22r domains, and they were statistically different. Both groups improved post-surgery compared to baseline scores in all but the function and subtotal domains. Bago et al. reported consistent results for older individuals with AIS (mean age 18.1 years) using the MCID of the subtotal score. The successful group had low pre-surgery scores and significantly improved compared with the unsuccessful group [47]. The high ceiling effects (over 20%) of the SRS-22 were the potential cause of this between group difference [47]. Having high scores close to the end of PROM limits the ability to score a big change, as it outweighs the width of PROM [48]. To ensure that a real change has been detected, re-evaluation of the SRS-22r domain that causes ceiling effect is needed. Findings implies that the unsuccessful group had minimal potential for clinically meaningful improvement. Thus, it is essential for surgeon to discuss surgery's objectives with their patients and, managing expectations which may lead to different treatment strategy.

Predictors of relevant changes

In this study, poor preoperative scores predicted the attainment of SDC values in long-term. Consistent findings showed that inactivity pre-surgery predicted reaching the MCID post-surgery [49]. Factors such as a large curve size >70° [50] and functional limitation [51] are associated with poor preoperative scores.

Age was a significant predictor of SDC pain and function. Previously shown older patients with AIS had poor pre-surgery scores as well as more back pain and cosmetic concerns [44,53]. Those

who underwent surgery at a later stage of their adolescence had more pain than younger patients with AIS [52] and young age could be a protective factor [51]. Our results support previous suggestions of revision of SRS-22r since the ceiling effect is reported more with young AIS, which limits the questionnaire discriminative ability [53].

The length of hospital stay predicted SDC at pain scores. Increased hospital stay was associated with an increase in the amount of instrumentation and operation time [54] and was reduced in those who had an enhanced recovery program after surgery [55]. In contrast, a shorter hospital stay was associated with being active and having better preoperative function scores [49].

Male sex also predicted the SDC value for pain post-surgery. Males are known to have higher functional requirements and expectations than females, which may explain their poor pre-surgery scores. In contrast, females had better outcomes in terms of function and self-image after surgery [55].

Strength and limitations

Strengths include use of SDC values to evaluate real clinical benefit of surgery for AIS, in both short and long terms. Current study used SDC as a cutoff point to classify AIS into groups, based on achievement of real change [6,20,21]. The SDC is more likely to be consistent in individuals with AIS who have had surgery, as there is no evidence that it will differ between surgical and non-surgical or sub-surgical populations [56]. Previous studies used approaches such as activity question of SRS-22r; to classify patients into active and inactive groups [49], or the Minimal Important Change of SRS-22 subtotal score [47]. Future studies may estimate cutoff score of SRS-22r, such as SDC, to enable discrimination between AIS, which would be clinically meaningful.

The main limitation of this study is the high loss at follow up rate, which may introduce bias into the results. Despite this limitation, this study data is based on a registry, indicating that the data

collection process was standardized and comprehensive. Additionally, a reminder system was put in place to encourage participants to complete the follow-up questionnaire. Furthermore, it is worth noting that poor response rates have been reported in previous studies, and this is not an isolated issue [57-59]. There could be various reasons for this, including patients feeling well and not requiring further evaluation, finding alternative treatments, or simply not feeling obliged to complete the questionnaire. Moreover, the low compliance rate could also be related to the design and content of the (PROM) used to collect data [57] . It is important to highlight that a comparison between the study cohort and those lost at follow-up revealed no difference in their pre-surgery characteristics. This indicates that the findings of this study are therefore valid, and the sample is representative of individuals with AIS treated at this spine centre.

Conclusion

Findings indicate that poor domain scores for corrective surgery in AIS, being male, older in age, and increasing length of hospital stay increase the likelihood of having clinical benefit from surgery and attainment of the SDC. The findings demonstrate the utility of the SDC for assessing the benefits and attributable factors for gaining benefits from corrective surgery.

References

1. Bettany-Saltikov J, Weiss HR, Chockalingam N, et al. Surgical versus non-surgical interventions in people with adolescent idiopathic scoliosis. *Cochrane Database of Systematic Reviews* 2015.

2. Tsirikos AI, Roberts SB, Bhatti E. Incidence of spinal deformity surgery in a national health service from 2005 to 2018: an analysis of 2,205 children and adolescents. *Bone & Joint Open* 2020;1:19-28.

3. von Heideken J, Iversen MD, Gerdhem P. Rapidly increasing incidence in scoliosis surgery over 14 years in a nationwide sample. *European Spine Journal* 2018;27:286-92.

4. Vigneswaran HT, Grabel ZJ, Eberson CP, et al. Surgical treatment of adolescent idiopathic scoliosis in the United States from 1997 to 2012: an analysis of 20, 346 patients. *Journal of Neurosurgery: Pediatrics* 2015;16:322-8.

5. Lee AC, Feger MA, Singla A, et al. Effect of surgical approach on pulmonary function in adolescent idiopathic scoliosis patients: a systemic review and meta-analysis. *Spine* 2016;41:E1343-E55.

6. Fernandes P, Do Brito JS, Flores I, et al. Impact of surgery on the quality of life of adolescent idiopathic scoliosis. *The Iowa Orthopaedic Journal* 2019;39:66.

7. Helenius L, Diarbakerli E, Grauers A, et al. Back pain and quality of life after surgical treatment for adolescent idiopathic scoliosis at 5-year follow-up: comparison with healthy controls and patients with untreated idiopathic scoliosis. *JBJS* 2019;101:1460-6.

8. Weiss H-R, Goodall D. Rate of complications in scoliosis surgery–a systematic review of the Pub Med literature. *Scoliosis* 2008;3:1-18.

9. Bastrom TP, Marks MC, Yaszay B, et al. Prevalence of postoperative pain in adolescent idiopathic scoliosis and the association with preoperative pain. *Spine* 2013;38:1848-52.

10. Rullander A-C, Isberg S, Karling M, et al. Adolescents' experience with scoliosis surgery: a qualitative study. *Pain Management Nursing* 2013;14:50-9.

11. Wong G, Yuen V, Chow B, et al. Persistent pain in patients following scoliosis surgery. *European Spine Journal* 2007;16:1551-6.

12. Aghdasi B, Bachmann KR, Clark D, et al. Patient-reported outcomes following surgical intervention for adolescent idiopathic scoliosis: a systematic review and meta-analysis. *Clinical Spine Surgery* 2020;33:24-34.

13. Kakar RS, Simpson KJ, Das BM, et al. Review of physical activity benefits and potential considerations for individuals with surgical fusion of spine for scoliosis. *International journal of exercise science* 2017;10:166.

14. Hughes J, Yaszay B, Bastrom TP, et al. Long-term Patient Perception Following Surgery for Adolescent Idiopathic Scoliosis if Dissatisfied at 2-year Follow-up. *Spine* 2021;46:507-11.

15. Hays RD, Woolley JM. The Concept of Clinically Meaningful Difference in Health-Related Quality-of-Life Research. *PharmacoEconomics* 2000;18:419-23.

16. de Vet HCW, Terwee CB, Mokkink LB, et al. *Measurement in Medicine: A Practical Guide*ed. Cambridge: Cambridge University Press, 2011.

17. Spratt KF. Minimal clinically important difference based on clinical judgment and minimally detectable measurement difference: a rationale for the SF-36 Physical Function scale in the SPORT Intervertebral disc herniation cohort. *Spine* 2009;34:1722.

18. Mokkink LB, Terwee CB, Patrick DL, et al. The COSMIN study reached international consensus on taxonomy, terminology, and definitions of measurement properties for health-related patient-reported outcomes. *Journal of Clinical Epidemiology* 2010;63:737-45.

19. Alamrani S, Rushton AB, Gardner A, et al. Physical functioning in adolescents with idiopathic scoliosis: a systematic review of outcome measures and their measurement properties. *Spine* 2021;46:E985-E97.

20. Kelly MP, Lenke LG, Sponseller PD, et al. The minimum detectable measurement difference for the Scoliosis Research Society-22r in adolescent idiopathic scoliosis: a comparison with the minimum clinically important difference. *The Spine Journal* 2019;19:1319-23.

21. Carreon LY, Sanders JO, Diab M, et al. The minimum clinically important difference in Scoliosis Research Society-22 Appearance, Activity, And Pain domains after surgical correction of adolescent idiopathic scoliosis. *Spine* 2010;35:2079-83.

22. Kelly MP, Kim HJ, Ames CP, et al. Minimum Detectable Measurement Difference for Health-Related Quality of Life Measures Varies With Age and Disability in Adult Spinal Deformity: Implications for Calculating Minimal Clinically Important Difference. *Spine (Phila Pa 1976)* 2018;43:E790-e5.

23. Asher MA, Lai SM, Glattes RC, et al. Refinement of the SRS-22 Health-Related Quality of Life questionnaire Function domain. *Spine* 2006;31:593-7.

24. Glattes RC, Burton DC, Lai SM, et al. The reliability and concurrent validity of the Scoliosis Research Society-22r patient questionnaire compared with the Child Health Questionnaire-CF87 patient questionnaire for adolescent spinal deformity. *Spine* 2007;32:1778-84.

25. Mens RH, Bisseling P, Kleuver Md, et al. Relevant impact of surgery on quality of life for adolescent idiopathic scoliosis. *The Bone & Joint Journal* 2022;104-B:265-73.

26. Djurasovic M, Glassman SD, Sucato DJ, et al. Improvement in Scoliosis Research Society-22R Pain Scores After Surgery for Adolescent Idiopathic Scoliosis. *Spine (Phila Pa 1976)* 2018;43:127-32.

27. Bailey KM, Howard JJ, El-Hawary R, et al. Pain trajectories following adolescent idiopathic scoliosis correction: analysis of predictors and functional outcomes. *JBJS Open Access* 2021;6:e20.

28. Rushton PR, Grevitt MP. What is the effect of surgery on the quality of life of the adolescent with adolescent idiopathic scoliosis? A review and statistical analysis of the literature. *Spine (Phila Pa 1976)* 2013;38:786-94.

29. von Elm E, Altman DG, Egger M, et al. The Strengthening the Reporting of Observational Studies in Epidemiology (STROBE) Statement: guidelines for reporting observational studies. *Int J Surg* 2014;12:1495-9.

30. Asher M, Min Lai S, Burton D, et al. Scoliosis research society-22 patient questionnaire: responsiveness to change associated with surgical treatment. *Spine* 2003;28:70-3.

31. Lenke LG, Betz RR, Harms J, et al. Adolescent idiopathic scoliosis: a new classification to determine extent of spinal arthrodesis. *JBJS* 2001;83:1169-81.

32. Emanuelson U, Egenvall A. The data – Sources and validation. *Preventive Veterinary Medicine* 2014;113:298-303.

33. Razali NM, Wah YB. Power comparisons of Shapiro-Wilk , Kolmogorov-Smirnov , Lilliefors and Anderson-Darling tests, 2011.

34. Howe CJ, Cole SR, Lau B, et al. Selection Bias Due to Loss to Follow Up in Cohort Studies. *Epidemiology (Cambridge, Mass.)* 2016;27:91-7.

35. Kim HY. Statistical notes for clinical researchers: the independent samples t-test. *Restor Dent Endod* 2019;44:e26.

36. Shan G, Gerstenberger S. Fisher's exact approach for post hoc analysis of a chi-squared test. *PLoS One* 2017;12:e0188709.

37. Adogwa O, Elsamadicy AA, Han JL, et al. Do measures of surgical effectiveness at 1 year after lumbar spine surgery accurately predict 2-year outcomes? *J Neurosurg Spine* 2016;25:689-96.

38. Sperandei S. Understanding logistic regression analysis. *Biochemia medica* 2014;24:12-8.

39. Madera M, Brady J, Deily S, et al. The role of physical therapy and rehabilitation after lumbar fusion surgery for degenerative disease: a systematic review. *Journal of Neurosurgery: Spine* 2017;26:694-704.

40. Fabricant PD, Admoni S-h, Green DW, et al. Return to athletic activity after posterior spinal fusion for adolescent idiopathic scoliosis: analysis of independent predictors. *Journal of Pediatric Orthopaedics* 2012;32:259-65.

41. Seki H, Ideno S, Ishihara T, et al. Postoperative pain management in patients undergoing posterior spinal fusion for adolescent idiopathic scoliosis: a narrative review. *Scoliosis Spinal Disord.* 2018;13:17.

42. Fletcher D, Stamer UM, Pogatzki-Zahn E, et al. Chronic postsurgical pain in Europe: An observational study. *Eur J Anaesthesiol* 2015;32:725-34.

43. Carrasco MIB, Ruiz MCS. Perceived self-image in adolescent idiopathic scoliosis: an integrative review of the literature. *Revista da Escola de Enfermagem da USP* 2014;48:748-57.

44. Sanders JO, Carreon LY, Sucato DJ, et al. Preoperative and perioperative factors effect on adolescent idiopathic scoliosis surgical outcomes. *Spine* 2010;35:1867-71.

45. Payne WK, III, Ogilvie JW, Resnick MD, et al. Does Scoliosis Have a Psychological Impact and Does Gender Make a Difference? *Spine* 1997;22.

46. Andersen M, Andersen GR, Thomsen K, et al. Early weaning might reduce the psychological strain of Boston bracing: a study of 136 patients with adolescent idiopathic scoliosis at 3.5 years after termination of brace treatment. *J Pediatr Orthop B* 2002;11:96-9.

47. Parent EC, Hill D, Moreau M, et al. Score distribution of the Scoliosis Quality of Life Index questionnaire in different subgroups of patients with adolescent idiopathic scoliosis. *Spine (Phila Pa 1976)* 2007;32:1767-77.

48. Bago J, Perez-Grueso FJ, Pellise F, et al. How do idiopathic scoliosis patients who improve after surgery differ from those who do not exceed a minimum detectable change? *Eur Spine J* 2012;21:50-6.

49. Copay AG, Subach BR, Glassman SD, et al. Understanding the minimum clinically important difference: a review of concepts and methods. *The Spine Journal* 2007;7:541-6.

50. Raad M, Neuman BJ, Jain A, et al. The use of patient-reported preoperative activity levels as a stratification tool for short-term and long-term outcomes in patients with adult spinal deformity. *Journal of Neurosurgery: Spine* 2018;29:68-74.

51. Tarrant RC, O'Loughlin PF, Lynch S, et al. Timing and predictors of return to short-term functional activity in adolescent idiopathic scoliosis after posterior spinal fusion: a prospective study. *Spine* 2014;39:1471-8.

52. Sieberg CB, Simons LE, Edelstein MR, et al. Pain prevalence and trajectories following pediatric spinal fusion surgery. *The Journal of Pain* 2013;14:1694-702.

53. Rodrigues LMR, Gotfryd AO, Machado AN, et al. Adolescent idiopathic scoliosis: surgical treatment and quality of life. *Acta Ortop Bras* 2017;25:85-9.

54. Parent EC, Dang R, Hill D, et al. Score distribution of the scoliosis research society-22 questionnaire in subgroups of patients of all ages with idiopathic scoliosis. *Spine* 2010;35:568-77.

55. Basques BA, Bohl DD, Golinvaux NS, et al. Patient factors are associated with poor short-term outcomes after posterior fusion for adolescent idiopathic scoliosis. *Clin Orthop Relat Res* 2015;473:286-94.

56. White SF, Asher MA, Lai SM, et al. Patients' perceptions of overall function, pain, and appearance after primary posterior instrumentation and fusion for idiopathic scoliosis. *Spine (Phila Pa 1976)* 1999;24:1693-9; discussion 9-700.

57. Sauerland S, Lefering R, Neugebauer EA. Retrospective clinical studies in surgery: potentials and pitfalls. *J Hand Surg Br* 2002;27:117-21.

Table 1: Characteristics of study participants

Variables	**Study cohort n=124**	**Lost at follow up n=180**	**P value**
Age, median (IQR) (years)	14.9 (1.6)	15.5 (3.3)	.097
Sex, n (%) Female Male	 106 (85.4) 18 (14.5)	 156 (86.6) 24 (13.3)	.467
Major Cobb angle, median (IQR) (degrees)	63.0 (22)	64.4 (28.8)	.769
Lenke classification, n (%) Lenke 1 Lenke 2 Lenke 3 Lenke 4 Lenke 5 Lenke 6	 70 (56.5) 1 (0.8) 17 (13.7) 2 (1.6) 31 (25.0) 3 (2.4)	 93 (51.7) 7 (3.9) 34 (18.9) NA 43 (23.9) 3 (1.7)	 .200
Procedure, n (%) Posterior approach Anterior approach Anterior and posterior approach	 115 (92.7) 7 (5.6) 2 (1.6)	 159 (88.3) 18 (10.0) 3 (1.7)	 .396
Length of hospital stay, Median (IQR)	7(3)	7 (3)	.889
SRS-22r pre-surgery scores, median (IQR) Function Pain Self-Image Mental Health Satisfaction Sub-total Total	 4.0 (1.2) 3.6 (1.4) 2.8 (1.0) 3.8 (1.4) 3.5 (1.5) 3.6 (1.2) 3.5 (1.0)	 3.8 (0.9) 3.6 (1.2) 2.8 (1.0) 3.7 (1.4) 3.5 (1.0) 3.4 (0.9) 3.5 (0.8)	 .037 * .511 .682 .511 .218 .182 .532

n indicates sample size; IQR, Interquartile Range, SRS-22r, scoliosis research society questionnaire-22 revised, * significant at the 0.05 level (2-tailed)

Table 2: Descriptive values of SRS-22r domain scores per time points

SRS-22r domains	Baseline Mean (SD)	6 Weeks Mean (SD)	6 Months Mean (SD)	1 Year Mean (SD)	2 Years Mean (SD)	Change at 1 year	Change at 2 years
Total	3.4 (0.7)	3.5 (0.5)	3.8 (0.6)	4.0 (0.5)	4.1 (0.5)	0.6	0.7
Subtotal	3.5 (0.7)	3.4 (0.7)	3.7 (0.6)	4.0 (0.5)	4.0 (0.5)	0.5	0.6
Function	3.8 (0.7)	3.1 (0.6)	3.6 (0.5)	4.0 (0.6)	4.1 (0.5)	0.2	0.3
Pain	3.5 (0.9)	3.0 (0.9)	4.0 (0.7)	3.5 (0.9)	4.1 (0.7)	-0.1	0.5
Self-image	2.8 (0.8)	3.9 (0.7)	4.0 (0.7)	4.0 (0.7)	4.1 (0.6)	1.2	1.3
Mental health	3.6 (0.9)	3.5 (0.9)	3.8 (0.8)	4.0 (0.8)	4.1 (0.8)	0.4	0.5
Satisfaction	3.3 (1.4)	4.3 (0.8)	4.2 (1.0)	4.3 (0.9)	4.3 (0.9)	1.0	1.0

SRS-22r indicates Scoliosis Research Society-22r; SD standard deviation, CI: confidence intervals.

Table 3: Comparison analysis between successful and unsuccessful groups of SRS-22r domain scores

SRS-22r domain	Successful ≥ SDC n (%) mean (SD)	Unsuccessful < SDC n (%) mean (SD)	P value	SDC
Function	33 (26.6)	91 (73.3)		
Pre-surgery	3.2 (0.7)	4.1 (0.5)	<.001**	
1 year	4 (0.6)	3.9 (0.5)	0.7	0.24
Difference	0.8	0.2	<.001**	
Pain	51 (41.1)	73 (59.3)		
Pre-surgery	3.1 (0.8)	3.9 (0.9)	<.001**	
1 year	4.2 (0.6)	3.7 (0.9)	.003**	0.3
Difference	1.1	-0.2	<.001**	
Mental health	44 (29.6)	54 (70.4)		
Pre-surgery	3.19 (0.7)	4.1 (0.7)	<.001**	
1 year	4.17 (0.7)	3.7 (0.8)	.004**	0.27
Difference	0.9	0.4	<.001**	
Self-image	76 (81.7)	17 (18.3)		
Pre-surgery	2.7 (0.8)	3.4 (0.8)	<.001**	
1 year	4.1 (0.6)	3.3 (0.6)	<.001**	0.3
Difference	1.4	-0.1	<.001**	
Subtotal	59 (63.4)	39 (41.9)		
Pre-surgery	3.2 (0.7)	3.9 (0.6)	<.001**	
1 year	4.0 (0.5)	4.1 (0.5)	0.32	0.23
Difference	0.8	0.2	0.03	

SDC indicates the Minimal Detectable Minimal Difference, n: sample size, SRS-22r: Scoliosis Research Society-22r, SD: standard deviation,

* Significant at the 0.05 level (2-tailed)

** significant at the 0.01 level (2-tailed)

Table 4: Multiple logistic regression analysis for factors predicted **function** outcome† at 1 year follow up

Backward selection method (final model)	**b (SE)**	**p**	**95% confidence interval for Exp (b)**		
Independent factors			**Exp (b)**	**Lower**	**Upper**
Age	0.5 (0.2)	.029 *	1.6	1.1	2.2
Function(pre-surgery)	-2.3 (0.4)	<.001**	0.11	0.1	0.2

† Dependent variables were dichotomized into successful and unsuccessful using SDC value for function (0 .24)

R2 = .578 (Hosmer and Lemeshow), .238 (Cox and Snell), .447 (Nagelkerke)

* p< 0.05

**p<.001

Table 5: Multiple logistic regression analysis for factors predicted **pain** outcome† at 1 year follow up

Backward selection method (final model)	**b (SE)**	**p**	**95% confidence interval for Exp (b)**		
Independent factors			**Exp (b)**	**Lower**	**Upper**
Sex	1.5 (0.7)	.036 *	4.5	1.1	18.5
Age	1.0 (0.3)	<.001 **	2.7	1.7	4.5
Length of hospital stay	0.1 (0.1)	.040 *	1.1	1.1	1.3
Function (pre-surgery)	1.3 (0.5)	.011*	3.5	1.3	9.4
Pain (pre-surgery)	-2.2 (0.5)	<.001**	0.12	0.1	0.3

† Dependent variables were dichotomized into successful and unsuccessful using SDC value for pain (0 .3)

R2 = .107 (Hosmer and Lemeshow), .390 (Cox and Snell), .526 (Nagelkerke)

* P < 0.05

** P <.001

Appendix 2. Alamrani (2023a)

PLOS ONE

RESEARCH ARTICLE

Content validity of the Scoliosis Research Society questionnaire (SRS-22r): A qualitative concept elicitation study

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Abstract

Introduction

Scoliosis Research Society-22 revised (SRS-22r) is the common questionnaire used to evaluate health related quality of life (HRQOL) for young people with adolescent idiopathic scoliosis (AIS). The aim of this study is to evaluate its content validity for this population.

Methods

In-depth semi-structured interviews were conducted with a purposive sample of young people with AIS (Cobb angle ≥25˚, aged 10–18 years). Concept elicitation was used to evaluate the influence of AIS on participants' HRQOL. Participant information sheets and consent/assent forms were age relevant. Topic guide was informed by the SRS-22r and existing evidence. Interviews were audio and video recorded, transcribed verbatim, coded, and analysed using thematic analysis. Derived themes/codes were compared with SRS-22r contents (domains/items).

Results

Eleven participants (mean age 14.9 years [SD = 1.8]; 8 female) were recruited. The mean curve size was 47.5˚ [SD = 18˚] and participants had been managed via different approaches. Four main themes emerged with associated subthemes: 1) Physical effects related to physical symptoms (back hurt, stiffness) and body asymmetry (uneven shoulders), 2) Activity-related effects showed impact on mobility (sitting for long periods), self-care (dressing), and school activities (focus during lessons), 3) Psychological effects revealed emotional (feel worried), mental (sleep quality), and body image effects (hide back from others), 4) Social effects (participation in school and leisure activities), and school, friends and mental health support. A weak association was found between items of the SRS-22r and the identified codes.

Conclusion

The SRS-22r does not adequately capture important concepts that relate to HRQOL of adolescents with AIS. These findings support revision of the SRS-22r, or the development of a new patient reported outcome measure to evaluate HRQOL of adolescents with AIS.

Introduction

Scoliosis Research Society (SRS) questionnaire is the common patient reported outcome measure (PROM) for young people with adolescent idiopathic scoliosis (AIS) [1, 2]. It was developed by Haher et al. in 1999 by incorporating questions from previous questionnaires used to evaluate surgical treatment of adults with scoliosis [3]. The developed questionnaire (SRS-24) was refined and updated to enhance its internal consistency, and two items were removed producing the SRS-22r [4]. The SRS-22r has been included in the Core Outcome Set (COS) for adolescents and young adults with spinal deformity to evaluate self-image, physical functioning and pain [5].

As directed by the initiatives of the Core Outcome Measures in Effectiveness Trials (COMET) consortium, if the SRS-22r has been recommended as a PROM to be selected in a COS, it should exhibit at least high-quality evidence of good content validity [6]. Content validity is the first, and most important measurement property to consider when selecting any PROM [6]. Adequate content validity indicates that the contents of a PROM are consistent with the perspective, words, and experiences of the population of interest [7, 8]. The use of a PROM that does not appropriately reflect the population being measured might undermine the validity of study findings. On the other hand, using a PROM having content validity for the intended purpose and other essential psychometric qualities can lead to advances in theory and practise [9]. Qualitative research methods such as the use of concept elicitation methodology may usefully assess content validity by interviewing the population of interest to understand their perspectives regarding the impact of the disease on their own health condition [10–12].

The psychometric properties of the SRS-22r have been extensively studied [4, 13–15], with the SRS-22r translated and culturally adapted into more than ten languages [1]. However, participants in these studies were older than 18 years, (range 19–34 years), and therefore they may not be representative of an AIS population. Furthermore, the mental health domain of the SRS-22r includes questions from the SF-36 survey, which is a generic questionnaire designed for adults [15]. The SRS-22r is reported to have ceiling effects (20–44%) [4], which limit the reported content validity and reliability [6]. The Scoliosis Quality of Life Index (SQOLI) is another PROM which was developed to address the limitations of the SRS-22r and improve relevance to AIS [16]. However, the SQOLI is not widely utilised, and the SRS-22r remains the commonly used PROM for health-related quality of life (HRQOL) evaluation in people with AIS. Because adolescence is a distinct demographic age group with discrete emotional and social characteristics, it is essential that their PROM represents their developmental stage [17].

A recent scoping review exploring the experiences of AIS and families about the diagnosis and treatment of scoliosis revealed a need for more qualitative research for this population [18]. The majority of studies evaluated decision for treatment [19] and focused on the experience of surgery [20], mainly its effects on the psychological aspects for the patient and parents [21, 22].

To the best of our knowledge, no previous qualitative study has interviewed individuals with AIS to elicit concepts about their scoliosis and their impact on HRQOL. Evidence for

content validity of the SRS-22r is therefore lacking. The aim of this study was to determine whether the content of the SRS-22r questionnaire is relevant and important to participants with AIS, by exploring the impact of scoliosis and its associated treatment on HRQOL.

Methods

Ethics approval

Ethical approval to conduct the study was sought from the Health Research Authority and Health and Care Research Wales approval (REC reference: 21/WM/0076).

Design

This qualitative study design is reported in line with the COnsolidated criteria for Reporting qualitative study [23], and has been described in a published study protocol [1].

Theoretical framework

Phenomenology was used as theoretical framework and adapted grounded theory as the methodology for collecting data and analysis [11, 24, 25]. Phenomenology is a philosophical approach focusing on the experience the individual experiencing the phenomenon [26]. It was used to explore in depth adolescents' lived experiences with idiopathic scoliosis and the related treatments. Adapted ground theory is an approach in which previous knowledge grounded in existing literature and expert opinion is used to develop domains and probes in the topic guide [7]. It provides a framework for data generation and coding which guide analysis process [27]. The existing information about the SRS-22r, was used to inform study design, the topic guide, and to identify themes and concepts (domains and items) and to interpret the results [24].

Participant selection

A heterogenous purposive sampling approach was used to recruit individuals with AIS aiming for n = 10–15, which is deemed sufficient to reach concept saturation [28]. This approach enabled a wide range of experience and opinions to be explored and ensured diversity in the sample characteristics [29]. Participants with a Cobb angle >25˚, aged 10–18 years, and who had access to a video/audio call platform, were considered eligible. Individuals with other forms of scoliosis, and those who were unable to speak English fluently were excluded.

Data collection and setting

Participants were recruited from a tertiary scoliosis centre in the United Kingdom. Due to COVID-19 restrictions semi-structured interviews were conducted virtually (using Zoom/Microsoft Teams platforms). Written consents to participate in the study were collected from participants and parents by the research nurse, during their visit to the clinic. The lead researcher (SA) contacted the participant/parent, and the interview time was chosen based on participant/parent preferences. Data collection took place between January and April 2022.

Topic guide

The HRQOL concept is a "complex, multidimensional concept, including social, emotional and physical functioning or well-being, related to the patient's health state" [30]. To ensure that the concept of HRQOL was elicited from participants during the interview, a topic guide was developed using a hypothesised conceptual framework (Fig 1), as well as the recommended guidelines on the development of PROM for measuring HRQOL [31, 32]. The topic

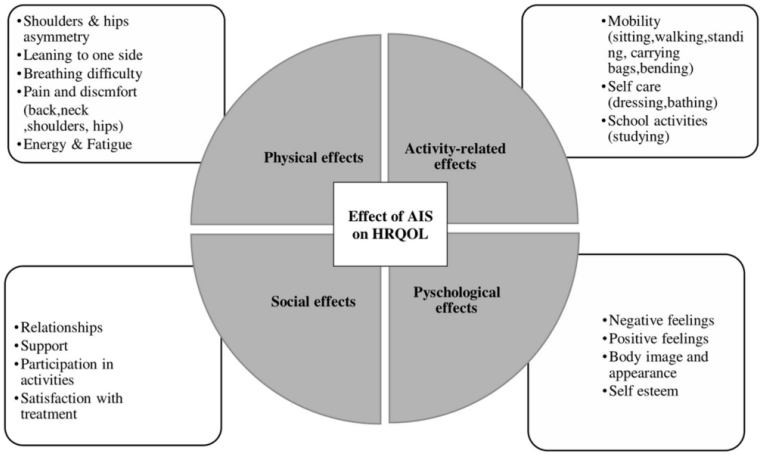

Fig 1. Hypothesised conceptual framework using HRQOL dimensions.

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guide was age appropriate and consisted of open-ended questions that explored concepts of interest (i.e., HRQOL) as well as other concepts from the SRS-22r such as satisfaction. Ended by exploring other areas that are important to participants with AIS, such as participation in sport and other physical activities (S1 Appendix).

Protocol amendments

An amendment was made to the topic guide after piloting with a participant with AIS. The revision was to improve clarity of some questions, as they may lead to confusion. Furthermore, details about confidentiality were explained and simplified.

Interview procedure

At the beginning of the interview, the interview format was explained, and any questions were answered. The confidentiality, voluntary participation and withdrawal process was also explained. Demographic and clinical data were collected electronically before the interviews took place. Participants were encouraged to talk for as long as was needed during the interview, which lasted a mean time of 49 minutes (SD = 9.7). Participants were unknown to the interviewer (SA) prior to the interview in all cases. The interviewer and participants were alone during interview in all but three instances, where the interview was observed by a parent. Interviews were audio and video recorded, and field notes were collected following the interview. Audio recordings were transcribed verbatim by an official transcription service. Interview transcripts were sent to participants to check accuracy with an opportunity for participants to add any further details if required. No revisions or amendments were received.

Research team and reflexivity

An experienced musculoskeletal physiotherapist researcher (SA) conducted the interviews, with support from specialist co-authors in spinal research (surgeon, physiotherapists), and qualitative research. No specific relationship was established prior to the commencement of

the interviews and participants were informed about the professional background of the interviewer and that the study is part of a PhD thesis.

Patient and public involvement (PPI)

This study was conceived as a direct consequence of gaps identified in a previous systematic review of physical functioning outcome measures amongst participants with AIS [1]. The PPI representative was part of the study management group, and her feedback had been sought on the study protocol, the topic guide as well as versions of the participant information sheet and consent forms. Furthermore, the PPI representative was involved in data analysis and interpretation of study findings through provision of a plain English summary.

Data storage and management

All study investigators complied with requirements of the Data Protection Act 2018 with regards to the collection, storage, processing and disclosure of personal information and upheld the Act's core principles. Personal data were coded and depersonalised and replaced with a participant identification number. and stored electronically on a password-protected computer at the University of Birmingham. Secure maintenance of the data ensured that the linking code were kept securely in a separate location using encrypted digital files within password- protected folders and storage media. Only the Chief Investigator and the study Research Fellow had access to the data as necessary for quality control, audit and analysis. Data will be stored for 10 years in line with the University of Birmingham's Research Governance procedures only accessible for the research team.

Data analysis

Data were analysed according to the recommended guidelines for the evaluation of the content validity of a PROM [7, 8, 24], with guidance from thematic analysis [33]. A coding framework was developed using the topic guide and the hypothesised conceptual framework [24]. Coding was performed by SA with guidance from the co-authors. A saturation table (S2 Appendix) was created to ensure that concept saturation was achieved, at the point where two consecutive interviews failed to elicit any new themes [28]. To ensure that the results were representative of the concept of interest among different participant characteristics or subgroups [8]. Coding was stratified according to participant characteristics (i.e., curve severity and management approach). Deductive analysis was used to organise the data into themes, which were predetermined by the topic guide or emerged from the data related to the concept [7]. Themes that emerged were mapped to the SRS-22r contents (domain and items) [8]. The words and phrases in the derived themes and subthemes were compared to words used in the SRS-22r questionnaire [7, 8]. If a poor agreement was present, indicating inadequate coverage of the concept of interest, an adaption to the PROM, or the development of a new PROM, should be made to support its use [8].

Findings

Participants

Demographic and clinical characteristics of participants are presented in Table 1. From 19 recruited and consented participants, 11 took part in the study. Four participants could not be contacted because they did not respond to the invitation from the researcher. Two withdrew due to school commitments and one withdrew because of parental withdrawal of consent. One participant withdrew once the interview had commenced as they denied speaking and

Table 1. Participants demographic and clinical characteristics.

ID	Age (years)	Gender	Curve severity	Location of curve	Lenke type	Management	Duration Following surgery	Number of X-rays	Pain intensity (0–10)	Duration of pain	Physical activity level per day	SRS-22r score Mean (SD)
1	15	Female	51˚	Thoracic	1	Observation	NA	2	5	> 6 mos.	< 60 min.	3.2 (.89)
2	17	Female	32˚*	Thoracic	3	Post-surgery	20 mos.	12	5	> 6 mos.	> 60 min.	3.7 (.72)
3	12	Female	31˚	Thoracolumbar junction	5	Brace	NA	5	3	> 6 mos.	> 60 min.	4.4 (.46)
4	12	Female	25˚	Thoracolumbar junction	5	Observation	NA	1	0	-	> 60 min.	4.6 (.81)
5	16	Female	40˚*	Thoracic	1	Post-surgery	10 mos.	8	5	> 6 mos.	60 min.	4 (.76)
6	17	Male	57˚	Thoracic	1	Pre-surgery	NA	6	4	> 6 mos.	< 60 min.	3.3 (.71)
7	16	Female	70˚	Thoracic	1	Pre-surgery	NA	2	8	> 6 mos.	< 60 min.	3.7 (.52)
8	16	Female	70˚	Thoracolumbar junction	1	Post-surgery	3 mos.	11	4	> 6 mos.	< 60 min.	3.4 (.68)
9	14	Male	40˚	Thoracic	1	Brace	NA	2	3	3–6 mos.	< 60 min.	4.1 (.68)
10	13	Female	86˚	Thoracic	1	Pre-surgery	NA	3	7	> 6 mos.	> 60 min.	2.8 (.66)
11	16	Male	40˚	Thoracic	1	Pre-surgery	NA	2	7	> 6 mos.	60 min.	3 (.52)

* Indicates curve severity post-surgery

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participating because of feeling shyness. Therefore, their demographic data were discarded. The sample was representative of the population of adolescents with AIS, with the majority being female (n = 8), aged between 10–18 years old.

A slight modification was made to the hypothesised conceptual framework (Fig 1), when compared to the updated conceptual framework (Fig 2) that was developed from the data. The identified themes were: (1) physical effects, (2) activity-related effects), (3) psychological effects, and (4) social effects. Some of the hypothesised subthemes were discarded as no data were collected to support their inclusion. Each theme and subthemes with appropriate code (participants quotes) are presented in (S3 Appendix).

Physical effects

This theme was consistent with hypothesised subthemes across the two conceptual frameworks. In addition to various new subthemes were identified.

Physical symptoms. Back hurt was the main physical symptom reported by study participants. Those with severe curves expressed high pain intensity compared to participants with milder curves. Pain was associated with exercise performance such as stretching exercises as a form of sport (e.g., yoga), or in a form of treatment (e.g., physiotherapy). Participants noted that exercise was the leading cause of pain and the need for subsequent analgesia, which affected their participation in sport or physical education in school.

> *"Sometimes it hurts my back when I do some exercises.., when I play football, it hurts my back"* (P11-pre-surgery)

Management of AIS (e.g., surgery) contributed also to the feel of pain. It affected school attendance, performance and the need for analgesia for several months following surgery. Bracing also can contribute to the experience of pain, and it was described as uncomfortable, and causes bruises.

> *"Whereas now if I do get pain, it's because of the surgery" (*P8-post surgery)

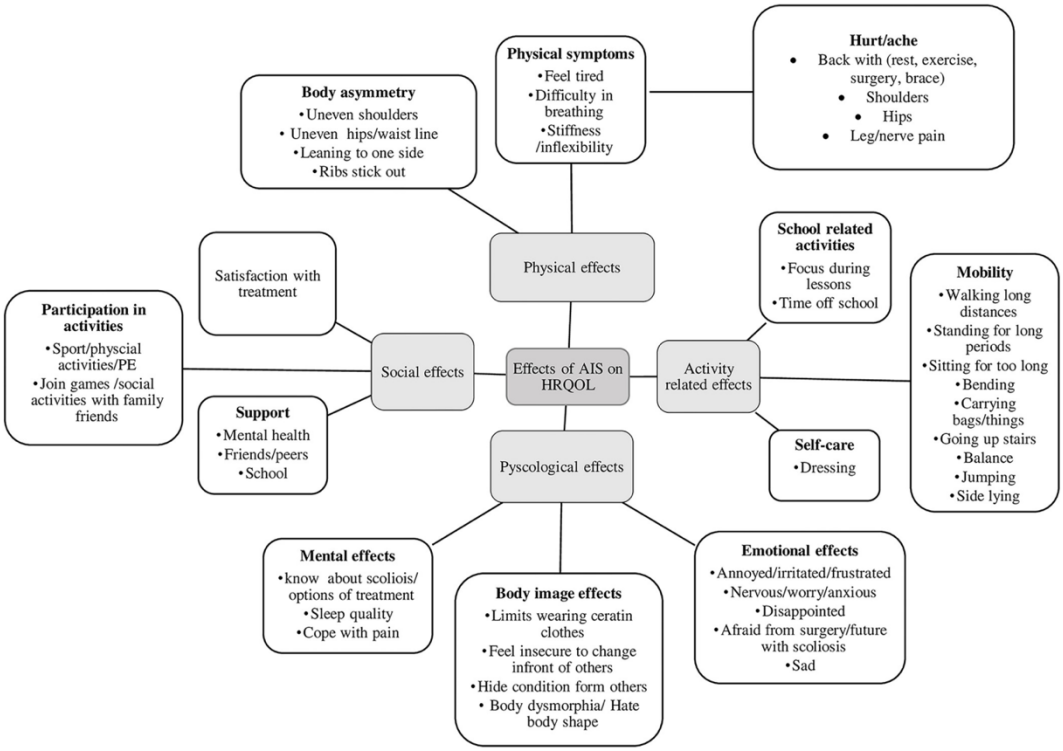

Fig 2. Updated conceptual framework.

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Participants also reported shoulder and hip pain, which was associated with body asymmetry. Shoulder and hip asymmetry were thought to affect being able to find a comfortable sleeping position and having a good quality of sleep.

"I think just my shoulders and my hips sometimes because it's kind of like disjointed everything. They ache most of the time and no matter how many medications I can take for it, it will always be there because it just reoccurs" (P1-observation)

Nerve or leg pain was also reported by those with severe curves and waiting surgery, contributed to the feeling of weak legs and the increased need of taking pain medications.

*"When I get nerve pain, I have to take three [painkillers] to kill it" (*P10-Pre-surgery).

Participant reported feeling more tired when comparing themselves to their peers at school, and they felt that they needed to take more breaks in the day. Difficulty in breathing was also reported especially with running, and among those with severe curves.

"I definitely feel more tired than other people after I do a lot of things and I have to take more breaks than most of my friends when they do the same things" (P1-Observation).

Our study participants have not reported any limitation in range of movements; however, the terms "stiffened", and 'inflexibility' were used by some.

"I'm less flexible; I've never really been that flexible. I used to be really; I could do anything. But now it feels kind of stiffened and I've definitely lost quite a lot of flexibility" (P1-observation)

Body asymmetry. Participants did not see or report a back hump that reflects the asymmetry of the posterior torso and is a physical sign of scoliosis, whilst a parent had noticed it. They noted different shoulder and hip levels, and with severe curves they noticed the ribs being more prominent. They also reported feelings of leaning over to one side.

"My hips are about five centimetres apart so like one's there, one's there. . ." (P10-presurgery)

Activity-related effects

Three main themes were identified related to the effects of scoliosis on activity (mobility, self-care, school related activities). Compared with the hypothesised framework, multiple sub-themes emerged from collected data for mobility but not for self-care.

Mobility. Sitting for too long was the frequently reported mobility problem by participants. They described that they cannot sit for a long time without feeling back pain and discomfort. This often led to a change in their position, either by standing or moving around. This was problematic in school, especially with lessons of a long duration without breaks, or where chairs had no back support. Further, standing for long time" and "walking for long distances" were also reported as the cause of back pain. Some participants avoided joining activities with friends and family that require walking a long distance.

"Sitting down because I have two-hour lessons, instead of one hour. And sitting down for two hours is uncomfortable". . . "*Sometimes, I'm allowed a break like 10 minutes in between the lesson. But sometimes, I don't, and it can get kind of painful"* (P6-Presurgery)

Bending activities (i.e., picking up things from floor or vacuuming) was described as painful and challenging activity. Those who had surgery or wore brace found that it was difficult to tie their shoelaces or put socks on. Avoidance behaviour, asking for help or finding alternative ways to perform the activity was reported.

"When I keep having to bend down and pick things up, it's quite hard to sometimes clean my room and especially vacuum. . ..and it hurts quite a lot at the end" (P1-Observation)

"Bending over, that kind of thing; I tend to avoid bending over" (P2-post surgery).

Activities such as jumping and running at a pace that is more than a jog, were reported as difficult, and would be avoided.

"The first time I realised that something was quite painful about it was when I was out with friends at a trampolining place. I started to jump,.this was the only time that it actually started to hurt. And I had to stop" (P1-Observation)

Participants found that "carrying things" such as school bags could increase of back discomfort. Other reported that scoliosis "hindered their balance", which make them feel dizzy

and unable to walk in a straight line. Some adolescents reported that they were unable to go upstairs, and they have been allowed to use the lift instated of stairs at school. Side lying and difficulties finding a comfortable position have an impact on sleep quality as well.

"Walking in straight. . . I can't walk in like. . . well, I feel like I'm not walking in a straight line" (P7- Pre-surgery)

"Sometimes I do think it hinders my balance as well, so maybe that's why I can't stand up for very long periods of time without feeling dizzy." (P1- Observation).

Self-care. Dressing was the only challenging activity identified in this theme, causing discomfort for some participants. Activities such as washing oneself was discarded as no data supported its inclusion. Those who had undergone surgery reported needing help in self-care activities in early post-surgery period, but not after several months following surgery. Dressing also included tying shoes laces and putting socks on, which remained challenging following surgery.

"Dressing, putting my socks on can be a bit of a task, or pulling up my trousers, but it's not that hard that it's a problem" (P8-Post-surgery)

School-related activities. Participants reported that their focus and concentration during lesson time was impacted because of pain they felt with prolonged sitting. Thinking about pain and changing sitting position to find a comfortable spot affected concentration. Also, wearing a brace increased their discomfort and further affected their ability to focus.

"Sometimes it's definitely difficult to focus when my back is hurting that much and I'm trying to get on with the lesson. It is worse in science because we have these chairs, they don't have a back on and so I have nothing to lean against. It makes me struggle because I'm trying to focus, but then trying to make sure that my back is not bending or hunching over because it would hurt more". (P1-Observation)

Participants reported that pain, surgery, and the follow-up appointments affected school attendance and performance. Surgery alone led up to 5 months away from school for one participant. For another participant she decided not to carry on schooling, because she had missed a lot of school days due to surgery.

"It did affect my GCSEs because I literally had it done right before my exams. I had two months off school, . . .so I obviously missed a lot of school..now I still have follow-up appointments, obviously you miss time off school.. so it does add up" (P5-Post-surgery)

Psychological effects

Three main subthemes were identified related to this theme. Hypothesized factors such as self-esteem and positive feelings has been discarded, as no data supported its inclusion.

Emotional effects. Many terminologies were used interchangeably by participants with AIS to describe their negative feelings and emotions about scoliosis and it associated treatment (Fig 2). While no positive feelings were elicited. Participants used terms like "sad" when they felt pain, when they couldn't explain their feelings to others, or when they think about surgery.

"It makes me sad because it reminds me of being in pain a lot before the surgery, because the biggest thing I wanted out of the surgery was to help my pain, even if it was just a little bit". (P8-post-surgery)

Feelings nervous, worried and anxious were used when they compared themselves with others, or when thinking about the need of surgery and how it will affect their future life. They felt worried also because their parents felt worried too.

"I feel a bit worried, like how it will affect me in the future and how it's going to impact on my life. Just a bit nervous about if it will get worse in the future or not" (P11-pre-surgery)

Participants reported feelings of being "annoyed" and, "irritated" because of having scoliosis or because they still have back problems following surgery. Wearing a brace made them feel annoyed as well. Feeling "afraid" was used when describing their emotions to have surgery, or that they might need surgery in the future. Disappointed was expressed when they believed there was nothing that could be done to treat their condition.

"It can make me feel very annoyed, like it's just a bit like why me? Why?" (P10-presurgey)

"I feel quite disappointed that there can't be anything done. And I understand why there cannot be anything done. Yeah, I think just disappointment is the biggest one" (P1- Observation)

Body image effects. This theme has a significant impact on participants lives and many subthemes were found. Participants reported feeling insecure about their body shape, making them unable to change when being with their friends or peers. They tend to hide their back from others by wearing large clothes. They felt that scoliosis limits wearing certain clothes such as a swimming kit, which limits participation in swimming. The term "dysmorphia" was used by one participant describing how she feels about her body shape.

*"I don't know if this is the right thing, but it's getting changed in front of people, I can't do that and that's the issue usually also in PE, . . . Or it's when I'm with friends and we're all getting ready for going out . . .. I just can't get changed in front of them" (*P1- Observation).

This was also associated with preference of some of participants with AIS, to hide their condition from others.

"I didn't tell anyone about it because I didn't like it. . . I distanced myself from people.. I just wanted to be on my own. But it's fine now" (P5-Postsurgey).

Mental effects. Participants described different methods to cope with pain, including taking analgesia, changing positions, and taking rest when pain was associated with activity.

*"It's usually just a case of I just have to lie down for a minute. Obviously when I'm resting it doesn't hurt, that's literally the way to quickly cure it, is lie down" (*P5-Post-surgery).

Participants reported that they felt shocked and confused, because of the lack of knowledge about scoliosis and its treatment. They reported that they felt anxious and worried which affect quality of sleep.

"I kind of had a stage of denial, I didn't believe that it was real, and I felt that they were wrong. But then I saw the x-rays and I was shocked, to be honest. I didn't realise what it actually looked like and what it was meant to look like basically" (P1-Observation).

Social effects

Participation. Sport and exercise participation was limited or discontinued due to back discomfort. Likewise, following surgery, some people were recommended to avoid contact sports, therefore their involvement in sports was limited.

"I've always been a big sporty person, which is why it impacted me so much having scoliosis. I went from being very competitive and fast and loved sports to not being able to participate" (P8-Posturgery).

Scoliosis also has an influence on "recreation and leisure" activities with family and friends. Going out with family and friends was difficult because of the severe pain.

"*Sometimes. When they were going trampolining, and I couldn't't go because it hurt that much. Or when it's a birthday party and everybody is doing piggybacks and I have to say, hmm, maybe that's not a good idea"* (P1-Observation).

Support. Friendship support was found to be important to participants and would assist in mental health, particularly during the recovery time following surgery.

"My school has been amazing.. has provided a chair with lumbar support. It's a very comfortable chair for me to sit in in exams, so I don't have to sit for two hours to do a mock on an uncomfortable chair." (P8-Post-surgery).

"Mental health support" has been shown to be necessary since scoliosis has various effects on the psychological part of AIS life.

"I think, definitely, the treatment you get post-op; you don't get any mental support, and I think there should be more of it" (P2-Post-surgery)

Satisfaction. Treatment satisfaction was essential to study participants. They felt that having observation without intervention was frustrating, and that performing exercises was not an effective treatment. Some were dissatisfied when surgery was the only possible option associated with long waiting periods.

"*So, I literally got diagnosed, they were like, "You've got this," and then they were like, "We can't help you with anything and you've just got to wait for surgery." I've basically been given nothing, absolutely nothing"* (P10-Pre-surgery).

Stratification analysis

Table 2 shows the stratification of data based on the participant's management approach (i.e., observation, brace, pre-/ post-surgery). Although, some codes are not expressed by some

Table 2. Stratification of subthemes according to participants management approach.

Subthemes	Observation	Brace	Pre-surgery	Post-surgery
1a. Back hurt at rest	✓			✓
1b. Back hurt with brace		✓		
1c. Back hurt when exercising	✓	✓	✓	✓
1d. Back hurt because of surgery				✓
Hips hurt	✓	✓	✓	
leg ache/nerve pain			✓	
Shoulder aches	✓		✓	
Feel stiffened/inflexible	✓		✓	✓
Difficult breathing			✓	
Feel tired	✓		✓	
Uneven Shoulders	✓	✓	✓	✓
Uneven hips/waist	✓	✓	✓	
Leaning to one side			✓	
Ribs stick out			✓	
Time off school		✓	✓	✓
Focus during lessons	✓	✓	✓	
Ache with dressing	✓	✓	✓	✓
Jumping/jog	✓		✓	
Hinder balance/walking straight	✓		✓	
Bending	✓	✓	✓	✓
Carrying bags/things	✓		✓	
Going up stairs			✓	
Walking long distances			✓	
Sitting for long time			✓	✓
Side lying	✓	✓	✓	
Stand for too long	✓		✓	✓
Annoyed-irritated-frustrated	✓	✓	✓	✓
Sad	✓		✓	✓
Disappointed	✓			
Nervous-worry-anxious	✓		✓	✓
Afraid	✓		✓	
Feel insecure to change in front of others	✓		✓	
Hate body shape/dysmorphia			✓	
Limit wearing certain clothes			✓	
Hide back condition			✓	
Pain coping	✓	✓	✓	✓
Know about the scoliosis	✓	✓	✓	✓
Know about options of treatment	✓	✓	✓	✓
Sleep quality			✓	✓
School support		✓	✓	✓
Friends support	✓		✓	
Mental health support		✓	✓	✓
Participation In sport/physical activities/physical education	✓	✓	✓	✓
Participation in games /social activities with family friends	✓		✓	
Satisfaction about given treatment	✓	✓	✓	✓

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participants it could be returned to individual variation in experience and personality. Overall, the majority of codes are expressed by participants across the three-management approach.

Comparison analysis

Table 3 presents the comparison and association performed between the codes elicited from the interviews and the items of the SRS-22r. Only one question from the SRS-22r was linked to the physical effects codes. Whereas other physical symptoms and body asymmetry were not linked to the any of the SSRS-22r items. Similarly, for activity effects only one question from the SRS-22r was linked to the codes, while three questions were linked to psychological effects cods and two questions to the body image codes. Participation was also linked to the SRS-22r and the satisfaction theme. Only 10 questions from the SRS-22r were connected to the codes raised from the qualitative data. This may indicate that the SRS-22r may not accurately reflect the perspectives of those with AIS.

Discussion

This is the first qualitative study that has used in depth semi-structured interviews to evaluate the impact of scoliosis on the HRQOL of adolescents with AIS. The study was designed to evaluate the content validity of a currently used PROM (i.e., the SRS-22r) through a concept elicitation format, focusing on the adolescent experience with scoliosis and its associated treatment. Previous research studies have used quantitative methods to evaluate the validity of the SRS-22r [13, 14], with no attention being given to qualitative approaches. This study used words and phrases from participants with AIS in the generation of concepts, to ensure that the data are a true reflection of participants' perspectives. When the qualitative data from participants with AIS was compared to the contents of the SRS-22r, a poor match was found indicating a lack of content validity of the SRS-22r. Furthermore, the terminology used in the SRS-22r does not reflect the language used by participants with AIS. Findings from this study provide strong evidence for the need to revise the SRS-22r, or development of a new PROM using data similar to that generated from this study that represents the voice of adolescents with AIS and is language and age relevant.

Physical effects

Back hurt/pain was the most reported physical effect of scoliosis on HRQOL of participants, consistent with all previous reviews, where the prevalence of back pain amongst adolescents with AIS ranges between 37% to 42% [34]. Performing exercise improves HRQOL of AIS, by improving pain and function [35, 36]. However, most participants experienced back pain when exercising, which affected their adherence and participation in sport and PE at school. Consistent with previous studies, surgery contributed to back pain which may extend to two years post-surgery [37, 38]. This has been identified as the major concern in the presurgical period [22, 39]. The adolescents in this study used terms such as "hurt" or "ache" rather than "pain", to describe their pain experience. This is not reflected in the terminology used by the SRS-22r [4], which may not be representative of this category of population. An important aspect of content validity is that the PROM should be age and language relevant [17, 40]. Other symptoms were also reported such as difficulty in breathing, stiffness, hip, and shoulder pain in accordance with earlier reports [22, 41, 42]. However, these symptoms are not assessed by the SRS-22r. Body asymmetry was also reported by participants as a major concern consistent with previous studies, where body asymmetry associated with scoliosis had a social and psychological impact on life [43]. This aspect is not assessed by the SRS-22r. These findings

Table 3. Comparison between SRS Scoliosis Research Society (SRS-22r) contents with themes, and subthemes elicited from interviews.

Qualitative data		SRS-22r
Themes	**Subthemes**	**Questions**
Physical effects	1a. Back hurt at rest	8- Do you experience back pain when at rest?
	1b. Back hurt with brace	
	1c. Back hurt when exercising	
	1d. Back hurt because of surgery	
Physical Symptoms	Hips hurt	
	leg ache/nerve pain	
	Shoulder aches	
	Feel stiffened/inflexible	
	Difficult breathing	
	Feel tired	
Body asymmetry	Uneven Shoulders	
	Uneven hips/waist	
	Leaning to one side	
	Rips stick out	
Activity-related effects	Time off school	17-In the last 3 months have you taken any days off work, including household work, or school because of back pain?
School-related activities	Focus during lessons	
Self-care	Ache with dressing	
Mobility	Jumping/jog	
	Hinder balance/walking straight	
	Bending	
	Carrying bags/things	
	Going up stairs	
	Walking long distances	
	Sitting for long time	
	Side lying	
	Stand for too long	
Psychological-related effects	Annoyed-irritated- frustrated	
	Sad	7-In the past 6 months have you felt so down in the dumps that nothing could cheer you up?
	Disappointed	16-In the past 6 months have you felt down hearted and blue?
Emotional effects	Nervous-worry-anxious	3-During the past 6 months have you been a very nervous person?
	Afraid from surgery/future with scoliosis	
Body image	Feel insecure to change in front of others	
	Hate body shape/dysmorphia	4- If you had to spend the rest of your life with your back shape as it is right now, how would you feel about it? 19. Do you feel attractive with your current back condition?
	Limit wearing certain clothes	
	Hide back condition	
Mental effects	Coping with pain	
	Know about scoliosis	
	Know about other treatment options	
	Sleep quality	
Social effects	Friends' support	
Support	School support	
	Mental health support	

(*Continued*)

Table 3. (Continued)

Qualitative data		SRS-22r
Themes	**Subthemes**	**Questions**
Participation	Sport/physical activities/physical education	
	Join games /social activities with family friends	18-Does your back condition limit your going out with friends/family?
Satisfaction	Satisfaction about given treatment	21-Are you satisfied with the results of your back management? 22-Would you have the same management again if you had the same condition?

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reflect the strength of this study in using a concept elicitation interview to capture participant's own perceptions of their condition to inform content validity [24].

Activity-related effects

Carrying out normal daily activities such as sitting, standing and walking, if performed for a long time, was reported to aggravate symptoms. These activities were reported in a previous qualitative study in a presurgical period [22]. Scoliosis also affects standing stability and balance [44]. Walking in a straight line was altered because of scoliosis, which has been assessed in earlier studies [45]. Bending was limited generally, and for those who had a surgery in particular, since spinal mobility and flexibility is reduced following surgery [46, 47]. Participants reported discomfort when carrying backpacks, which has been explained by the increase in the compressive forces on the curve apex [48], that may cause additional strains on muscles around the spine. Jumping was avoided as they realised that it caused back pain [22]. Going up stairs was also reported as difficult, although it is not reflected in any scoliosis literature. The association between performing functional tasks such as climbing stairs and having back pain has been previously noted in other groups [49]. Participants described that their focus during lesson time was affected because of a feeling of back hurt during sitting for prolonged time. A study on the association between pain and functioning in school found that adolescents with pain were more likely to have low school engagement, attendance and performance [50]. Participants also reported that they missed school days because of feelings of pain, surgery or hospital appointments which affected their academic achievements. In some countries, surgery is performed during summer time to reduce distribution on school performance [51]. However, for some people with AIS, time off school may extend to 6 months [12, 52]. Regarding self-care, participants reported that dressing caused aching and was a challenging. In summary, these findings demonstrated the significance of these aspects to HRQOL of adolescent with AIS, and therefore should be reflected in a PROM that evaluates HRQOL of this population.

Psychological effects

The emerged themes revealed that scoliosis has a significant impact on the psychological aspect of individuals with AIS life. Different negative feelings were reported associated with both diagnosis and treatment. Feelings of frustration, irritation, and annoyance as well sadness, when experiencing pain were described by participants [12]. Disappointment was also discussed, relating to the treatment decisions, as mentioned in a recent review [18]. Worry and fear about the need for surgery and a future living with scoliosis contributed to feelings of nervousness and anxiety, as discussed before [18, 22, 53], and is also reported by the parents of adolescents with AIS in other studies [19, 21, 39]. Different questions in the SRS-22r evaluated mental health of adolescents, however, they are driven from the SF-36 which is an adult rather than a paediatric questionnaire [1].

The effects of scoliosis on body image of adolescents with AIS is another important factor found is this study. Participants disclosed feeling insecure to change in front of others and hid their back. Scoliosis also limited their ability to wear certain clothes [18, 22]. Body dysmorphia and hate of body shape were raised by one participant with a severe curve. This was associated with dissatisfaction about body image [54]. The self-image domain in the SRS-22r assesses the effect of AIS on body image [4]. Only two questions from this domain seems relevant to the driven subthemes.

A pain coping subtheme was revealed by our participants. A prior study found that coping with pain is dependent on the personality of the individual and affects the recovery process [51]. Since the HRQOL of adolescents with AIS may be related more to psychosocial effects than to physical effects and its consequences [55], it is recommended to train clinicians who are caring of individuals with AIS, in stress reducing techniques including pain coping strategies [51, 56]. Information about scoliosis, and the options for treatment and associated consequences, has a psychological impact on participants life. This has been studied among adolescents with AIS and their parents prior to surgery [22, 39], wherein providing support and information to adolescents and their families helps in reducing stress and anxiety associated with scoliosis [18, 51].

Participants also reported that their sleep quality was affected by scoliosis. A recent study assessed the sleep profile of adolescents with AIS and revealed poor sleep quality associated with high pain intensity [57]. Furthermore, following surgery reports indicated anxiety, nightmares and sleeping difficulties, both after the hospital visit and for a long time after the recovery period [20]. These results highlight the significant impact of scoliosis on the psychological aspects of life with AIS and therefore, should be included in a PROM evaluate their HRQOL.

Social effects

Participation in activities of daily life is important for the physical and psychological development of adolescents [58]. In this study, participants reported reduced participation in social and sport activities consistent with previous studies [59]. Fear of pain or injury, and reduced self-image, may limit social participation, and has a negative impact on wellbeing [20]. Spinal fusion surgery has been shown to have a long term effect on an individuals' social life, and that providing support in the form of information and coping techniques increased the level of social participation [51]. In our study, participants reported the need for mental health support to overcome the psychological consequences of scoliosis and surgery. Earlier studies recommended providing psychological support to adolescents with AIS and their parents to minimize stress and uncertainty about surgery [20, 21]. Satisfaction about the treatment, assessed based on the answers of the SRS-22r was relevant to our study participants. Two questions in the SRS-22r assess satisfaction and were linked to the identified themes.

Strengths and limitations

This study is reported in line with published guidance of qualitative research (COREQ) [23], and is based on a published protocol [1] to ensure rigor and comprehensiveness of the findings. Findings of this study using concept elicitation format allow capturing participant's own perceptions of their condition to inform content validity [24]. Furthermore, patient perspective was central in this study, through involvement of PPI representative (ER) in the study design and interpretation of the results.

Limitations are in a number of areas with several factors that have influenced data collection and analysis, and thus may influence the findings reported. The sample was recruited from a single hospital in the UK limiting transferability of the findings. However, we assured

diversity in population characteristics using a heterogenous purposive sampling technique [29], which ensures the recruitment of participants with various demographic, clinical characteristics, and treatment approaches. The children/adolescents were not talkative, tended to give short answers and were reluctant despite prompting to elaborate on their responses and give further details. The assessment of concept saturation was performed following data collection, although, it has been recommended to be evaluated during data collection [28]. Coding of data was performed by one researcher (SA) and evaluated later by the research team. However, the coding framework and the codebook provide an evidence that coding is a true reflection of participants quotes [24].

Implications for practice and future research

The findings highlight that the content of the SRS-22r does not adequately capture the experience of adolescents with AIS and the effect of scoliosis on adolescent HRQOL. Themes and codes driven from participant data should inform the adaptation of the SRS-22r, or the development of a new PROM, which is relevant to AIS and based directly on qualitative input from adolescents. Furthermore, qualitative research is needed to assess the content validity of the developed PROM through conducting cognitive testing to test its relevance, comprehension, and comprehensiveness from both adolescent and practitioner perspectives [6].

Conclusion

Concept elicitation interviews with adolescents with AIS revealed that scoliosis and its associated treatment has a broad impact on life, including physical, activity-related, psychological, and social effects. Evaluation of content validity of SRS-22r by comparing between the concepts and codes elicited from interview data, and the SRS-22r contents, revealed that the SRS-22r does not accurately reflect the words and phrases of adolescents with AIS, indicating a lack of its content validity. Themes and subthemes that have resulted from this qualitative study could be used to develop a new PROM for AIS or update the existing SRS-22r to enhance its use for this population.

Supporting information

S1 Appendix. Topic guide.
(PDF)

S2 Appendix. Saturation table.
(PDF)

S3 Appendix. Themes, subthemes, codes.
(PDF)

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References

1. Alamrani S, Gardner A, Falla D, Russell E, Rushton AB, Heneghan NR. Content validity of Scoliosis Research Society questionnaire-22 revised (SRS-22r) for adolescents with idiopathic scoliosis: protocol for a qualitative study exploring patient's and practitioner's perspectives. BMJ Open. 2021; 11(12): e053911. Epub 20211214. https://doi.org/10.1136/bmjopen-2021-053911 PMID: 34907066; PubMed Central PMCID: PMC8672051.
2. Alamrani S, Rushton AB, Gardner A, Bini E, Falla D, Heneghan NR. Physical Functioning in Adolescents With Idiopathic Scoliosis: A Systematic Review of Outcome Measures and Their Measurement Properties. Spine 2021. Epub 2021/01/27. https://doi.org/10.1097/BRS.0000000000003969 PMID: 33496543.
3. Haher TR, Gorup JM, Shin TM, Homel P, Merola AA, Grogan DP, et al. Results of the Scoliosis Research Society instrument for evaluation of surgical outcome in adolescent idiopathic scoliosis: a multicenter study of 244 patients. Spine. 1999; 24(14):1435.
4. Asher MA, Lai SM, Glattes RC, Burton DC, Alanay A, Bago J. Refinement of the SRS-22 Health-Related Quality of Life questionnaire Function domain. Spine 2006; 31(5):593–7. Epub 2006/03/02. https://doi.org/10.1097/01.brs.0000201331.50597.ea PMID: 16508558.
5. de Kleuver M, Faraj SSA, Holewijn RM, Germscheid NM, Adobor RD, Andersen M, et al. Defining a core outcome set for adolescent and young adult patients with a spinal deformity. Acta Orthop. 2017; 88 (6):612–8. Epub 2017/09/16. https://doi.org/10.1080/17453674.2017.1371371 PMID: 28914116; PubMed Central PMCID: PMC5694805.
6. Terwee C, Prinsen C, Chiarotto A, Westerman M, Patrick D, Alonso J, et al. COSMIN methodology for evaluating the content validity of patient-reported outcome measures: a Delphi study. Quality of Life Research. 2018; 27. https://doi.org/10.1007/s11136-018-1829-0 PMID: 29550964
7. Brod M, Tesler LE, Christensen TL. Qualitative research and content validity: developing best practices based on science and experience. Qual Life Res. 2009; 18(9):1263–78. Epub 2009/09/29. https://doi.org/10.1007/s11136-009-9540-9 PMID: 19784865.
8. Rothman M, Burke L, Erickson P, Leidy NK, Patrick DL, Petrie CD. Use of existing patient-reported outcome (PRO) instruments and their modification: the ISPOR Good Research Practices for Evaluating and Documenting Content Validity for the Use of Existing Instruments and Their Modification PRO Task Force Report. Value Health. 2009; 12(8):1075–83. Epub 2009/10/07. https://doi.org/10.1111/j.1524-4733.2009.00603.x PMID: 19804437.
9. Vogt D, King D, King L. Focus Groups in Psychological Assessment: Enhancing Content Validity by Consulting Members of the Target Population. Psychological assessment. 2004; 16:231–43. https://doi.org/10.1037/1040-3590.16.3.231 PMID: 15456379
10. Holman HR. Qualitative inquiry in medical research. Journal of Clinical Epidemiology. 1993; 46(1):29–36. https://doi.org/10.1016/0895-4356(93)90006-m PMID: 8433110
11. Brédart A, Marrel A, Abetz-Webb L, Lasch K, Acquadro C. Interviewing to develop Patient-Reported Outcome (PRO) measures for clinical research: eliciting patients' experience. Health Qual Life Outcomes. 2014; 12:15. Epub 20140205. https://doi.org/10.1186/1477-7525-12-15 PMID: 24499454; PubMed Central PMCID: PMC3933509.
12. Toye F, Williamson E, Williams MA, Fairbank J, Lamb SE. What value can qualitative research add to quantitative research design? An example from an adolescent idiopathic scoliosis trial feasibility study. Qualitative health research. 2016; 26(13):1838–50. https://doi.org/10.1177/1049732316662446 PMID: 27509903
13. Asher MA, Min Lai S, Burton DC. Further development and validation of the Scoliosis Research Society (SRS) outcomes instrument. Spine (Phila Pa 1976). 2000; 25(18):2381–6. Epub 2000/09/14. https://doi.org/10.1097/00007632-200009150-00018 PMID: 10984792.
14. Asher M, Lai SM, Burton D, Manna B. The reliability and concurrent validity of the scoliosis research society-22 patient questionnaire for idiopathic scoliosis. Spine. 2003; 28(1):63–9. https://doi.org/10.1097/00007632-200301010-00015 PMID: 12544958

15. Asher M, Lai SM, Burton D, Manna B. Scoliosis research society-22 patient questionnaire: responsiveness to change associated with surgical treatment. Spine. 2003; 28(1):70–3. https://doi.org/10.1097/00007632-200301010-00016 PMID: 12544959

16. Feise RJ, Donaldson S, Crowther ER, Menke JM, Wright JG. Construction and validation of the scoliosis quality of life index in adolescent idiopathic scoliosis. Spine (Phila Pa 1976). 2005; 30(11):1310–5. Epub 2005/06/02. https://doi.org/10.1097/01.brs.0000163885.12834.ca PMID: 15928558.

17. Matza LS, Patrick DL, Riley AW, Alexander JJ, Rajmil L, Pleil AM, et al. Pediatric Patient-Reported Outcome Instruments for Research to Support Medical Product Labeling: Report of the ISPOR PRO Good Research Practices for the Assessment of Children and Adolescents Task Force. Value Health. 2013; 16(4):461–79. https://doi.org/10.1016/j.jval.2013.04.004 PMID: 23796280

18. Essex R, Bruce G, Dibley M, Newton P, Thompson T, Swaine I, et al. A systematic scoping review and textual narrative synthesis of the qualitative evidence related to adolescent idiopathic scoliosis. International Journal of Orthopaedic and Trauma Nursing. 2022; 45:100921. https://doi.org/10.1016/j.ijotn.2022.100921 PMID: 35217471

19. Donnelly MJ, Dolan LA, Grande L, Weinstein SL. Patient and parent perspectives on treatment for adolescent idiopathic scoliosis. The Iowa orthopaedic journal. 2004; 24:76–83. PMID: 15296211; PubMed Central PMCID: PMC1888409.

20. Rullander A-C, Isberg S, Karling M, Jonsson H, Lindh V. Adolescents' experience with scoliosis surgery: a qualitative study. Pain Management Nursing. 2013; 14(1):50–9. https://doi.org/10.1016/j.pmn.2010.07.005 PMID: 23452527

21. Bull J, Grogan S. Children having spinal surgery to correct scoliosis: a qualitative study of parents' experiences. J Health Psychol. 2010; 15(2):299–309. https://doi.org/10.1177/1359105309351607 PMID: 20207673.

22. Motyer GS, Kiely PJ, Fitzgerald A. Adolescents' Experiences of Idiopathic Scoliosis in the Presurgical Period: A Qualitative Study. Journal of Pediatric Psychology. 2022; 47(2):225–35. https://doi.org/10.1093/jpepsy/jsab095 PMID: 34524430; PubMed Central PMCID: PMC8841985.

23. Tong A, Sainsbury P, Craig J. Consolidated criteria for reporting qualitative research (COREQ): a 32-item checklist for interviews and focus groups. Int J Qual Health Care. 2007; 19(6):349–57. https://doi.org/10.1093/intqhc/mzm042 PMID: 17872937

24. Patrick DL, Burke LB, Gwaltney CJ, Leidy NK, Martin ML, Molsen E, et al. Content validity—establishing and reporting the evidence in newly developed patient-reported outcomes (PRO) instruments for medical product evaluation: ISPOR PRO good research practices task force report: part 1—eliciting concepts for a new PRO instrument. Value Health. 2011; 14(8):967–77. Epub 2011/12/14. https://doi.org/10.1016/j.jval.2011.06.014 PMID: 22152165.

25. Lasch K, Marquis P, Vigneux M, Abetz-Webb L, Arnould B, Bayliss M, et al. PRO development: Rigorous qualitative research as the crucial foundation. Quality of life research: an international journal of quality of life aspects of treatment, care and rehabilitation. 2010; 19:1087–96. https://doi.org/10.1007/s11136-010-9677-6 PMID: 20512662

26. Connelly LM. What Is Phenomenology? Medsurg Nursing. 2010; 19(2):127–8. PMID: 20476524.

27. Bulawa P. Adapting Grounded Theory in Qualitative Research: Reflections from Personal Experience. International Research in Education. 2014; 2:145. https://doi.org/10.5296/ire.v2i1.4921

28. Kerr C, Nixon A, Wild D. Assessing and demonstrating data saturation in qualitative inquiry supporting patient-reported outcomes research. Expert Rev Pharmacoecon Outcomes Res. 2010; 10(3):269–81. https://doi.org/10.1586/erp.10.30 PMID: 20545592

29. Etikan I. Comparison of Convenience Sampling and Purposive Sampling. American Journal of Theoretical and Applied Statistics. 2016; 5:1. https://doi.org/10.11648/j.ajtas.20160501.11

30. Seid M, Varni JW, Jacobs JR. Pediatric Health-Related Quality-of-Life Measurement Technology: Intersections between Science, Managed Care, and Clinical Care. Journal of Clinical Psychology in Medical Settings. 2004; 7:17–27.

31. Ravens-Sieberer U, Erhart M, Wille N, Wetzel R, Nickel J, Bullinger M. Generic health-related quality-of-life assessment in children and adolescents: methodological considerations. Pharmacoeconomics. 2006; 24(12):1199–220. https://doi.org/10.2165/00019053-200624120-00005 PMID: 17129075.

32. Rajmil L, Herdman M, Fernandez de Sanmamed M-J, Detmar S, Bruil J, Ravens-Sieberer U, et al. Generic health-related quality of life instruments in children and adolescents: a qualitative analysis of content. Journal of Adolescent Health. 2004; 34(1):37–45. https://doi.org/10.1016/s1054-139x(03)00249-0 PMID: 14706404

33. Braun V, Clarke V. Using thematic analysis in psychology. Qual Res Psychol. 2006; 3(2):77–101.

34. Théroux J, Stomski N, Hodgetts CJ, Ballard A, Khadra C, Le May S, et al. Prevalence of low back pain in adolescents with idiopathic scoliosis: a systematic review. Chiropractic & Manual Therapies. 2017; 25(1):10. https://doi.org/10.1186/s12998-017-0143-1 PMID: 28439404

35. Thompson JY, Williamson EM, Williams MA, Heine PJ, Lamb SE. Effectiveness of scoliosis-specific exercises for adolescent idiopathic scoliosis compared with other non-surgical interventions: a systematic review and meta-analysis. Physiotherapy. 2019; 105(2):214–34. Epub 20181027. https://doi.org/10.1016/j.physio.2018.10.004 PMID: 30824243.

36. Monticone M, Ambrosini E, Cazzaniga D, Rocca B, Ferrante S. Active self-correction and task-oriented exercises reduce spinal deformity and improve quality of life in subjects with mild adolescent idiopathic scoliosis. Results of a randomised controlled trial. European Spine Journal. 2014; 23(6):1204–14. https://doi.org/10.1007/s00586-014-3241-y PMID: 24682356

37. Bastrom TP, Marks MC, Yaszay B, Newton PO, Group HS. Prevalence of postoperative pain in adolescent idiopathic scoliosis and the association with preoperative pain. Spine. 2013; 38(21):1848–52. https://doi.org/10.1097/BRS.0b013e3182a4aa97 PMID: 23883827

38. Perry-Eaddy M. "I thought I was going to die": A meta-synthesis exploring pediatric pain after scoliosis surgery. International Journal of Orthopaedic and Trauma Nursing. 2018;31. https://doi.org/10.1016/j.ijotn.2018.09.001 PMID: 30342906

39. Motyer G, Dooley B, Kiely P, Fitzgerald A. Parents' information needs, treatment concerns, and psychological well-being when their child is diagnosed with adolescent idiopathic scoliosis: A systematic review. Patient Educ Couns. 2021; 104(6):1347–55. Epub 20201125. https://doi.org/10.1016/j.pec.2020.11.023 PMID: 33280964.

40. Haynes S, Richard D, Kubany E. Content Validity in Psychological Assessment: A Functional Approach to Concepts and Methods. Psychological Assessment. 1995; 7:238–47. https://doi.org/10.1037/1040-3590.7.3.238

41. Weinstein SL, Dolan LA, Cheng JC, Danielsson A, Morcuende JA. Adolescent idiopathic scoliosis. Lancet. 2008; 371(9623):1527–37. Epub 2008/05/06. https://doi.org/10.1016/S0140-6736(08)60658-3 PMID: 18456103.

42. Altaf F, Gibson A, Dannawi Z, Noordeen H. Adolescent idiopathic scoliosis. The BMJ. 2013; 346:f2508. Epub 20130430. https://doi.org/10.1136/bmj.f2508 PMID: 23633006.

43. Aroeira RMC, de Las Casas EB, Pertence AEM, Greco M, Tavares JMRS. Non-invasive methods of computer vision in the posture evaluation of adolescent idiopathic scoliosis. Journal of Bodywork and Movement Therapies. 2016; 20(4):832–43. https://doi.org/10.1016/j.jbmt.2016.02.004.

44. Nault ML, Allard P, Hinse S, Le Blanc R, Caron O, Labelle H, et al. Relations between standing stability and body posture parameters in adolescent idiopathic scoliosis. Spine. 2002; 27(17):1911–7. https://doi.org/10.1097/00007632-200209010-00018 PMID: 12221357.

45. Yang JH, Suh S-W, Sung PS, Park W-H. Asymmetrical gait in adolescents with idiopathic scoliosis. European Spine Journal. 2013; 22(11):2407–13. https://doi.org/10.1007/s00586-013-2845-y PMID: 23732766

46. Danielsson AJ, Romberg K, Nachemson AL. Spinal Range of Motion, Muscle Endurance, and Back Pain and Function at Least 20 Years After Fusion or Brace Treatment for Adolescent Idiopathic Scoliosis: A Case-Control Study. Spine. 2006; 31(3):275–83. https://doi.org/10.1097/01.brs.0000197652.52890.71 PMID: 16449899-200602010-00005.

47. Nokariya S, Kotani T, Sakuma T, Iijima Y, Okumura T, Katogi T, et al. Trunk flexibility using a sit-and-reach test after surgery for adolescent idiopathic scoliosis. Spine deformity. 2022. Epub 20221104. https://doi.org/10.1007/s43390-022-00608-3 PMID: 36331800.

48. Schmid S, Burkhart KA, Allaire BT, Grindle D, Bassani T, Galbusera F, et al. Spinal Compressive Forces in Adolescent Idiopathic Scoliosis With and Without Carrying Loads: A Musculoskeletal Modeling Study. Frontiers in Bioengineering and Biotechnology. 2020; 8. https://doi.org/10.3389/fbioe.2020.00159 PMID: 32195239

49. Lima M, Ferreira AS, Reis FJJ, Paes V, Meziat-Filho N. Chronic low back pain and back muscle activity during functional tasks. Gait & Posture. 2018; 61:250–6. https://doi.org/10.1016/j.gaitpost.2018.01.021.

50. Groenewald CB, Tham SW, Palermo TM. Impaired School Functioning in Children With Chronic Pain: A National Perspective. Clinical Journal of Pain. 2020; 36(9):693–9. https://doi.org/10.1097/AJP.0000000000000850 PMID: 32487871; PubMed Central PMCID: PMC7429324.

51. LaMontagne LL, Hepworth JT, Cohen F, Salisbury MH. Adolescent scoliosis: Effects of corrective surgery, cognitive-behavioral interventions, and age on activity outcomes. Applied Nursing Research. 2004; 17(3):168–77. https://doi.org/10.1016/j.apnr.2004.06.007 PMID: 15343550

52. Danielsson AJ, Wiklund I, Pehrsson K, Nachemson AL. Health-related quality of life in patients with adolescent idiopathic scoliosis: a matched follow-up at least 20 years after treatment with brace or surgery. European spine journal. 2001; 10(4):278–88. https://doi.org/10.1007/s005860100309 PMID: 11563612

53. Rullander A-C, Lundström M, Östlund U, Lindh V. Adolescents' Experiences of Scoliosis Surgery and the Trajectory of Self-Reported Pain. Orthopaedic Nursing. 2017; 36(6):414–23.

54. Bertuccelli M, Cantele F, Masiero S. Body Image and Body Schema in Adolescents with Idiopathic Scoliosis: A Scoping Review. Adolescent Research Review. 2022. https://doi.org/10.1007/s40894-022-00187-4

55. Gallant JN, Morgan CD, Stoklosa JB, Gannon SR, Shannon CN, Bonfield CM. Psychosocial Difficulties in Adolescent Idiopathic Scoliosis: Body Image, Eating Behaviors, and Mood Disorders. World Neurosurgery. 2018; 116:421–32.e1. Epub 20180523. https://doi.org/10.1016/j.wneu.2018.05.104 PMID: 29803063.

56. Rullander AC, Lundström M, Lindkvist M, Hägglöf B, Lindh V. Stress symptoms among adolescents before and after scoliosis surgery: correlations with postoperative pain. Journal of Clinical Nursing. 2016; 25(7–8):1086–94. https://doi.org/10.1111/jocn.13137 PMID: 26898698

57. Yakut Y, Pelin Z, Yagci G. An investigation of sleep profiles in individuals with idiopathic scoliosis. Sleep Science. 2022; 15(2):172–8. https://doi.org/10.5935/1984-0063.20220038 PMID: 35755911; PubMed Central PMCID: PMC9210568.

58. Huus K, Schlebusch L, Ramaahlo M, Samuels A, Berglund IG, Dada S. Barriers and facilitators to participation for children and adolescents with disabilities in low- and middle-income countries—A scoping review. Afr J Disabil. 2021; 10:771. Epub 20210308. https://doi.org/10.4102/ajod.v10i0.771 PMID: 33824860; PubMed Central PMCID: PMC8008013.

59. Kakar RS, Simpson KJ, Das BM, Brown CN. Review of physical activity benefits and potential considerations for individuals with surgical fusion of spine for scoliosis. International journal of exercise science. 2017; 10(2):166. PMID: 28344731

Appendix 3. Alamrani et al. (2021b)

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Content validity of Scoliosis Research Society questionnaire-22 revised (SRS-22r) for adolescents with idiopathic scoliosis: protocol for a qualitative study exploring patient's and practitioner's perspectives

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ABSTRACT

Introduction Content validity is the most important measurement property for any patient-reported outcome measure (PROM). It being the extent that the PROM measures important concepts that are relevant to the population of interest. Adolescent with idiopathic scoliosis (AIS) is the most common spinal deformity in paediatric populations, with the Scoliosis Research Society questionnaire-22 revised (SRS-22r) a commonly used PROMof quality of life. In the absence of existing evidence, a content validity study for SRS-22r is needed to confirm its suitability for AIS. Thus, this study aims to investigate the content validity of SRS-22r for AIS. A secondary aim is to explore healthcare professional (HCP) perspectives of the barriers and facilitators to using outcome measures in AIS.

Methods and analysis Qualitative study reported according to COnsolidated criteria for Reporting Qualitative Studies. A purposive sample of AIS (n=10–15, Cobb angle >25°, aged 10–18 years) will be recruited for online semi-structured interviews. A convenience sample (n=10–12) of HCP with clinical and/or research experience in AIS will be recruited for a focus group discussion. Topic guides and age-relevant documents are informed by existing evidence and developed using a framework of concept elicitation and cognitive debriefing. Audio-recordings will be transcribed verbatim, coded, analysed and synthesised using interpretive phenomenology analysis. Themes that generated from the analysis will be used as codes that will then be mapped to the SRS-22r contents.

Ethics and dissemination The Health Research Authority and Health and Care Research Wales approval have been granted (IRAS 289888). Study findings will be disseminated through publications in peer-reviewed journals and conference presentations.

Strengths and limitations of this study

- This will be the first study to explore the content validity of the Scoliosis Research Society questionnaire-22 revised and its suitability for adolescent with idiopathic scoliosis (AIS).
- Purposive sampling technique will be used to ensure diversity of the sample.
- Interpretive phenomenology analysis will be used to understand experiences of AIS to their health condition and its associated treatment.
- The small sample size and recruitment from one site may limit the transferability of study findings.

INTRODUCTION

Adolescent with idiopathic scoliosis (AIS) is a scoliosis of unknown cause between the ages of 10–18 years.[1] It is the most common spinal deformity among paediatric patients,[2] with prevalence ranging from 1% to 3%.[3] The AIS may experience health problems including back pain,[4] psychological stress and respiratory dysfunction[5] which can significantly impact quality of life (QoL).[6]

Patient-reported outcome measures (PROM) are commonly used to evaluate QoL and provide patients' perceptions about their health condition and its associated management.[7] A PROM should exhibit good content validity to be recommended for use,[8] this being 'the degree to which elements of an assessment instrument are relevant to, and representative of, the targeted construct for a particular assessment purpose'[9 (p238)]. It is an imperative to the other forms of validity as it shows that all aspects of interest have been sufficiently captured in the PROM and that it is suitable for the intended use.[8 10]

The Scoliosis Research Society-22 revised (SRS-22r) questionnaire is the frequently used PROM for AIS.[11] It has been selected as the preferred PROM in the Core Outcome

Study for young individuals with spine deformity for evaluating the core outcome domains such as self-image, physical functioning, pain and participation.[12] The first version of the SRS questionnaire was developed by Haher *et al* comprising 24 items[13] and later modified to include 22 items (SRS-22).[14 15] The population involved in these studies had a mean age that was older than AIS (25 years old, ranging 19–34 years), and therefore, may not be truly representative of AIS. Additionally, the mental health domain includes some questions from the SF-36 survey,[16] which was designed for an adult population, and is a generic rather than condition-specific measure. Further, the SRS-22 has reported ceiling effects (20%–44%),[14 17] which undermine its reported content validity and reliability.[8]

Using the International Classification of Functioning, Disability and Health (ICF), AIS reported limitations in their functioning.[6] The SRS-22 does not include items about the cardiovascular system, weight management or leisure, all of which are considered important to AIS.[6] These findings raise doubt as to the appropriateness of the SRS-22 and its revised version (SRS-22r) of this discrete population, and highlight the need for a content validity study of the SRS-22r in an English-speaking population to assess its suitability.

Our recent systematic review of measurement properties of physical functioning outcome measures used in AIS, shows that the SRS-22 is a widely used PROM and has been adapted and translated into more than eleven languages.[11] The common use of the SRS-22r may suggest acceptance within clinical and research practice. However, it is necessary to document its content validity with qualitative interviews from the population of interest with diverse characteristics to ensure that different perspectives of patients are fully captured within the PROM.[8] For adolescents, it is essential that a PROM is relevant to their age group and not just a use of adult measures,[18] considering their developmental stage and unique emotional and social characteristics.[18]

Although a PROM provides important information from a patients perspective, it might be influenced by a patient's perception of change,[19] and other factors such as pain and psychological stress, which often reported in this population.[4 20–22] The body structure and function measures, such as range of motion gives indication about the dysfunction in structure or function, but it fails to fully capture the functional limitations.[19] Conversely, performance-based outcome measures (eg, walking speed) may achieve a reproducible and unbiased assessment of function.[19] There is a preference in the literature towards using a PROM compared with performance-based outcome measure. Furthermore, studies that evaluate the measurement properties of these outcome measures were relatively limited, which may limit its use in AIS.[11] Thus, as well as understanding the content validity of the SRS-22r, to optimise the use of PROM in practice there is a need to understand perceptions of healthcare professional (HCP) who manage this specific population and explore the barriers and facilitators to use outcome measures.

Aim

To assess the content validity of the SRS-22r questionnaire with AIS and HCP. A secondary aim is to explore barriers and facilitators of HCP use of outcome measure in AIS.

Objectives

1. To elicit concepts that are most relevant and important to AIS.
2. To determine the relevance, comprehensiveness and comprehensibility of SRS-22r contents from the perspectives of AIS.
3. To determine the relevance, comprehensiveness of SRS-22r contents from perspectives of HCP.
4. To explore perceptions of HCP on the barriers and facilitators of using outcome measures in AIS.

METHODS AND ANALYSIS

Design and methods

This study will be reported in line with the COnsolidated criteria for Reporting Qualitative studies.[23] It will consist of semistructured interviews with AIS and a focus group discussion with HCP. The interpretive phenomenology analysis (IPA) will be used to explore and understand perceptions and experiences of AIS to their health condition and its associated treatment.[24] The IPA is an approach to collect and analyse qualitative data that seeks to understand the lived experience.[25] The IPA provide an open-ended approach which is needed for the concept elicitation of PROM items, to capture important aspects to patients.[26] To understand barriers and facilitators to use outcome measure among AIS, thematic analysis following Braun and Clarke framework will be used.[27]

The assessment of content validity of an existing PROM consists of both concept elicitation and cognitive debriefing. In concept elicitation, important concepts related to construct of the PROM are elicited from the target population and then mapped to the content of the existing PROM.[28] Meanwhile cognitive debriefing exploring participants understanding about three aspects of PROM, that is, relevance, comprehensiveness and comprehensibility of the items included in the PROM.[8] A flow chart of the study design is shown in (figure 1).

Participant recruitment and eligibility criteria

- Individuals should meet the following inclusion criteria to participate in semi-structured interviews: diagnosis of AIS by their respective physician (Cobb angle >25°); age 10–18 years old; and have access to a video/audio call platform. A Cobb angle of 25° has been chosen as the minimum curve size that may necessitate a change in management (eg, bracing, surgery) and going beyond observation and monitoring.[29]
- Exclusion criteria: individuals with other forms of scoliosis, and those who unable to speak English fluently.

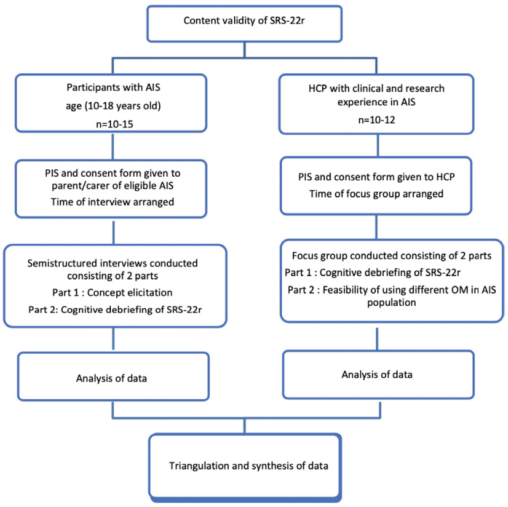

Figure 1 Study flow chart. AIS, adolescent idiopathic scoliosis; HCP, healthcare professional; OM, outcome measure; PIS, participants information sheet; SRS-22r, Scoliosis Research Society questionnaire-22 revised.

- A sample size of 5–25 participants was suggested for studies using phenomenology.[30 31] Thus, a purposive sample of n=15 participants was estimated to be sufficient to test the content validity of SRS-22r. Purposive sampling will be used to recruit participants, which is a method of identification and selection of participants who are experienced with the phenomenon of interest.[32] This is to ensure diversity in the sample in terms of age-categories, gender, ethnicity, different curve severity (mild, moderate and sever curves), that could be managed through observation, bracing and surgery. Those who treated with surgery would be sampled to recruit individual at different surgery status (eg, 3 months, 6 months, 1 year). This approach will enable a wide range of experience and opinions to be explored.[28 33]
- It has been recommended when examining the content validity of a PROM, data collection to be continued till point of saturation, to ensure that the items in the PROM appropriately represent the content of the concept.[7] Sampling will continue until data saturation is achieved, and whereby no new concepts emerge from participants' interviews or are considered as missing during the cognitive interviews.[28]
- Participants will be identified by a research nurse and the study will be outlined with them. Those who show interest in the study will be given an age relevant participant information sheet (PIS). Then, their parent/carer will be contacted by the lead researcher to confirm eligibility, discuss the PIS, answer any questions about the study and arrange the time of the interview.
- Clinicians who are treating AIS (physiotherapists, spinal surgeons, nurses), or individuals who have research experience in AIS will be invited to participate in a focus group discussion (expected n=10–12). We anticipate that two focus groups will be sufficient to fulfil the aims and objectives of the study.[34]

Study settings

Participants for the interviews and the focus group will be recruited from The Royal Orthopaedic Hospital NHS Trust, Birmingham, UK (figure 1). Both interviews and the focus group will be conducted virtual via Zoom/Microsoft Teams or by a phone call if video calling is not possible. A parent/carer or an adult family member will be asked to be present with child participants <16 years old throughout the interview. However, to ensure that the interview is reflecting the child's perspective, they will be asked to not actively participate in the interview unless invited to do so to lend support.

In instances where the parents/carers are not able to present with their child, the interviewer (SA) will make sure that the child is comfortable/happy to conduct the interview without their parent. The interview will be terminated if the child is unwilling to do so. The interviewer (SA) has both DBS check and safeguarding training to work with children. The timing of the focus group discussion will be selected at the convenience to all HCP participants.

Data collection and procedure

Objectives 1 and 2: semistructred interviews

The semistructured interview will last approximately 60–90 min, and it will adhere to PROM development format,[28] consisting of concept elicitation and cognitive debriefing.[8] The concept elicitation aims to elicit key concepts related to the influence of spine deformity on adolescents' QoL, while the cognitive debriefing aims to gather feedback on the content of the SRS-22r, including relevance, comprehensiveness and comprehensibility.[8 35] As well as exploring if there are any additional/important areas that need to be covered in the SRS-22r, participants will be invited to raise new topics and/or issues during the interviews.[28]

The topic guide (online supplemental file 1) was developed using existing evidence,[33] as well as perspectives of AIS through patient and public involvement. Additionally, the ICF framework[36] has been used to ensure all important aspects of the life of adolescents are covered. The topic guide is age-relevant and will be piloted with an AIS ahead of the main study to identify areas that do not flow easily or may confuse participants.[7] The topic guide consists of open-ended questions and it is written in way that allows participants to provide detailed information with unlimited responses.[7] The topic guide will be modified when new themes are discovered from the completed interviews.[33]

Objectives 3 and 4: focus group

HCP and researchers will be invited to join a focus group discussion, which will last approximately 60–90 min. It will consist of two parts, first to evaluate the content validity of SRS-22r and second to explore barriers and facilitators to the use of PROM for AIS. The topic guide (online supplemental file 2) has been developed and will be piloted ahead of the main study.[7]

Research team and reflexivity

Semistructured interviews will be undertaken by the chief researcher (SA). She is an experienced musculoskeletal physiotherapist with experience of working with adolescents. The focus group discussion of HCP will be led by an experienced musculoskeletal physiotherapy researcher with specialism in spinal research and experience in conducting qualitative research, as well as involvement of a spinal surgeon with experience working with AIS, and involvement of (ER) as Patient and Public Involvement (PPI). The lead interviewer for the interviews will be present as an observer and she will take field notes. Although no specific relationship will be established prior to the commencement of the interviews or focus group, participants will be informed about the professional background of the interviewer and that the study is part of a PhD thesis.

Data management and data analysis

Demographic and clinical information of participants will be collected prior to the interviews and focus group discussion which will then be used to characterise the sample. Interviews and the focus group will be audio recorded and transcribed verbatim. The transcripts will be emailed to participants or their parent/carer to enable member checking and allow any further details to be added. Participants will be given 2 weeks to make any alterations or suggest changes. The interviewer (SA) will take supplementary field notes during the interviews and focus group discussion to allow data triangulation. A saturation table will be used to document data saturation. The information elicited from interviews will be arranged by concept code.[28 37] Further, the depth of analysis will be evaluated using a code book.[28]

Coding of the first interview will be performed by two researchers (SA and NRH) to develop a coding plan and to ensure that researchers concur in coding. The lead researcher will then code the remaining interviews independently. Any new themes or codes arising from the transcripts will be reviewed by coders, for every 2–3 transcripts coded.

Qualitative interview data will be analysed according to a four-stage approach of IPA[38] :

Stage 1: Multiple reading and making notes. The initial stage will involve close reading of the transcripts multiple times. The researcher will then make notes about her observations and reflections about the interview experience.

Stage 2: Transforming notes into Emergent Themes. Notes from initial stage will be transformed into emerging themes. Preliminary themes will then be presented and discussed within the research team.

Stage 3: Seeking relationships and clustering themes. The emerging themes will be grouped together in clusters.

Stage 4: Production of summary table. Themes will be presented in a summary table with verbatim extract and it will be subsequently discussed with the research team.[38 39]

Themes that are generated from the analysis will be used as codes that will then be mapped to the content of the SRS-22r.[28]

Qualitative data produced from focus group discussion with HCP will be analysed using the Braun and Clarke 6-step process.[27]

1. Familiarise yourself with the data.
2. Generate initial codes.
3. Searching for themes.
4. Reviewing themes.
5. Defining and naming themes.
6. Producing the report.[27]

Data storage, access and disposal

Digital audio recordings of the interviews and the focus group will be uploaded securely and transcribed by the approved service provider. Participants' data will be kept on a secure, password protected, database on the site at the University of Birmingham for 10 years, accessible only to the research team. It will be kept in accordance with General Data Protection Regulation, the Data Protection Act 2018 and University of Birmingham's research governance framework.

Patient and public involvement

This study was conceived directly because of gaps identified in a systematic review of physical functioning outcome measures among AIS.[40] The protocol of systematic review was informed following discussions at a patient and public involvement meeting at the Centre of Precision Rehabilitation for Spinal Pain at the University of Birmingham. The PPI representative is a part of the study management group (comprise all coinvestigators involved in the design of the study, interpretation of the study and identification of the participants). Their feedback has been sought on the study protocol, the topic guide as well as the PISs and consent forms. Further, they will be involved in data analysis/interpretation and study findings through provision of a plain English summary.

Implications of this study

The AIS is a common spinal deformity that can significantly affect the adolescents QoL.[6] It is important that the PROM used for evaluating their QoL has sufficient content validity.[8] Adolescents comprise a unique population and a PROM should be relevant, comprehensible and capture all areas which are relevant and important to them.[18] A qualitative study with AIS is, therefore, needed to understand their experiences with their health

condition and its management, which then allows an evaluation of the SRS-22r contents. Findings from this study will provide important evidence on the content validity of SRS-22r, which will build confidence in the PROM findings[41] and further support its use in practice and for future research.

Ethics and dissemination

The Health Research Authority and Health and Care Research Wales approval has been granted (IRAS 289888). AIS participants will be advised that participation in study will not affect their current and future healthcare. Minimal risk is associated with this study. Informed consent from children >16 years old, and informed consent from the parent/carer, along with assent from the child <15 years old will be obtained prior to inclusion in the study. The study findings will be presented without identification of any study participants and any protocol amendment will be documented. Findings will be disseminated through publications in peer-reviewed journals as well as international and national conference presentations.

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Contributors All authors conceptualised and designed the protocol. SA is a PhD student and NRH (lead supervisor), ABR and DF are supervisors and AG is a spinal surgeon. SA drafted the initial manuscript with NRH, ABR, DF and AG providing guidance on design, topic, methodology and analyses. ER acted as patient and public involvement representative, contributing to the design of the study. All authors reviewed and commented on each draft of the protocol. All authors have approved and contributed to the final manuscript.

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REFERENCES

1 James JI. Idiopathic scoliosis; the prognosis, diagnosis, and operative indications related to curve patterns and the age at onset. *J Bone Joint Surg Br* 1954;36-B:36–49.
2 Konieczny MR, Senyurt H, Krauspe R. Epidemiology of adolescent idiopathic scoliosis. *J Child Orthop* 2013;7:3–9.
3 Weinstein SL, Dolan LA, Cheng JCY, *et al*. Adolescent idiopathic scoliosis. *Lancet* 2008;371:1527–37.
4 Makino T, Kaito T, Kashii M, *et al*. Low back pain and patient-reported QOL outcomes in patients with adolescent idiopathic scoliosis without corrective surgery. *Springerplus* 2015;4:397.
5 Durmala J, Tomalak W, Kotwicki T. Function of the respiratory system in patients with idiopathic scoliosis: reasons for impairment and methods of evaluation. *Stud Health Technol Inform* 2008;135:237–45.
6 Du C, Yu J, Zhang J, *et al*. Relevant areas of functioning in patients with adolescent idiopathic scoliosis on the International classification of functioning, disability and health: the patients' perspective. *J Rehabil Med* 2016;48:806–14.
7 Patrick DL, Burke LB, Gwaltney CJ, *et al*. Content validity--establishing and reporting the evidence in newly developed patient-reported outcomes (PRO) instruments for medical product evaluation: ISPOR PRO good research practices task force report: part 1--eliciting concepts for a new PRO instrument. *Value Health* 2011;14:967–77.
8 Terwee CB, Prinsen CAC, Chiarotto A, *et al*. COSMIN methodology for evaluating the content validity of patient-reported outcome measures: a Delphi study. *Qual Life Res* 2018;27:1159–70.
9 Haynes SN, Richard DCS, Kubany ES. Content validity in psychological assessment: a functional approach to concepts and methods. *Psychol Assess* 1995;7:238–47.
10 U.S. Department of Health and Human Services FDA Center for Drug Evaluation and Research, U.S. Department of Health and Human Services FDA Center for Biologics Evaluation and Research, U.S. Department of Health and Human Services FDA Center for Devices and Radiological Health. Guidance for industry: patient-reported outcome measures: use in medical product development to support labeling claims: draft guidance. *Health Qual Life Outcomes* 2006;4:79.
11 Alamrani S, Rushton AB, Gardner A, *et al*. Physical functioning in adolescents with idiopathic scoliosis: a systematic review of outcome measures and their measurement properties. *Spine* 2021;46:E985–97.
12 de Kleuver M, Faraj SSA, Holewijn RM, *et al*. Defining a core outcome set for adolescent and young adult patients with a spinal deformity. *Acta Orthop* 2017;88:612–8.
13 Haher TR, Gorup JM, Shin TM, *et al*. Results of the scoliosis research Society instrument for evaluation of surgical outcome in adolescent idiopathic scoliosis. A multicenter study of 244 patients. *Spine* 1999;24:1435.
14 Asher MA, Lai SM, Glattes RC, *et al*. Refinement of the SRS-22 health-related quality of life questionnaire function domain. *Spine* 2006;31:593–7.
15 Asher M, Min Lai S, Burton D, *et al*. The reliability and concurrent validity of the scoliosis research society-22 patient questionnaire for idiopathic scoliosis. *Spine* 2003;28:63–9.
16 Asher M, Min Lai S, Burton D, *et al*. Scoliosis research society-22 patient questionnaire: responsiveness to change associated with surgical treatment. *Spine* 2003;28:70–3.
17 Glattes RC, Burton DC, Lai SM, *et al*. The reliability and concurrent validity of the scoliosis research Society-22r patient questionnaire compared with the child health Questionnaire-CF87 patient questionnaire for adolescent spinal deformity. *Spine* 2007;32:1778–84.
18 Matza LS, Patrick DL, Riley AW, *et al*. Pediatric patient-reported outcome instruments for research to support medical product labeling: report of the ISPOR pro good research practices for the assessment of children and adolescents Task force. *Value Health* 2013;16:461–79.

19 Reiman MP, Manske RC. The assessment of function: how is it measured? A clinical perspective. *J Man Manip Ther* 2011;19:91–9.
20 Seki H, Ideno S, Ishihara T, *et al*. Postoperative pain management in patients undergoing posterior spinal fusion for adolescent idiopathic scoliosis: a narrative review. *Scoliosis Spinal Disord* 2018;13:17.
21 Bastrom TP, Marks MC, Yaszay B, *et al*. Prevalence of postoperative pain in adolescent idiopathic scoliosis and the association with preoperative pain. *Spine* 2013;38:1848–52.
22 Leszczewska J, Czaprowski D, Pawłowska P, *et al*. Evaluation of the stress level of children with idiopathic scoliosis in relation to the method of treatment and parameters of the deformity. *ScientificWorldJournal* 2012;2012:538409.
23 Tong A, Sainsbury P, Craig J. Consolidated criteria for reporting qualitative research (COREQ): a 32-item checklist for interviews and focus groups. *Int J Qual Health Care* 2007;19:349–57.
24 Smith TLJA, Flower P, Larkin M. Interpretative phenomenological analysis: theory, method and research. *Qualitative Research in Psychology* 2009;2009:346–7.
25 Lasch KE, Marquis P, Vigneux M, *et al*. PRO development: rigorous qualitative research as the crucial Foundation. *Qual Life Res* 2010;19:1087–96.
26 Cheng KKF, Clark AM. Qualitative methods and patient-reported outcomes. *Int J Qual Methods* 2017;16:160940691770298.
27 Braun V, Clarke V. Using thematic analysis in psychology. *Qual Res Psychol* 2006;3:77–101.
28 Rothman M, Burke L, Erickson P, *et al*. Use of existing patient-reported outcome (PRO) instruments and their modification: the ISPOR good research practices for evaluating and documenting content validity for the use of existing instruments and their modification pro Task force report. *Value Health* 2009;12:1075–83.
29 Gardner A, Berryman F, Pynsent P. What is the variability in shoulder, axillae and waist position in a group of adolescents? *J Anat* 2017;231:221–8.
30 Creswell JW, Poth CN. *Qualitative inquiry and research design: choosing among five approaches*. Sage publications, 2016.
31 Morse JM. Designing funded qualitative research 1994.
32 Etikan I, Musa SA, Alkassim RS. Comparison of convenience sampling and purposive sampling. *Am J Theoretical Appl Stats* 2016;5:1–4.
33 Brod M, Tesler LE, Christensen TL. Qualitative research and content validity: developing best practices based on science and experience. *Qual Life Res* 2009;18:1263–78.
34 Hennink MM, Kaiser BN, Weber MB. What influences saturation? estimating sample sizes in focus group research. *Qual Health Res* 2019;29:1483–96.
35 Patrick DL, Burke LB, Gwaltney CJ, *et al*. Content validity--establishing and reporting the evidence in newly developed patient-reported outcomes (PRO) instruments for medical product evaluation: ISPOR PRO Good Research Practices Task Force report: part 2--assessing respondent understanding. *Value Health* 2011;14:978–88.
36 World Health O.. *International classification of functioning, disability and health : ICF*. Geneva: World Health Organization, 2001.
37 Kerr C, Nixon A, Wild D. Assessing and demonstrating data saturation in qualitative inquiry supporting patient-reported outcomes research. *Expert Rev Pharmacoecon Outcomes Res* 2010;10:269–81.
38 Smith JA, Flowers P, Larkin M. *Interpretative phenomenological analysis: theory, method and research*. SAGE Publications, 2009.
39 Smith JA, Shinebourne P. *Interpretative phenomenological analysis*. American Psychological Association, 2012.
40 Alamrani S, Rushton A, Gardner A, *et al*. Outcome measures evaluating physical functioning and their measurement properties in adolescent idiopathic scoliosis: a protocol for a systematic review. *BMJ Open* 2020;10:e034286.
41 Newman I, Lim J, Pineda F. Content validity using a mixed methods approach: its application and development through the use of a table of specifications methodology. *J Mix Methods Res* 2013;7:243–60.

Appendix 4. Alamrani et al. (2021b)

Spine

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LITERATURE REVIEW

Physical Functioning in Adolescents with Idiopathic Scoliosis

A Systematic Review of Outcome Measures and Their Measurement Properties

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Study Design. A systematic review.

Objective. To summarize evidence on measurement properties of Outcome Measures (OM) used to assess physical functioning in adolescents with idiopathic scoliosis (AIS).

Summary of Background Data. The AIS is a common spine deformity in those aged 10 to 18 years old. Associated health problems (*e.g.*, back pain) significantly impact the quality of life (QoL). One important domain in QoL is physical functioning, which can be measured with patient-reported outcome measures (PROM), performance-based outcome measures (PBOM), and body structure and function OM. Adequate measurement properties of OM are important for precision in research and practice.

Methods. A two-staged search strategy was performed on electronic databases up to December 2019. Search one revealed a list of OM was used for physical functioning assessment in AIS. Search two identified studies that evaluated the measurement properties of OM in AIS; using the list identified in search one. Two independent reviewers determined study eligibility, risk of bias assessment (COnsensus-based Standards for the selection of health Measurement INstruments [COSMIN] checklist), and performed data extraction. The level of evidence was established using a modified GRADE approach.

Results. Search one yielded: 28 PROM, 20 PBOM, and 10 body structure and function OM. Search two revealed: 16 measurement properties studies for PROM, one for PBOM, and three for body structure and function measures. Construct validity, reliability, and responsiveness of most PROM has been established in AIS, but not content validity or internal consistency (moderate evidence). Construct validity was sufficient for the Timed Up and Go test and body structure and function measures (very low to low evidence).

Conclusion. Currently, physical functioning is evaluated with a variety of measures in AIS. The majority of measurement properties studies evaluated PROM with a paucity of information on measurement properties of PBOM and body structure and function OM. Based on COSMIN methodology, none of the OM identified in this review can be recommended with confidence in individuals with AIS.

Key words: idiopathic scoliosis, measurement properties, outcome assessment, physical functioning, reliability, systematic review, validity.

Level of Evidence: 2

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Adolescent idiopathic scoliosis (AIS) is the most common spine deformity among children aged 10 to 18 years old,[1] with prevalence ranging 1% to 3%.[2] Comprising of a lateral curvature and axial rotation of spinal vertebrae, the cause is unknown in most cases.[3] AIS has been linked to back pain,[4] psychological stress,[5] and respiratory dysfunction,[6] potentially impacting on quality of life (QoL).[7]

A dimension of any QoL measurement is "physical functioning" this being the ability to carry out activities of daily living.[8] Physical functioning limitations have been associated with an increased risk of disability and predictive of social and healthcare use.[9] Limitations include walking and maintaining body positions,[7] as well as pain related functional restriction.[10] Corrective surgery is used for some, necessitating a long recovery period and often associated with pain and immobility in adolescence.[11] Measuring the impact of AIS is therefore important in both research and clinical practice.

Physical functioning can be evaluated with patient-reported outcome measures (PROM), performance-based outcome measures (PBOM), and measures of body structure and function.[12] Each measure assesses different, but complementary, aspects of physical functioning,[12] with PROM for self-report, PBOM for the performance of a specific activity (*e.g.*, chair stand test),[12,13] and body structure and function providing anatomical data (*e.g.*, range of motion) or a physiological process (*e.g.*, muscle strength).[12]

Outcome measures need adequate measurement properties to assure truthfulness of results and avoid risk of bias.[14] The COnsensus-based Standards for the selection of health Measurement INstruments (COSMIN) group developed a taxonomy of measurement properties to enable this.[15] Three main domains are validity, reliability, and responsiveness.[15] The COSMIN group provide guidelines for conducting a systematic review for PROM, which can be adapted for other OM.[16]

The Scoliosis Research Society questionnaire (SRS-22) and its' variants are the most widely used PROM in this population.[17–19] From the core outcome study (COS), SRS-22 revised (SRS-22r) is recommended and the considered reference standard for evaluating physical functioning for adolescents and young adults with spine deformity.[20] However, SRS-22r does not capture all aspects of physical functioning, such as mobility and self-care.[7] Furthermore, the COS study included all forms of spinal deformities; the heterogeneity limiting applicability to individuals with AIS. Furthermore, little is known about PBOM and body structure and function measures for individuals with AIS.

In the absence of existing relevant reviews,[21] the purpose of this review was to identify OM used to assess physical functioning in individuals with AIS, and secondly to evaluate their measurement properties.

METHODS

Design

This review was conducted according to a registered (PROSPERO CRD42019142335) and published protocol.[22] Designed in line with COSMIN methodology for systematic review of PROM,[16] the review is reported in line with Preferred Reporting Items for Systematic Review and Meta-Analysis (PRISMA) statement.[23]

Search Strategy

The search was conducted in two parts. Search one identified and generated a list of OM used for assessment of physical functioning in AIS. Search two identified the studies of measurement properties using the list from search one. Details of both search are listed in Table 1.

Data Sources

A comprehensive search was performed using MEDLINE, PsycINFO, EMBASE, CINAHL, SPORTdiscus, Web of Science, and PubMed databases from date of inception until December 2019. As well as searches on key journals, reference lists, conference proceedings, and grey literature were also searched. The search terms were first developed for MEDLINE and then adapted with relevant syntax and subject headings for the other databases. Supplemental digital content 1, http://links.lww.com/BRS/B720 shows example of search one and two.

TABLE 1. Search One and Search Two Strategy

	Search One (Inventory of Outcome Measure)	Search Two (Measurement Properties)
Inclusion criteria	Individuals with AIS (≥10° Cobb angle)[1] Age 10–18 years old	Individuals with AIS (≥10° Cobb angle)[1] Age 10–18 years old Mixed cohort studies >50% of participants with AIS
	Any study design that included assessment of physical functioning for individuals with AIS. No limitations were applied on type of outcome measure, language or location.	Measurement properties studies (*i.e.*, content validity, structural validity, construct validity, reliability, and responsiveness) of outcome measure identified in search one.
	Outcome measure defined as following: PROM in form of questionnaires, scales or sub-scales, designed to evaluate physical functioning in AIS. PBOM, meaning a clinician- observer measure of an ''activity'' such as the execution of a task or action by an individual,[2] measured by/or time, or distance. Body structure and function measures defined as ''the physiological function of body systems and/or the anatomical parts of body.''[12,25]	
Exclusion criteria	Radiographs, laboratory-based measures, anthropometric measures.[26–32]	Studies in non-English speaking population Systematic reviews Studies providing normative data Studies providing indirect evidence on measurement properties.

AIS indicates adolescent idiopathic scoliosis; PBOM, performance based outcome measure; PROM, patient reported outcome measure.

Study Selection
Two independent reviewers (S.A., E.B.) assessed studies based on the title and abstract for eligibility. In case of insufficient information, full text articles were retrieved and screened for eligibility. The reviewers discussed findings and reached consensus on eligibility of studies. The percentage agreement between reviewers was estimated using the κ statistic (IBM SPSS Statistics for Windows, Version 25.0. Armonk, NY: IBM Corp).

Data Extraction
Two reviewers (S.A., E.B.) independently extracted data of eligible studies. Information about study, participants characteristics, outcome measures, and measurement properties were extracted. If information was not clear or unavailable in studies, corresponding authors were contacted.

Risk of Bias Assessment
The risk of bias for each measurement properties was assessed using COSMIN checklist.[14] Adaptions were made for studies of body structure and function, for example, interobserver reliability. This involved removal of inapplicable standards, that is, "was the time interval appropriate?" Each item of measurement property was rated as either "very good," "adequate," "doubtful," or "inadequate quality."[14] Subsequently overall methodological quality of measurement property was rated based on "the worst score counts principle."[14] Two independent reviewers (S.A., E.B.) assessed study quality and inconsistencies were resolved by discussion.

Hypotheses for Construct Validity and Responsiveness
Hypotheses for evaluating construct validity and responsiveness assessed in included studies, were pre-defined[33] and listed in supplemental digital content 2, http://links.lww.com/BRS/B721.

Data Analysis and Synthesis
The necessary homogeneity in studies results was insufficient, thus meta-analysis was not performed. Results were therefore synthesized and qualitatively summarized.[16] The measurement property for each study was rated according to updated criteria for good measurement properties as sufficient (+), insufficient (−), or indeterminate (?).[16] Then, evidence was graded using modified Grading of Recommendations Assessment, Development and Evaluation (GRADE) approach.[16] Five factors determine quality of evidence: risk of bias, inconsistency, indirectness, imprecision, and publication bias.[34] For evaluating measurement properties in systematic reviews of PROM, only four factors were assessed, with fifth factor (publication bias) removed.[33]

RESULTS
The PRISMA flow diagram shows results of both searches, selection process, and reasons for exclusion (Figure 1).

Search One: Inventory of Outcome Measure
A list of OM was generated and classified into 28 PROM, 20 PBOM, and 10 body structure and function OM are listed in supplemental digital content 3, http://links.lww.com/BRS/B722. The International Classification of Functioning, Disability and Health (ICF) model[25] was used to classify OM into either PBOM or body structure and function OM. Agreement between reviewers (S.A., E.B.) for title and abstract assessment was excellent (94.0%, Kappa = 0.91) and full-text (92.5%, Kappa = 0.80). The third reviewer (N.R.H.) was consulted twice.

Search Two: Measurement Properties
There were 16 studies for measurement properties of PROM, one study for PBOM, and three studies for body structure and function OM (Table 2). Excellent agreement between reviewers (S.A., E.B.) for titles/abstracts (95%, Kappa = 0.92) and substantial agreement for full-text articles (90%, Kappa = 0.78).[35] Eleven authors responded from 21 who were contacted clarifying participants age, language of PROM utilized, or for missing data. The third reviewer (N.R.H.) was consulted four times.

Study and Outcome Measure Characteristics
Detailed information on studies and participant characteristics are shown in Table 2. The OM included were nine PROMs (six disease-specific and three generic), one PBOM, and six body structure and function OM. Detailed description of OMs and their characteristics are shown in Tables 3 and 4.

Risk of Bias
Evaluated measurement properties included, development (n = 1), internal consistency (n = 3), reliability (n = 5), measurement invariance (n = 2), measurement error (n = 2), hypothesis testing for construct validity (n = 18), responsiveness (n = 2). Results of risk bias assessment are presented in supplemental digital content 4, http://links.lww.com/BRS/B723.

Measurement Properties and Synthesis of Evidence
Table 5 shows the summary of findings table for results of measurement properties and the overall evidence for measurement properties against COSMIN and GRADE approach.

Patient-Reported Outcome Measures
Functional scales of SRS-24[19] displayed sufficient discriminative validity in pre and postsurgery individuals with AIS.[38] While, construct validity of SRS-22 function scale was rated insufficient (moderate-quality evidence),[38,40,41] and sufficiently responsive[39] (very low-quality evidence). Measurement invariance of this scale was rated indeterminate since no multiple group factor analysis was performed,[43] and the measurement error rated insufficient.[42] The activity scale of SRS-22r was rated sufficiently reliable as the Interclass Correlation Coefficient (ICC) was 0.76 (0.56– 0.80)

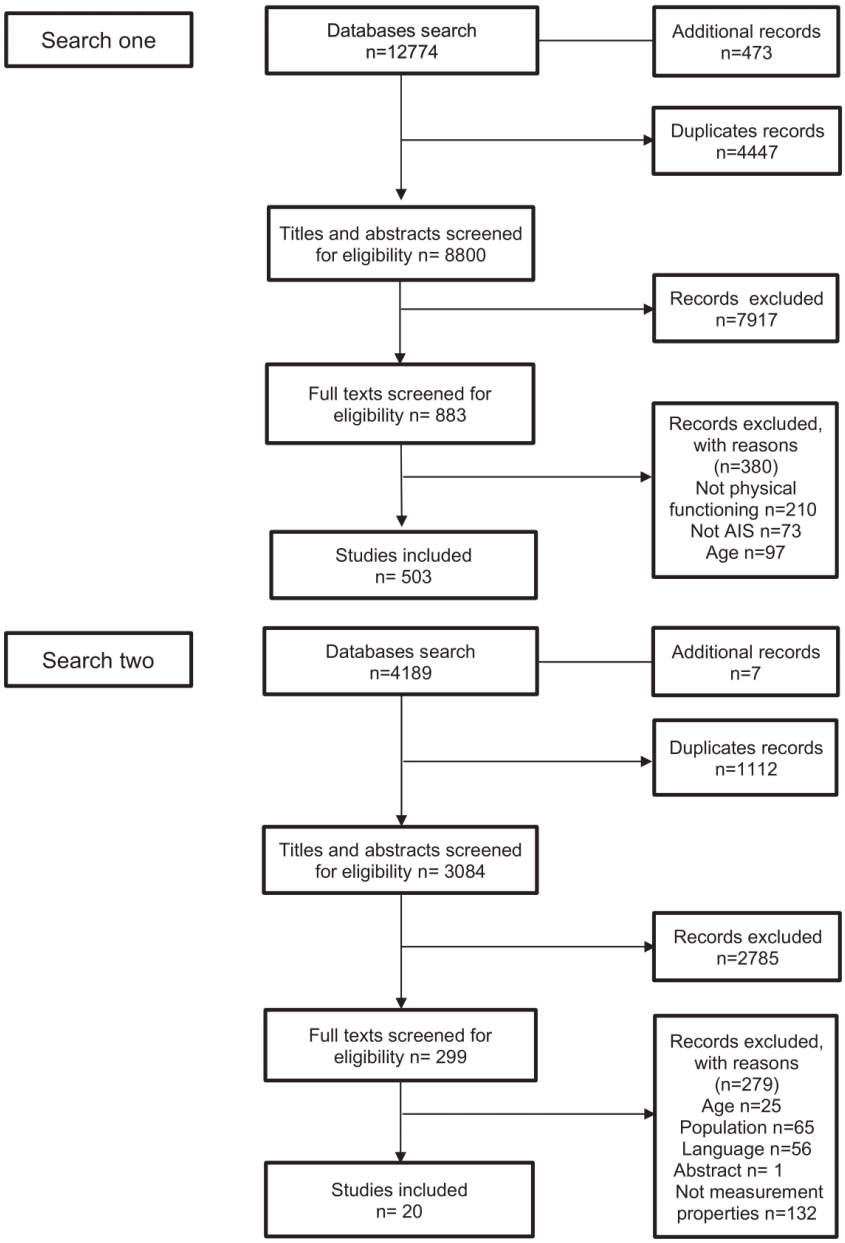

Figure 1. PRISMA flow diagram of both searches and selection process. PRISMA indicates Preferred Reporting Items for Systematic Review and Meta-Analysis.

supported by low-quality evidence. However, internal consistency[18] was rated indeterminate.[33] The SRS-22r showed insufficient measurement error[45] (moderate-quality evidence). A strong correlation between function scale of SRS-22r with mobility scale of Child Health Questionnaire-Child Self-Report Form 87 (CHQ-CF87) (Pearson $r = 0.73$)[18] indicating sufficient convergent validity. While, hypothesis of discriminative validity was not met.[44] Thus,

TABLE 2. Studies and Participants Characteristics

Patient Reported Outcome Measure									
Reference	Name of OM	Country	Age (Mean ± SD) Range	Gender (n)	Sample Size (n)	Curve Type (%), (n)	Curve Size Degree ± SD (n)	Type of Intervention (n)	Score (Mean ± SD)
Feise *et al*[36]	SQLI	Canada	14.9 ± 2.4 (10–18)	F (70) M (14)	84	NR	Unbraced 26.1° ± 10° Braced 34.3° ± 8.7° Postsurgical 31.0 ± 11.4°	Postsurgical (16) Braced (30) Unbraced (24) Control (14)	81.1 ± 15.7
Parent *et al*[37]	SQLI	Canada	14.7 ± 1.9 (8–20)	F (95)	95	Main thoracic (29) Double thoracic (4) Double major (23) Triple major (2) Thoracolumbar/ lumbar (20) Thoracolumbar/lumbar, main thoracic (17)	<30° (34) 30°–50° (44) > 50° (17)	Surgery	NR
Bastrom *et al*[38]	SRS-24, SRS-22	USA	14.8 ± 2 (10–21)	F (81%)	829	Lenke 1 (43%) Lenke 2 (20%) Lenke 3 (7%) Lenke 4 (4%) Lenke 5 (16%) Lenke 6 (10%)	Presurgery 55° ± 13 Post surgery 20° ± 9	Pre and Postsurgery	Presurgery 45° Cobb SRS-22 (4.6 ± 0.5) SRS-24 (4.1 ± 0.5) > 80° Cobb SRS-22 (4.2 ± 0.7) SRS-24 (3.8 ± 0.7) Postsurgery <11° Cobb SRS-22 (4.6 ± 0.5) SRS-24 (4.17 ± 0.5) >29° Cobb SRS-22 (4.61 ± 0.5) SRS-24 (4.17 ± 0.6)
Asher *et al*[39]	SRS-22	USA	16.4 (10.6–47.3)	F (48) M (10)	58	Single (36) Double (19) Triple (3)	63°	Surgery	Function (0 mo) 4.1 Function (3 mo) 3.3 Function (6 mo) 3.9 Function (12 mo) 4.2 Function (24 mo) 4.3
Asher *et al*[40]	SRS-22	USA	Control 13 (10.7–15.4) Non-surgical 14 (9.9–16) Non-surgical untreated 14 (10.8–16) Non-surgical braced 13 (9.9 to –15.2) Presurgery 14 (10.6–15.8)	Control F (15) M (4) Non-surgical F (57) M (11) Non-surgical untreated F (44) M (10) Non-surgical braced F (13) M (1) Presurgery F (31) M (1)	Total (119) Control (19) Nonsurgical (68) Untreated (54) Braced (14) Presurgery (32)	Thoracic, thoracolumbar, lumbar, double triple	Largest cobb angle Non-surgical untreated 27° Braced 31° Presurgery 61°	Brace, presurgery, control	Control (4.5 ± 0.35) Nonsurgical (4.4 ± 0.36) Non-surgical untreated (4.4 ± 0.37) Non-surgical braced (4.5 ± 0.32) Presurgery (4.2 ± 0.42)
Parent *et al*[41]	SRS-22	Canada	13.5–20 (153) Total (18.6 ± 9.2)	F (153)	153	NR	30° (58) 30°–50° (66) 50° (4)	Observation (107) Brace (32) Presurgery (22) Postsurgery (62)	Observation (4.3 ± 0.59) Brace (4.5 ± 0.59) Presurgery (4.2 ± 0.58) Postsurgery (4.1 ± 0.60)
Carreon *et al*[42]	SRS-22	USA	14.3 ± 1.9 (10–18)	F (735) M (152)	887	NR	53° ± 18°	Pre and 1 year postsurgery	Presurgery 4.15 ± 0.55 Postsurgery 4.23 ± 0.46
Verma *et al*[43]	SRS-22	USA and Ghana	15.4	F (100) M (60)	160	NR	Ghana 67.2° USA 52°	Presurgery	Ghana 3.7 ± 0.8 USA 4.2 ± 0.4
Berliner *et al*[44]	SRS-22r	USA	13.8 (11.0–17.2)	F (115) M (40)	155	Non-surgical thoracic (56.5%) Thoracolumbar (38.7%) Lumbar (4.8%) Presurgical thoracic (65.2%) Thoracolumbar (34.8%) Lumbar (0%)	Total 43.1° Nonsurgical 21.9° Presurgical 57.2°	Non-surgical and presurgical	0°–19° (4.5 ± 0.47) 20°–40° (4.4 ± 0.37) 41°–50° (4.1 ± 0.69) 51°–60° (4.2 ± 0.54) >60° (4.3 ± 0.55)

TABLE 2 (*Continued*)

Reference	Name of OM	Country	Age (Mean ± SD) Range	Gender (n)	Sample Size (n)	Curve Type (%), (n)	Curve Size Degree ± SD (n)	Type of Intervention (n)	Score (Mean ± SD)
Patient Reported Outcome Measure									
Kelly *et al*[45]	SRS-22r	USA	14.6 (10–22)	F (1,034) M (247)	1,281	Lenke 1 (552) Lenke 2 (272) Lenke 3 (93) Lenke 4 (46) Lenke 5 (196) Lenke 6 (120)	NR	1, 2 year Postsurgery	Activity MCID (0.08) MDMD (0.24)
Glattes *et al*[18]	SRS-22r, CHQ-CF87	USA	14.1 ± 2.7 (8–18)	F (58) M (12)	Total (70)	NR	29.8° ± 12.3°	Presurgery	SRS-22r (4.5 ± 0.65) CHQ-CF87 (91 ± 15.6)
Fedorak *et al*[46]	PROMIS, SRS22r	USA	14.4 ± 2.1 (11.4–17.4)	F (78.8%) M (21.2%)	113	Thoracic (67%) Thoracolumbar (21.7%) Lumbar (11.3%)	Thoracic kyphosis 34.1 ± 14.9° Lumbar lordosis 54.8 ± 13.3°	Observed, Pre or post-bracing (69.0%) Brace (27.4%) Surgery (3.5%)	PROMIS, Mobility (50.93 ± 9.80) SRS-22r, Function (4.5 ± 0.5)
Roberts *et al*[47]	SRS-30	USA	F (14.0) M (15.2)	F (83.4%) M (16.5%)	744	Risser grade M (mean 3.5) F (mean 3.2)	F (53.3°) M (55.9°)	Presurgery, 2 yr. postsurgery	Presurgery F (4.2) M (4.2) Postsurgery F (4.3) M (4.4)
Lubicky *et al*[48]	SRS-30	USA	15.6 ± 1.7	F (75%)	356	NR	NR	Presurgery, 2 yr. postsurgery	Presurgery (4.18 ± 0.55) Postsurgery (4.34 ± 0.51)
Sarwahi *et al*[49]	SAQ	USA	15 (13 to –17)	F (71) M (24)	95	NR	Presurgery 51.08° Postsurgery 15.98°	NR	NR
Lerman *et al*[20]	PODCI	North America	Parent 15.2 (11.7–18.8) Patient 15.3 (11.7–20.9)	Parent F (88) M (9) Patient F (86) M (9)	102	Thoracic (17) Thoracolumbar (6) Lumbar (7) Double curve (17)	10–29° (n = 23) 30–49° (n = 20) >50° (n = 4)	1 year postsurgery	Upper extremity (96.8 ± 9.9) Transfer (97.6 ± 4.7) Sport & physical function (85.5 ± 17.5) Global function (89.4 ± 9.8)
Performance based outcome measure									
Gao *et al*[61]	TUG	USA	Mild AIS 14.9 ± 1.7 Moderate AIS 16.4 ± 3.3 Severe AIS 15.3 ± 3.1	NR	AIS (30) Control (30)	Right-sided Thoracolumbar	Mild AIS 19.9° ± 4.3 Moderate AIS 31.8° ± 4.2 Severe AIS 53.4° ± 16.1	Pretreatment	TUG (seconds) Mild (6.8 ± 1.5) Moderate (6.9 ± 0.9) Severe (6.5 ± 0.8) Healthy control (6.0 ± 0.6)
Body structure and function outcome measure									
Hresko *et al*[62]	MST	USA	14.2 ± 1.9 (11.3–18.6)	F (37)	37	Thoracic Lumbar	Thoracic 40° ± 20° Lumbar 31° ± 12°	Pretreatment	5.7 ± 2.2 cm
Eyvazov *et al*[63]	MST, FTF test, Axial rotation, LSB, ΔC7-PSIS	China	15.7 ± 4.1	M (12) F (46)	58	Lenke 5 (Thoracolumbar/lumbar)	Group A 25° ± 7.1° Group B 49.8° ± 13.6° Total 34° ± 9.2°	Pretreatment	Modified Schober (cm) Group A: (20.6 ± 1.4) Group B: (20.3 ± 1.2) FTF test (cm) Group A: (10.1 ± 11.2) Group B: (11 ± 10.3) ΔC7-PSIS (27.6 ± 1.8%) LSB (degrees) Group A: (66.6 ± 13.4) Group B: (57.8 ± 14.3) Axial rotation (degrees) Group A: (90.1 ± 21.9) Group B: (5.9 ± 19.6)
Stepien *et al*[24]	TPHA test	Poland	AIS (12.7 ± 2.6) Control (11.8 ± 2.5)	F (98)	Control (49) AIS (49)	Risser sign Grade 0 (14) Grade 1 (11) Grade 2 (6) Grade 3 (3) Grade 4 (9) Grade 5 (6)	Thoracic 27.7° ± 13.4° Lumbar 25.8° ± 10.5°	Physiotherapy	AIS Left TPHA –10.93° ± 4.64° Right TPHA –2.37° ± 8.30° Control Left TPHA –11° ± 3.30° Right TPHA -8.64° ± 4.70°

AIS indicates adolescent idiopathic scoliosis; C7-PSIS, cervical 7 to posterior superior iliac spine; CHQ-CF87, Child Health Questionnaire-Child Self-Report Form 87; F, female; FTF, Fingertip To Floor test; LSB, lateral side bending; M, male; MCID, minimal clinically important difference; MDMD, minimal detectable minimal difference; MST, Modified Schober test; NR, not reported; OM, outcome measure; PODCI, Paediatrics Outcomes Data Collection Instrument; PROMIS, patient-reported outcomes measurement information system; SAQ, sport activity questionnaire; SD, standard deviation; SQLI, scoliosis quality of life index; SRS, Scoliosis Research Society; SRS-22r, Scoliosis Research Society-22 Revised; TPHA, Trunk Pelvis Hip Angle test; TUG, Timed Up and Go test; USA, United State of America.

TABLE 3. Patient-Reported Outcome Measures Characteristics

PROMS	Country	Sub-scale Items (n)	Target Population	Mode of Administration	Recall Period	Response Options	Scoring System	Available Translations
SRS-24[19]	USA	General Function (3) Function after surgery (2) Function-activity (3)	AIS	Self-administrated	Now, postsurgery	Five response options	1–5	–
SRS-22[39]	USA	Function/Activity (5)	AIS	Self-administrated	Now, postsurgery	5 response options	1–5	Turkish,[55] Italian,[56] Spanish,[57] Japanese,[58] Traditional Chinese,[59] Simplified Chinese,[60] Polish,[61] French,[62,63] Thai,[64] Norwegian[65]
SRS-22r[66]	USA	Function/Activity (5)	AIS	Self-administrated	Now, postsurgery	5 response options	1–5	German,[67] Greek,[68] Dutch,[69] Chinese,[59] Brazilian,[70] Italian,[71] Thai,[72] Arabic,[73] Persian,[74] Swedish[75]
SRS-30[76]	USA	Function/Activity (5) postsurgery questions (2)	AIS	Self-administrated	Now, postsurgery	Function/Activity (5 response options) Postsurgery (three response options)	Function (1–5) postsurgery (1–3)	Finnish[77] Brazilian[78]
CHQ-CF87[18]	USA	Physical functioning (9)	Generic	Self-administrated	NR	Four, five, six response options	0–100	–
SQLI[36]	Canada	Physical activity (5)	AIS	Self-administrated	Four weeks	Five response options	0–4	–
SAQ[49]	USA	Total (24) School, gym, carry backpack, bend over, running	AIS	Self-administrated	Postsurgery	NR	NR	–
PROMIS[46]	USA	Mobility	Generic	Self-administrated	7-day	Five response options	Mean T-score 50, SD 10	–
PODCI[50]	North America	Upper extremity functioning, Transfers & basic mobility Sport & physical function Global function	Generic Paediatric orthopaedic conditions	Self-administrated parent-report Adolescents report	NR	3–6	0–100	–

AIS indicates adolescent idiopathic scoliosis; CHQ-CF87, Child Health Questionnaire-Child Self-Report Form 87; NR, not reported; PODCI, Paediatrics Outcomes Data Collection Instrument; PROMIS, patient-reported outcomes measurement information system; SAQ, sport activity questionnaire; SD, standard deviation; SQLI, scoliosis quality of life index; SRS, Scoliosis Research Society; SRS-22r, Scoliosis Research Society-22 revised; USA, United State of America.

TABLE 4. Performance-Based and Body Structure and Function Outcome Measure Characteristics

Outcome Measure (Reference)	Activity	Required Equipment	Number of Trials	Parameter Measured
TUG[51]	Stand from chair, walk 3 m, return, sit down	Chair, stopwatch, walking space	Three trials	Average of time in seconds
MST[52,53]	Marks on PSIS, keep knees straight, bend forward and touch the floor	Tape measure	Two to three trials	Average of distance in cm
FTF test[53]	Stood upright, bend forward and touch the floor	Tape measure	Two trials	Average of distance in cm
C7-PSIS distance[53]	Stand upright, maximally flex and extend neck, distance measured between C7 spinous process and PSIS	Tape measure	Two trials	Average of distance in cm
LSB angles[53]	In upright posture, knees straight, bend to the side without rotation	Goniometer	Two trials	Average angle in degrees between lines joining PSIS and C7
Axial rotation[53]	Seated position, locked both arms in front of body with fixed pelvic, shoulder rotation controlled by a goniometer holder device	Goniometer	Two trials on left and right side	Average angle in degrees
TPHA[54]	Supine, flex and pull lower limbs, then move limbs to the left or right side	Plurimeter	Three times on each side of body	Average of angle in degrees

C7-PSIS indicates cervical 7 to posterior superior iliac spine; FTF, Fingertip To Floor test; LSB, lateral side bending; MST, Modified Schober Test; TPHA, Trunk Pelvis Hip Angle test; TUG, Timed Up and Go test.

evidence for construct validity was downgraded for inconsistency. Moreover, the scale was found unresponsiveness to change (low-quality evidence).[45]

The SRS-30 consists of questions from both SRS-24 and SRS-22. Although no study was identified evaluated its validity or reliability, high-quality evidence indicated that the construct validity of activity scale of SRS-30 was sufficient.[48] A difference in activity scores (0.50) observed at instrumentations construct before and after surgery,[48] while measurement invariance was rated indeterminate.[47]

Scoliosis quality of life index (SQLI) is a modified version of SRS-22 consisting of physical activity domain.[36] Very low evidence demonstrated that its content validity is sufficient based on reviewers' ratings only.[79] The questionnaire was tested for comprehensibility among healthy school children (9.9 years old) only.[36] Per COSMIN guidance, those children may not consider as representative to population of interest.[79] The internal consistency of activity scale was rated indeterminate, while its reliability was insufficient (ICC = 0.46, 0.29–0.63). The evidence was downgraded due to serious risk of bias and imprecision. Moderate-quality evidence showed that construct validity of this scale was sufficient.

Mobility scale of patient-reported outcomes measurement information system (PROMIS)[46] correlated with function scale of SRS-22r (Pearson $r = 0.65$)[46] indicating sufficient construct validity, while functional domains of Paediatrics Outcomes Data Collection Instrument (PODCI) had insufficient construct validity.[50]

Internal consistency of physical functioning scale of (CHQ-CF87)[18] rated indeterminate as evidence of sufficient structural validity is not available,[33] while its reliability scale was sufficient (ICC = 0.73, 0.20–0.85) based on low-quality evidence.

The sport activity questionnaire (SAQ) was developed based on a test-retest method, which is considered a reliability study based on COSMIN definitions.[15] A very low-quality evidence showed that reliability of SAQ was sufficient.

In conclusion, according to COSMIN methodology for a PROM to be recommended for use, it should exhibit any level of sufficient content validity and low level of evidence of sufficient internal consistency.[33] None of the identified PROMs in this review met these criteria, thus we are unable to recommend any of these PROMs for use in individuals with AIS. Furthermore, none of these PROM had a high evidence of insufficient measurement properties. Therefore, these PROMs can be used but it requires further assessment of the quality of its measurement properties to be recommended for use with individuals with AIS.[33]

Performance-Based Outcome Measure

Timed Up and Go test (TUG) is the only performance measure identified in this review with its measurement properties tested in AIS. A difference in the time to perform TUG test was found between individuals with AIS having different curve severity,[51] indicating sufficient construct validity.[51]

Body Structure and Function Measures

The Trunk Pelvis Hip Angle (TPHA) test is used to measure mobility of lumbo-pelvic-hip complex.[54] Moderate-quality

TABLE 5. Summary of Findings Table for the Measurement Properties of Outcome Measure

Measurement Property	Outcome Measure (Subscale)	Summary Result	Overall Rating	Quality of Evidence
Internal consistency	SRS-22r (activity)	$\alpha = 0.82$	?	Moderate (imprecision)
	SQLI (physical activity)	$\alpha = 0.82$ (0.76–0.88)	?	Moderate (imprecision)
	CHQ-CF87 (physical function)	$\alpha = 0.89$	?	Moderate (imprecision)
Reliability	SRS-22r (activity)	ICC = 0.76 (0.56–0.80)	+	Low (one study adequate quality, imprecision)
	SQLI (physical activity)	ICC = 0.46 (0.29 –0.63)	–	Low (one study adequate quality, imprecision)
	CHQ-CF87 (physical function)	ICC = 0.73 (0.20–0.85)	+	Low (one study adequate quality, imprecision)
	SAQ	Kappa $\kappa \geq 0.70$	+	Very low (one study of doubtful quality)
	TPHA test	ICC = 0.85 (0.95–0.98)	+	Moderate (imprecision)
Cross-cultural validity\measurement invariance	SRS-22 (activity)	No multiple group factor analysis performed	?	Very low (one study inadequate quality, imprecision)
	SRS-30 (Function/Activity)	No multiple group factor analysis performed	?	Moderate (one study adequate quality)
Measurement error	SRS-22 (Activity)	SDC (0.24) >MIC (0.08)	–	Moderate (one study of adequate quality)
	SRS-22r	SDC (0.41) > MIC (0.08)	–	Moderate (one study of adequate quality)
Construct validity	SRS-24 (Function)	Two hypotheses confirmed	+	High (one study very good quality)
	SRS-22 (Activity)	Two out of nine hypotheses confirmed	–	Moderate (inconsistency)
	SRS-22r (Function)	Four hypotheses confirmed	+	Moderate (inconsistency)
	SRS-30 (Function)	One hypothesis confirmed	+	High (one study very good quality)
	SQLI (physical activity)	Two hypotheses confirmed	+	Moderate (imprecision)
	PODCI (functional scales)	Two hypotheses out of five confirmed	–	Moderate (one study adequate quality)
	PROMIS (Mobility)	One hypothesis confirmed	+	Moderate (one study adequate quality)
	TUG test	Two hypotheses out of three confirmed	+	Moderate (one study adequate quality)
	MST, FTF Test, C7-PSIS	Three hypotheses not confirmed	–	Moderate (imprecision)
	LSB, axial rotation	Two hypotheses confirmed	+	Moderate (imprecision)
Criterion validity	MST	Not all information for "+" reported	?	Moderate
Responsiveness	SRS-22 (Activity)	Four hypotheses confirmed	+	Very low (one study of doubtful quality, imprecision)
	SRS-22r (Function)	One hypothesis not confirmed	–	Low (one study doubtful quality)

C7-PSIS indicates cervical 7 to posterior superior iliac spine; CHQ-CF87 Child Health Questionnaire-Child Self-Report Form 87; FTF, Fingertip To Floor test; ICC, interclass correlation coefficient; LSB, lateral side bending; MIC, minimal important change; MST, Modified Schober test; PODCI, Paediatrics Outcomes Data Collection Instrument; PROMIS, patient-reported outcomes measurement information system; SAQ, sport activity questionnaire; SDC, small detectable change; SQLI, scoliosis quality of life index; SRS, Scoliosis Research Society; SRS-22r, Scoliosis Research Society-22 revised; TPHA, Trunk Pelvis Hip Angle test; TUG, Timed Up and Go. α = Cronbach alpha, + = sufficient, ? = indeterminate, – = insufficient.

evidence supported sufficient inter-observer reliability of TPHA (ICC > 0.942).[33]

The criterion validity of Modified Schober Test (MST) was rated indeterminate (Very low evidence), as not all required information reported, that is, amount of correlation with radiographs.[52] While, its construct validity rated insufficient.[52]

The construct validity of the Fingertip To Floor Test (FTF) and 7th cervical vertebra to posterior superior iliac spine (C7-PSIS) distance was rated insufficient (moderate-quality

evidence).[53] No difference in scores of these tests was found between individuals with mild and severe curves.[53] On the other hand, construct validity of lateral side bending (LSB) angle and axial rotation was sufficiently different between individuals with severe curves.[53]

Interpretability and Feasibility

Information about interpretability and feasibility aspects of functional scales included in this review are available in supplemental digital content 5, http://links.lww.com/BRS/B724. The majority of these scales had high ceiling effect (20%–44%) and minimal floor effects. An exception to this is physical activity scale of SQLI (minimal ceiling and floor effects).[36,37] The minimal clinical important difference (MCID) reported for activity domain for SRS-22 is 0.08.[42] While minimum detectable measurement difference (MDMD) of activity for SRS-22r is 0.24.[45] Review studies did not report information about response shift and percentage of missing items. Moreover, limited information found about feasibility aspects. Most of the included PROMs are completed within 2 to 3 minutes, and it could be concluded that these PROMs are easy to complete, available in different settings, and available free of charge.

DISCUSSION

This is the first rigorous systematic review identifying OM used to assess physical functioning in individuals with AIS and evaluating their respective measurement properties. Search one enabled the generation of a list of OM and search two revealed a few measurement properties studies; comprising nine PROMs, just one PBOM, and six measures of body structure and function. None of the identified PROMs had evidence of sufficient content validity and sufficient internal consistency.[34] Thus, PROMs identified in this review have the potential to be recommended for use but are yet to have the measurement properties investigated. The current evidence showed limited information on the measurement properties of PBOM and body function and structure measure in individuals with AIS.

Patient-Reported Outcome Measure

This review highlights a gap in evidence on content validity of routinely used PROMs that evaluate physical functioning in individuals with AIS. As COSMIN suggested, content validity is the first and most important measurement property to consider when selecting any PROM.[79] It should be assessed with an interview with both professionals and patients to assess relevance, comprehensiveness, and comprehensibility of items within a PROM.[79] The identified PROMs lack adequate development process, as many were developed in a population whose mean age was higher than that of individuals with AIS.[17,19,66,80] The physical activity scale of SQLI was the only scale where its comprehensibility had been investigated,[36] however using healthy children[36] it is not representative of our population of interest.[79]

The majority of identified measurement properties' studies tested construct validity, which displayed sufficient ratings in most of OMs. Otherwise, internal consistency was undetermined due to lack of evidence of sufficient structural validity. Most of activity scales identified demonstrated high ceiling effects, which affect its ability to assess changes in patient's status.[36]

Performance-Based Outcome Measure

Compared with PROMs just one study has investigated measurement properties of a PBOM[51] where pain[10] and psychological distress[81] may influence the self-reporting of functional ability,[12] it is questionable if PROMs are providing adequate information about actual functional performance of this population. While the TUG test assesses balance, mobility, and walking ability,[51] more evidence-based PBOM are needed to evaluate important and meaningful activities of daily livings for individuals with AIS.

Body Structure and Function Measures

Radiographs, measured using Cobb angle, are the gold standard measure for evaluating spinal curvature.[24] While measurement properties of this measure have been studied before,[27] little attention has given to other measures, such as MST and FTF test. These tests are inexpensive, easy, quick measure that does not expose young spines to ionizing radiation. When adequate measurement properties of these OM established, it could serve as a surrogate to radiographs.

Strengths and Limitations

This review utilized two-search strategy to enable identification of all types of OM used in AIS. Risk of selection bias was minimized by involving two independent reviewers for all stages. Adherence to the COSMIN methodology as preferred approach for systematic review of measurement properties is another strength.[33] However, ratings of studies were determined using lowest score principle, which may underestimate a study's final quality score.[14] A potential limitation of this review is there are few studies investigating measurement properties in individuals with AIS, and some that were included where investigating of measurement property was not a primary aim.

CONCLUSION

A range of measures are used for physical functioning assessment in individuals with AIS. The majority of measurement properties studies identified were for PROM with a paucity of information on PBOM and body structure and function measures. Moreover none of the identified PROM can be recommended for use in AIS. More measurement properties studies are required to support recommendation of these measures for research and clinical practice.

➢ Key Points

- ❑ A two staged search strategy was performed on all types of outcome measure for physical functioning assessment for AIS.

- ❑ Most of studies investigating measurement properties evaluated PROM with paucity of information on PBOM, and body structure and function measures.
- ❑ Based on the COSMIN methodology, none of measures identified in this review can be recommended for use in individuals with AIS.

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References

1. Konieczny MR, Senyurt H, Krauspe R. Epidemiology of adolescent idiopathic scoliosis. *J Child Orthop* 2013;7:3–9.
2. Weinstein SL, Dolan LA, Cheng JC, et al. Adolescent idiopathic scoliosis. *Lancet* 2008;371:1527–37.
3. Hamad A, Ahmed EB, Tsirikos AI. Adolescent idiopathic scoliosis: a comprehensive approach to aetiology, diagnostic assessment and treatment. *J Orthop Trauma* 2017;31:343–9.
4. Makino T, Kaito T, Kashii M, et al. Low back pain and patient-reported QOL outcomes in patients with adolescent idiopathic scoliosis without corrective surgery. *Springerplus* 2015;4:397.
5. Leszczewska J, Czaprowski D, Pawłowska P, et al. Evaluation of the stress level of children with idiopathic scoliosis in relation to the method of treatment and parameters of the deformity. *Sci World J* 2012;2012:538409.
6. Durmala J, Tomalak W, Kotwicki T. Function of the respiratory system in patients with idiopathic scoliosis: reasons for impairment and methods of evaluation. *Stud Health Technol Inform* 2008;135:237–45.
7. Du C, Yu J, Zhang J, et al. Relevant areas of functioning in patients with adolescent idiopathic scoliosis on the International Classification of Functioning, Disability and Health: The patients' perspective. *J Rehabil Med* 2016;48:806–14.
8. Dodd S, Clarke M, Becker L, et al. A taxonomy has been developed for outcomes in medical research to help improve knowledge discovery. *J Clin Epidemiol* 2018;96:84–92.
9. Tomey KM, Sowers MR. Assessment of physical functioning: a conceptual model encompassing environmental factors and individual compensation strategies. *Phys Ther* 2009;89:705–14.
10. Bastrom TP, Marks MC, Yaszay B, et al. Prevalence of postoperative pain in adolescent idiopathic scoliosis and the association with preoperative pain. *Spine (Phila Pa 1976)* 2013;38:1848–52.
11. LaMontagne LL, Hepworth JT, Cohen F, et al. Adolescent scoliosis: effects of corrective surgery, cognitive-behavioral interventions, and age on activity outcomes. *Appl Nurs Res* 2004;17:168–77.
12. Reiman MP, Manske RC. The assessment of function: how is it measured? A clinical perspective. *J Man Manip Ther* 2011;19:91–9.
13. Bean JF, Olveczky DD, Kiely DK, et al. Performance-based versus patient-reported physical function: what are the underlying predictors?. *Phys Ther* 2011;91:1804–11.
14. Mokkink LB, de Vet HCW, Prinsen CAC, et al. COSMIN risk of bias checklist for systematic reviews of patient-reported outcome measures. *Qual Life Res* 2018;27:1171–9.
15. Mokkink LB, Terwee CB, Patrick DL, et al. The COSMIN study reached international consensus on taxonomy, terminology, and definitions of measurement properties for health-related patient-reported outcomes. *J Clin Epidemiol* 2010;63:737–45.
16. Prinsen CAC, Mokkink LB, Bouter LM, et al. COSMIN guideline for systematic reviews of patient-reported outcome measures. *Qual Life Res* 2018;27:1147–57.
17. Asher MA, Min Lai S, Burton DC. Further development and validation of the scoliosis research society (SRS) outcomes instrument. *Spine (Phila Pa 1976)* 2000;25:2381–6.
18. Glattes RC, Burton DC, Lai SM, et al. The reliability and concurrent validity of the Scoliosis Research Society-22r patient questionnaire compared with the Child Health Questionnaire-CF87 patient questionnaire for adolescent spinal deformity. *Spine (Phila Pa 1976)* 2007;32:1778–84.
19. Haher TR, Gorup JM, Shin TM, et al. Results of the Scoliosis Research Society instrument for evaluation of surgical outcome in adolescent idiopathic scoliosis. A multicenter study of 244 patients. *Spine (Phila Pa 1976)* 1999;24:1435–40.
20. de Kleuver M, Faraj SSA, Holewijn RM, et al. Defining a core outcome set for adolescent and young adult patients with a spinal deformity. *Acta Orthop* 2017;88:612–8.
21. Faraj SSA, van Hooff ML, Holewijn RM, et al. Measuring outcomes in adult spinal deformity surgery: a systematic review to identify current strengths, weaknesses and gaps in patient-reported outcome measures. *Eur Spine J* 2017;26:2084–93.
22. Alamrani S, Rushton A, Gardner A, et al. Outcome measures evaluating physical functioning and their measurement properties in adolescent idiopathic scoliosis: a protocol for a systematic review. *BMJ Open* 2020;10:e034286.
23. Moher D, Liberati A, Tetzlaff J, et al. Preferred reporting items for systematic reviews and meta-analyses: the PRISMA statement. *PLoS Med* 2009;6:e1000097.
24. Cobb J. Outline for the study of scoliosis. *Instr Course Lect AAOS* 1948;5:261–75.
25. Santé Omdl, Organization WH, Staff WHO. *International Classification of Functioning, Disability and Health*, ICFed. Geneva: World Health Organization; 2001.
26. Prowse A, Pope R, Gerdhem P, et al. Reliability and validity of inexpensive and easily administered anthropometric clinical evaluation methods of postural asymmetry measurement in adolescent idiopathic scoliosis: a systematic review. *Eur Spine J* 2016;25:450–66.
27. Langensiepen S, Semler O, Sobottke R, et al. Measuring procedures to determine the Cobb angle in idiopathic scoliosis: a systematic review. *Eur Spine J* 2013;22:2360–71.
28. Navarro I, Rosa BND, Candotti CT. Anatomical reference marks, evaluation parameters and reproducibility of surface topography for evaluating the adolescent idiopathic scoliosis: a systematic review with meta-analysis. *Gait Posture* 2019;69:112–20.
29. Wade R, Yang H, McKenna C, et al. A systematic review of the clinical effectiveness of EOS 2D/3D X-ray imaging system. *Eur Spine J* 2013;22:296–304.
30. Fong DY, Lee CF, Cheung KM, et al. A meta-analysis of the clinical effectiveness of school scoliosis screening. *Spine (Phila Pa 1976)* 2010;35:1061–71.
31. Wu HD, Liu W, Wong MS. Reliability and validity of lateral curvature assessments using clinical ultrasound for the patients with scoliosis: a systematic review. *Eur Spine J* 2020;29:717–25.
32. He C, Wong MS. Spinal flexibility assessment on the patients with adolescent idiopathic scoliosis: a literature review. *Spine (Phila Pa 1976)* 2018;43:E250–8.
33. Mokkink LB, Prinsen C, Patrick DL, et al. COSMIN methodology for systematic reviews of Patient-Reported outcome measures (PROMs).*User Manual*. 2018.
34. Balshem H, Helfand M, Schünemann HJ, et al. GRADE guidelines: 3. Rating the quality of evidence. *J Clin Epidemiol* 2011;64:401–6.
35. Cohen J. A coefficient of agreement for nominal scales. *Educ Psychol Meas* 1960;20:37–46.
36. Feise RJ, Donaldson S, Crowther ER, et al. Construction and validation of the scoliosis quality of life index in adolescent idiopathic scoliosis. *Spine (Phila Pa 1976)* 2005;30:1310–5.
37. Parent EC, Hill D, Moreau M, et al. Score distribution of the Scoliosis Quality of Life Index questionnaire in different subgroups of patients with adolescent idiopathic scoliosis. *Spine (Phila Pa 1976)* 2007;32:1767–77.
38. Bastrom TP, Bartley C, Marks MC, et al. Postoperative perfection: ceiling effects and lack of discrimination with both SRS-22 and -24 outcomes instruments in patients with adolescent idiopathic scoliosis. *Spine (Phila Pa 1976)* 2015;40:E1323–9.

39. Asher M, Min Lai S, Burton D, et al. Scoliosis research society-22 patient questionnaire: responsiveness to change associated with surgical treatment. *Spine (Phila Pa 1976)* 2003;28:70–3.
40. Asher M, Min Lai S, Burton D, et al. Discrimination validity of the scoliosis research society-22 patient questionnaire: relationship to idiopathic scoliosis curve pattern and curve size. *Spine (Phila Pa 1976)* 2003;28:74–8.
41. Parent EC, Hill D, Mahood J, et al. Discriminative and predictive validity of the scoliosis research society-22 questionnaire in management and curve-severity subgroups of adolescents with idiopathic scoliosis. *Spine (Phila Pa 1976)* 2009;34:2450–7.
42. Carreon LY, Sanders JO, Diab M, et al. The minimum clinically important difference in Scoliosis Research Society-22 Appearance, Activity, And Pain domains after surgical correction of adolescent idiopathic scoliosis. *Spine (Phila Pa 1976)* 2010;35:2079–83.
43. Verma K, Lonner B, Toombs CS, et al. International utilization of the SRS-22 instrument to assess outcomes in adolescent idiopathic scoliosis: what can we learn from a medical outreach group in Ghana?. *J Pediatr Orthop* 2014;34:503–8.
44. Berliner JL, Verma K, Lonner BS, et al. Discriminative validity of the Scoliosis Research Society 22 questionnaire among five curve-severity subgroups of adolescents with idiopathic scoliosis. *Spine J* 2013;13:127–33.
45. Kelly MP, Lenke LG, Sponseller PD, et al. The minimum detectable measurement difference for the Scoliosis Research Society-22r in adolescent idiopathic scoliosis: a comparison with the minimum clinically important difference. *Spine J* 2019;19:1319–23.
46. Fedorak GT, Larkin K, Heflin JA, et al. Pediatric patient-reported outcomes measurement information system is equivalent to scoliosis research Society-22 in assessing health status in adolescent idiopathic scoliosis. *Spine (Phila Pa 1976)* 2019;44:E1206–10.
47. Roberts DW, Savage JW, Schwartz DG, et al. Male-female differences in Scoliosis Research Society-30 scores in adolescent idiopathic scoliosis. *Spine (Phila Pa 1976)* 2011;36:E53–9.
48. Lubicky JP, Hanson JE, Riley EH. Instrumentation constructs in pediatric patients undergoing deformity correction correlated with Scoliosis Research Society scores. *Spine (Phila Pa 1976)* 2011;36:1692–700.
49. Sarwahi V, Wendolowski S, Gecelter R, et al. When do patients return to physical activities and athletics after scoliosis surgery?: A validated patient questionnaire based study. *Spine (Phila Pa 1976)* 2018;43:167–71.
50. Lerman JA, Sullivan E, Haynes RJ. The Pediatric Outcomes Data Collection Instrument (PODCI) and functional assessment in patients with adolescent or juvenile idiopathic scoliosis and congenital scoliosis or kyphosis. *Spine (Phila Pa 1976)* 2002;27:2052–7; discussion 2057-8.
51. Gao C-C, Chern J-S, Chang C-J, et al. Center of pressure progression patterns during level walking in adolescents with idiopathic scoliosis. *PLoS One* 2019;14:e0212161.
52. Hresko MT, Mesiha M, Richards K, et al. A comparison of methods for measuring spinal motion in female patients with adolescent idiopathic scoliosis. *J Pediatr Orthop* 2006;26:758–63.
53. Eyvazov K, Samartzis D, Cheung JPY. The association of lumbar curve magnitude and spinal range of motion in adolescent idiopathic scoliosis: a cross-sectional study. *BMC Musculoskelet Disord* 2017;18:51.
54. Stępień A, Guzek K, Pałdyna B, et al. The Trunk-Pelvis-Hip Angle test is a reliable measurement of the range of the lower trunk-pelvis rotation in adolescents. *J Orthop Ther* 2018;10:1124.
55. Alanay A, Cil A, Berk H, et al. Reliability and validity of adapted Turkish Version of Scoliosis Research Society-22 (SRS-22) questionnaire. *Spine (Phila Pa 1976)* 2005;30:2464–8.
56. Monticone M, Carabalona R, Negrini S. Reliability of the Scoliosis Research Society-22 Patient Questionnaire (Italian version) in mild adolescent vertebral deformities. *Eura Medicophys* 2004;40:191–7.
57. Bago J, Climent JM, Ey A, et al. The Spanish version of the SRS-22 patient questionnaire for idiopathic scoliosis: transcultural adaptation and reliability analysis. *Spine (Phila Pa 1976)* 2004;29:1676–80.
58. Hashimoto H, Sase T, Arai Y, et al. Validation of a Japanese version of the Scoliosis Research Society-22 Patient Questionnaire among idiopathic scoliosis patients in Japan. *Spine (Phila Pa 1976)* 2007;32:E141–6.
59. Cheung KM, Senkoylu A, Alanay A, et al. Reliability and concurrent validity of the adapted Chinese version of Scoliosis Research Society-22 (SRS-22) questionnaire. *Spine (Phila Pa 1976)* 2007;32:1141–5.
60. Li M, Wang CF, Gu SX, et al. Adapted simplified Chinese (mainland) version of Scoliosis Research Society-22 questionnaire. *Spine (Phila Pa 1976)* 2009;34:1321–4.
61. Glowacki M, Misterska E, Laurentowska M, et al. Polish adaptation of scoliosis research society-22 questionnaire. *Spine (Phila Pa 1976)* 2009;34:1060–5.
62. Beausejour M, Joncas J, Goulet L, et al. Reliability and validity of adapted French Canadian version of Scoliosis Research Society Outcomes Questionnaire (SRS-22) in Quebec. *Spine (Phila Pa 1976)* 2009;34:623–8.
63. Lonjon G, Ilharreborde B, Odent T, et al. Reliability and validity of the French-Canadian Version of the Scoliosis Research Society 22 Questionnaire in France. *Spine (Phila Pa 1976)* 2014;39:E26–34.
64. Leelapattana P, Keorochana G, Johnson J, et al. Reliability and validity of an adapted Thai version of the Scoliosis Research Society-22 questionnaire. *J Child Orthop* 2011;5:35–40.
65. Adobor RD, Rimeslatten S, Keller A, et al. Repeatability, reliability, and concurrent validity of the scoliosis research society-22 questionnaire and EuroQol in patients with adolescent idiopathic scoliosis. *Spine (Phila Pa 1976)* 2010;35:206–9.
66. Asher MA, Lai SM, Glattes RC, et al. Refinement of the SRS-22 Health-Related Quality of Life questionnaire Function domain. *Spine (Phila Pa 1976)* 2006;31:593–7.
67. Niemeyer T, Schubert C, Halm HF, et al. Validity and reliability of an adapted german version of scoliosis research society-22 questionnaire. *Spine (Phila Pa 1976)* 2009;34:818–21.
68. Antonarakos PD, Katranitsa L, Angelis L, et al. Reliability and validity of the adapted Greek version of scoliosis research society - 22 (SRS-22) questionnaire. *Scoliosis* 2009;4:14.
69. Schlosser TP, Stadhouder A, Schimmel JJ, et al. Reliability and validity of the adapted Dutch version of the revised Scoliosis Research Society 22-item questionnaire. *Spine J* 2014;14:1663–72.
70. Camarini PM, Rosanova GC, Gabriel BS, et al. The Brazilian version of the SRS-22r questionnaire for idiopathic scoliosis. *Braz J Phys Ther* 2013;17:494–505.
71. Monticone M, Baiardi P, Calabro D, et al. Development of the Italian version of the revised Scoliosis Research Society-22 Patient Questionnaire, SRS-22r-I: cross-cultural adaptation, factor analysis, reliability, and validity. *Spine (Phila Pa 1976)* 2010;35:E1412–7.
72. Sathira-Angkura V, Pithankuakul K, Sakulpipatana S, et al. Validity and reliability of an adapted Thai version of Scoliosis Research Society-22 questionnaire for adolescent idiopathic scoliosis. *Spine (Phila Pa 1976)* 2012;37:783–7.
73. Haidar RK, Kassak K, Masrouha K, et al. Reliability and validity of an adapted Arabic version of the Scoliosis Research Society-22r Questionnaire. *Spine (Phila Pa 1976)* 2015;40:E971–7.
74. Mousavi SJ, Mobini B, Mehdian H, et al. Reliability and validity of the persian version of the scoliosis research society-22r questionnaire. *Spine (Phila Pa 1976)* 2010;35:784–9.
75. Danielsson AJ, Romberg K. Reliability and validity of the Swedish Version of the Scoliosis Research Society–22 (SRS-22r) patient questionnaire for idiopathic scoliosis. *Spine (Phila Pa 1976)* 2013;38:1875–84.
76. Scoliosis Research Society web site. SRS-30 Patient Questionnaire. Available at: http://www.srs.org/professionals/outcomes/srs-30.pdf. Accessed March 2020.
77. Kyrola K, Jarvenpaa S, Ylinen J, et al. Reliability and validity study of the finnish adaptation of scoliosis research society questionnaire version SRS-30. *Spine (Phila Pa 1976)* 2017;42:943–9.
78. Carrico G, Meves R, Avanzi O. Cross-cultural adaptation and validity of an adapted Brazilian Portuguese version of Scoliosis Research Society-30 questionnaire. *Spine (Phila Pa 1976)* 2012;37:E60–3.

79. Terwee CB, Prinsen CAC, Chiarotto A, et al. COSMIN methodology for evaluating the content validity of patient-reported outcome measures: a Delphi study. *Qual Life Res* 2018;27:1159–70.
80. Asher M, Min Lai S, Burton D, et al. The reliability and concurrent validity of the scoliosis research society-22 patient questionnaire for idiopathic scoliosis. *Spine (Phila Pa 1976)* 2003;28:63–9.
81. Sanders AE, Andras LM, Iantorno SE, et al. Clinically significant psychological and emotional distress in 32% of adolescent idiopathic scoliosis patients. *Spine Deform* 2018;6:435–40.

Appendix 5. Alamrani et al. (2020)

BMJ Open

Outcome measures evaluating physical functioning and their measurement properties in adolescent idiopathic scoliosis: a protocol for a systematic review

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ABSTRACT

Introduction Physical functioning (PF) is the ability to carry out the physical activity of daily living. It is an important outcome that provides a meaningful evaluation of individuals' life. PF can be assessed using patient-reported outcome measures, performance-based outcome measures or body structure and function measure. Measures need to be valid, reliable and responsive to change to evaluate the effects of an intervention. Adolescent idiopathic scoliosis (AIS) is the most common deformity among the paediatric population and impacts on individuals' lives. This systematic review will appraise evidence on the measurement properties of PF tools in individuals with AIS.

Methods/analysis A protocol for systematic review and meta-analysis informed by Cochrane guidelines is reported in line with Preferred Reporting Items for Systematic Reviews and Meta-Analysis-P. MEDLINE, PsycINFO, EMBASE, CINAHL, SPORTdiscus, Web of Science and PubMed will be searched in two stages, from inception until December 2019. Search 1 will inventory all studies that assessed PF in participants with AIS, without any limitations. The search terms will be scoliosis, adolescent and PF-related terms. Search 2 will include studies which investigated instrument measurement properties in the same population for measures identified in search one. Two reviewers will independently perform study selection, data extraction, risk of bias and overall quality assessment. The COnsensus-based Standards for the selection of health Measurement INstruments (COSMIN) risk of bias and a modified Grading of Recommendations, Assessment, Development and Evaluation (GRADE) guidelines will be used. A meta-analysis will be conducted if possible, or the evidence will be synthesised and summarised per measurement property per outcome measure per measurement type.

Ethics and dissemination This review will provide recommendations for practice and future research, considering psychometric properties of outcome measures of PF in AIS. The results of this study will be disseminated through a peer-reviewed publication and conference presentation.

PROSPERO registration number CRD42019142335.

Strengths and limitations of this study

- This review will synthesise evidence of patient-reported, performance-based or body structure and function outcome measures of physical functioning (PF) for use in practice or research involving individuals with adolescent idiopathic scoliosis (AIS).
- The search strategy of this review comprises two stages. The first stage will retrieve all studies that assessed PF in individuals with AIS, while the second stage will retrieve studies that investigated measurement properties of the instrument identified in the first search.
- This study will employ rigorous methods and uses COnsensus-based Standards for the selection of health Measurement INstruments (COSMIN) risk of bias tool and modified Grading of Recommendations, Assessment, Development and Evaluation (GRADE) approach.
- This review will be limited to studies of the English language that assess measurement properties among adolescents with idiopathic scoliosis.

INTRODUCTION

Scoliosis is a complex three-dimensional deformity of the spine, including lateral curvature and rotation of the vertebrae,[1] and characterised by a curve angle ≥10°.[2] There are two main types of scoliosis, idiopathic and non-idiopathic with the latter arising from congenital, neuromuscular or mesenchymal causes.[3] While the aetiology of idiopathic scoliosis remains unknown, genetic, hormonal and mechanical factors are involved.[4] Adolescent idiopathic scoliosis (AIS) often develops between 10 and 16 years of age and represents ~85% of cases.[5] AIS is the most common spinal deformity among the paediatric population, with a prevalence ranging from 2% to 3%.[6] Nearly 80% of those affected presents with a curvature of the

thoracic or thoracolumbar/lumbar region.[3] While men and women are equally affected, women are reported to be at 10 times greater risk of curve progression.[1]

A number of health-related problems are reported among individuals with AIS including lower quality of life,[7] back pain,[8] pulmonary dysfunction,[9] stress[10] and mental health disorders.[11] A major component of health status and health-related quality of life is physical functioning (PF),[12] which can be used to identify individuals at risk of disability and to predict health and social care use.[13 14] Accordingly, PF is included in the core outcome set (COS) for use within clinical trials for many musculoskeletal conditions,[12 15 16] including adolescents with spinal deformity.[16] Where the COS study includes all types of spinal deformity, there is now need for a more specific systematic review of PF outcome measures for this unique population subset. Limitations in PF are reported by individuals with AIS, for example walking, moving around and maintaining body position.[7 17] Additionally, pain is often reported in individuals with AIS, which may cause functional limitations.[8 18 19]

PF can be assessed with patient-reported outcome measures (PROMs), performance-based outcome measures (PBOMs)[20] or a measure of body structure and function. The most widely used PROM for assessment of the quality of life as well as PF of individuals with spinal deformity is the Scoliosis Research Society (SRS) questionnaire[21] and its variants.[22–25] The SRS is mostly used among surgically treated individuals with AIS,[21 24 25] but may not be applicable to those treated conservatively.[25] Although relevant, PROMs should be used cautiously, as it influenced by patients' perception of their abilities to perform activities and lack sensitivity to change.[26] Measures such as PBOMs have the potential to provide unbiased and reproducible assessments of PF during the performance of activities of daily living,[26 27] such as walking speed and trunk endurance testing.[26] Within individuals with AIS, little is known about the available PBOMs for evaluating PF. The body structure and function measures such as radiographs can give an indication about dysfunctions in structure but fail to fully capture functional limitations.[26]

The SRS-22r questionnaire is the reference standard outcome measure of quality of life, which include PF items as recommended by the recent COS study for adolescents and young adults with spinal deformity.[16] However, the SRS-22r fails to fully capture important aspects of PF for individuals with AIS, for example, self-care and mobility.[7] The COS study included all forms of spinal deformities, the heterogeneity limits applicability to individuals with AIS as a discrete population.

Adequate measurement properties of outcome measures are important to avoid the risk of bias and ensure accuracy in the evaluation of test results.[28] The COnsensus-based Standards for the selection of health Measurement INstruments (COSMIN) group developed a taxonomy of measurement properties to improve the selection of outcome measures.[29] Three main domains identified reliability, validity and responsiveness with further subgrouping.[28] A systematic review is needed to evaluate the measurement properties of PF outcome measure for individuals with AIS. Review findings will inform clinicians and researchers on the best available tools for the assessment of PF in AIS. Furthermore, findings will inform future research drawing on a range of measures of PF to investigate health status in AIS.

Objective

To identify outcome measures used to assess PF in individuals with AIS. A secondary aim is to evaluate the measurement properties of PF outcome measures in AIS.

METHODS

This protocol has been informed by experts in musculoskeletal orthopaedics including a consultant spine surgeon, musculoskeletal rehabilitation experts including physiotherapists and individuals with review, measurement properties and research experience. It has been designed in line with the COSMIN methodology for systematic reviews of PROMs[30] and is reported in line with the Preferred Reporting Items for Systematic Reviews and Meta-Analysis-P (PRISMA-P)[31] The search for this systematic review will be conducted in two parts. Stage 1 to identify studies used an outcome measure to evaluate PF in individuals with AIS. This search will allow a list of outcome measures to be generated. Stage 2 will identify studies, which evaluated measurement properties of PF outcome measure identified in the first search.

Search 1: inventory of outcome measures

Eligibility criteria

Study design

All study designs including randomised clinical trials, cohort, observational studies and case studies will be included to identify all outcome measures of PF being used with individuals with AIS. No limitation on language or location will be applied at this stage.

Participants

Participants aged between 10 years and 18 years of age, with a diagnosis of idiopathic scoliosis and ≥10° Cobb angle, will be considered. No restrictions will be applied to the curve severity, evaluation settings and the type of treatment.

Outcome

Any study that includes assessments of the PF of AIS using a specific outcome measure will be included. PF is defined according to the Core Outcome Measures in Effectiveness Trials (COMET) taxonomy,[15] as any physical activities of daily living such as the ability to walk, independence, self-care, performance status and disability index.[15 32] The outcome measures are defined as any one of the following:

1. PROMs in the form of questionnaires or scales designed for AIS to evaluate PF or if it is included as a subscale within a questionnaire.

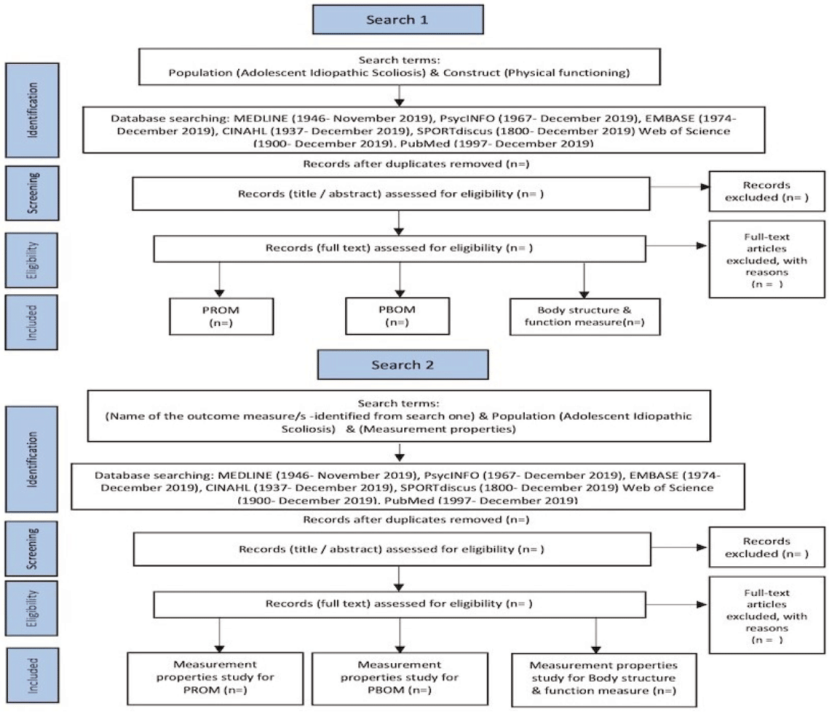

Figure 1 Flow diagram of search strategy (search 1 and search 2) and selection process. PBOM, performance-based outcome measure; PROM, patient-reported outcome measure.

2. PBOMs a measure of PF by clinician while the individual is performing a functional task, for example, walking and/or
3. Body structure and function measures which means any dysfunction in a specific body part or system which may limit function such as range of motion.[26]

Search strategy

A comprehensive, systematic and reproducible search strategy will be completed by one reviewer (SA). Databases will be searched to identify studies that assessed PF among individuals with AIS. To ensure that all relevant studies are included, the type of the outcome measure will not be specified at this stage (figure 1). Initial search terms will be developed for MEDLINE and then adapted with relevant syntax and subject headings for the other databases. An example of the search 1 strategy is available as an online supplementary file 1. As a result of this search, a list of outcome measures for PF used in AIS will be generated. Then, the outcome measures will be classified as, that is, PROM, PBOM or measure of body structure and function. The list will then be used to perform the search 2.

Search 2: measurement properties of outcome measures

Eligibility criteria

Study design

Any study that has evaluated one or more measurement properties of the identified outcome measures in search 1 will be eligible. Only full-text studies available in English will be included. Conference abstracts will be excluded due to the inability to effectively evaluate the quality of the study.

Participants

Participants aged between 10 and 18 years of age, with a diagnosis of idiopathic scoliosis and ≥10° Cobb angle, will be eligible. In studies with mixed cohorts, >50% of participants should be individuals with AIS for the study to be included. Authors of studies will be contacted in case of missing information about number of participants with AIS. Studies without original participant data (eg, systematic review) will be excluded.

Outcome

The outcomes of interest are the measurement properties: reliability including (internal consistency, test–retest, inter-rater and intrarater), measurement error, validity

(content validity, structural validity or criterion validity), hypothesis testing and responsiveness[29] of the outcome measures identified in the search 1 will be eligible. Studies that provide indirect evidence on the measurement properties (by testing an alternative test against an outcome measure of interest and studies in which the outcome measure is used to measure an outcome) will be excluded. Also, studies that only provide normative data will be excluded.

Search strategy

Using the list of outcome measures determined from search 1, one reviewer (SA) will conduct the search. Each category of outcome measure will be searched separately. The search terms will be consisting of the name of the outcome measure/s, the AIS and the measurement properties (figure 1). The recommended search filters specifically designed for retrieving articles on measurement properties will be adapted and used at this stage.[33] An example of the search 2 strategy is available as an online supplementary file 1.

Information sources

The electronic search of databases will be conducted including MEDLINE (1946–November 2019), PsycINFO (1967–December 2019) and EMBASE (1974–December 2019) through OVID interface, CINAHL (1937–December 2019) and SPORTdiscus (1800–December 2019) through EBSCO interface, Web of Science (1900–December 2019) and PubMed (1997–December 2019). No language limitations will be applied in search 1; however, the search 2 will be limited to the full-text article in English. The Web of Science database will be searched for conference proceedings for the last 5 years for search 1 only. A hand search in the key journals including Spine, the Spine Journal, Spine Deformity, Scoliosis and Spinal Disorders and European Spine Journal as well as contacting relevant leading researchers in the field. Further, searching for the grey literature, including British National Bibliography for report literature, opengrey, dissertation abstracts and Electronic Thesis Online Service (EThOS) will be conducted.

Data management

Search records will be imported into Endnote V.X9 (Clarivate Analytics). Using Endnote, the abstracts and full texts will be stored. The duplicates will be identified through the Endnote software and exact duplicates will be removed.

Selection process

A standardised eligibility assessment will be performed by two independent reviewers (SA, Elena Bini (EB)). All studies identified by the search strategy will be assessed based on title/abstract for eligibility. If there is insufficient information to include/exclude study, full text will be retrieved and then screened for eligibility. The study selection (included and excluded studies) with the reasons for exclusion will be summarised in a PRISMA flow diagram.[31] Articles will be included if both reviewers agreed that the eligibility criteria were met. Any disagreement will be first discussed and the third reviewer (NRH) will mediate situations of disagreement. At each assessment stage, agreement between reviewers will be estimated with percentage of agreement and the κ statistic using SPSS for Windows statistical software package (IBM SPSS Statistics V.25).

Data collection process

Two reviewers (SA, EB) will independently extract the data of eligible studies. A bespoke data extraction form will be used and piloted on three studies. Any disagreement between reviewers will be mediated through discussion with a third reviewer (NRH) if needed. If information is not clear or unavailable in the studies, corresponding authors will be contacted to request further details. A second and final reminder will then each be sent 2 weeks apart.

Data items

The data that will be extracted from each study at each stage is summarised in table 1. In the case of missing data, the authors of the study will be contacted.

Outcomes and prioritisation

The gold standard and the primary outcome measure for evaluation of body structure and function (eg, spinal curvature) are the radiographs using the Cobb method.[2] However, no primary PROM or PBOM of PF for individuals with AIS, can be identified for this review.

Risk of bias in individual studies

The COSMIN checklist for assessment of risk of bias and methodological quality in individual studies will be used.[28] It was revised and specifically designed for use in systematic reviews of PROMs to evaluate studies on measurement properties.[34] The methodological quality of each study for each measurement property will be assessed separately.[30] The items for each measurement property in the relevant standards box will be rated as either very good, adequate, doubtful or inadequate quality.[30] Then, the overall methodological quality of the measurement property will be rated based on 'the worst score counts principle', that is, the overall quality of the study for a specific measurement property is based on the lowest rating of any items in the standards' box.[30] The result of each item and overall rating will be reported in the final results. The COSMIN group recommend researchers to adapt the checklist to other measures (ie, PBOMs, body structure and function measure) since it was originally developed for PROMs.[30] Two independent reviewers (SA, EB) will assess the risk of bias for all included studies. Any disagreement will be resolved through discussion, and if no agreement is reached a third reviewer (NRH) will be consulted. The agreement between reviewers will be estimated with percentage agreement and the κ statistic using SPSS for Windows statistical software package (IBM SPSS Statistics V.25) and will be reported in the final results.

Table 1 Summary of items to be extracted from included studies

Study and participants characteristics	Reference, year, country, design of study, age, gender, sample size (used in the analysis), curve size, curve type, type of intervention (bracing, physiotherapy, exercise or surgery)
Outcome measure	PROM: Name of outcome measure, means of scores (SD), mode of administration, recall period, subscale, number of items, response option, response rate, missing items, setting, target population, scoring, original language, available translation
	PBOM: Name of outcome measure, equipment needed, number of assessments, outcome (eg, time needed, ability/disability), setting, scoring
	Body structure and function measure: Name of outcome measure, equipment needed, mode of administration, setting, scoring, outcome (eg, time needed, ability/disability)
Measurement properties	Validity: Name of outcome measure, type of validity, descriptive statistics, missing value, comparator outcome or predictor outcome, hypothesis, statistics method, CI, validation results
	Reliability: Name of outcome measure, type of reliability, descriptive statistic, time interval, reliability coefficient, measurement error
	Responsiveness: Name of outcome measure, Method of testing: *Hypothesis testing, Distribution based method* (ES, SRM and MDC), hypothesis, time to follow-up. *Anchor-based methods* (MIC or MCIC or MID), anchor/s.
	Interpretability: Name of outcome measure, distribution of score in the study population, percentage of missing items, floor and ceiling effects, scores and change scores available for relevant (sub)groups, MIC Or MID, information on response shift
	Feasibility: Patient's comprehensibility, Clinician's comprehensibility, Type and ease of administration, Length of instrument, Completion time, Patient's required mental and physical ability level, Ease of standardisation, Ease of score calculation, Copyright, Cost of an instrument, Required equipment, Availability in different settings, Regulatory agency's requirement for approval

ES, effects size; MCIC, minimal clinically important change; MDC, minimal detectable change; MIC, minimal important change; MID, minimal important difference; PBOM, performance-based outcome measure; PROM, patient-reported outcome measures; SRM, standardised response mean.

Data synthesis

The COSMIN guidelines for systematic reviews will be followed for the synthesis of the results.[30] Characteristics of the outcome measures, sample, measurement properties results, information about interpretability and feasibility of the scores of the included outcome measures will be presented in overview tables for each outcome measure.[30] Each measurement property for each study per tool will be rated against the updated criteria for good measurement properties as either sufficient (+), insufficient (–) or indeterminate (?).[30] The result of rating of measurement property and its methodological quality rating will be added to the overview table.[30] Then, the evidence will be pooled or summarised per measurement property per tool, with the overall result will be rated against the criteria for good measurement properties, and the quality of the evidence will be graded using a modified Grading of Recommendations Assessment, Development and Evaluation (GRADE) approach.[30]

The results on measurement properties from different studies will be pooled in a meta-analysis if there is enough clinical and methodological homogeneity. The data will be statistically pooled when: (1) individuals with AIS displayed similar characteristics in terms of curve severity and intervention; (2) similar base-line score; (3) same time interval; and (4) same statistical parameters. If inconsistent results of measurement properties were present due to different subgroups (ie, mild and sever curve), the consistent results will be separately summarised per subgroup.[30] Pooled estimate of measurement properties will be obtained by calculating weighted means and 95% CI. If deemed not possible to pool the results, a qualitative synthesis will be conducted, for example the percentage of confirmed hypotheses for construct validity will be provided.[30] The pooled or summarised evidence will be rated as sufficient when at least 75% of the results met the criteria. For example, for structural validity, 'at least 75% of the confirmatory factor analysis studies should found the same factor structure'.[30]

The recommendation of an outcome measure will be depending on the measurement properties, as well as interpretability and feasibility results. The tool should have sufficient content validity and at least low-quality evidence for sufficient internal consistency to be recommended for use and the results of this tool is trustworthy.[30]

Confidence in cumulative evidence

Two independent reviewers will assess the quality and strength of evidence for the pooled or summarised result. Using the modified (GRADE) approach, each measurement property per outcome measure in each category will be evaluated. The GRADE approach uses five factors to determine the quality of the evidence: risk of bias (quality of the studies), inconsistency (of the results of the studies), indirectness (evidence comes from different populations, interventions or outcomes than the ones

of interest in the review), imprecision (wide confidence intervals) and publication bias (negative results are less often published).[35] For evaluating measurement properties in systematic reviews of PROMs, only four factors will be assessed as recommended by COSMIN group, while the fifth factor (publication bias) will be removed as there is no registry exists for measurement properties.

DISCUSSION

PF is considered as an important outcome domain in health-related quality of life.[12] It can be used to predict future disability as well as health and social care use.[13] Individuals with AIS reported a limitation in their PF.[7] Thus, measurement of its impact is important in research and clinical practice. Numerous of tools are available for the assessment of PF, ranging from patient-reported to PBOMs. However, it is essential to confirm the psychometric properties of these tools before recommending for clinical use. The COS study for 'all spine deformities' identified the SRS-22r as the recommended PROM for assessment for PF among young adults with spinal deformities.[16] However, there is still a need for a more specific review that evaluate the quality of all outcome measures used in the assessment of PF in AIS including patient-reported and performance-based as well as measures of body structure and function. This systematic review will retrieve all tools that have been used to assess PF among individuals with AIS. Then, it will evaluate and synthesise the quality of studies that report psychometric properties of PF outcome measures in AIS. This review will provide a comprehensive assessment of current evidence which may benefit: (1) health practitioners in selection of the most suitable tools to assess PF in AIS; (2) patients who need a good outcome measures that reflect their actual health status; and (3) researchers and policy maker who can use the recommend measures in designing research trials and defining the COS for individuals with AIS, which in turn will improve health assessment and patient care. Limitations of this review are a focus on individuals with AIS specifically, so recommendations cannot be generalised to other forms of scoliosis.

Patient and public involvement

A study question and systematic review protocol were informed following discussion at a patient and public involvement meeting at the Centre of Precision Rehabilitation for Spinal Pain at the University of Birmingham. The group consisted of individuals with different musculoskeletal and spinal complaints. They actively contributed to research question and to establish the need for systematic review. Since no patient data is needed, patients will not be involved in data collection or analysis. However, the results of the study will be shared at public engagement events.

Implications of this study

AIS is a complex deformity of the spine and causes a significant impact on physical activities of individuals' daily living such as walking and maintaining body position.[7 17] In consequence, the quality of life is affected. PF gives an indication about the current health status and identifies people at risk of disability.[12 13] Therefore, PF is considered as one of the outcomes that should be assessed and reported in clinical trials of musculoskeletal conditions.[15] A systematic review is needed to evaluate current practice in the assessment of PF among individuals with AIS. The results of this review will inform clinicians and researchers on the best available tools for assessment of PF in AIS. This review could provide a research agenda that may highlight the gap in the literature around PF measure and their measurement properties among individuals with AIS.

Ethics and dissemination

No ethics approval is required for this systematic review. The results of this systematic review will be disseminated through peer-reviewed journals as well as international and national conferences presentation. The publications will be split into different publications according to the volume of data. Each category of outcome measures will be published in a separate article.

Twitter Alison Rushton @abrushton, Deborah Falla @Deb_Falla and Nicola R Heneghan @HeneghanNicola

Contributors All authors conceptualised and designed the protocol. SA is a PhD student and NRH (lead supervisor), AR and DF are supervisors and AG is a spinal surgeon. SA drafted the initial manuscript with NRH, AR, DF and AG providing guidance on design, topic, methodology and analyses. All authors reviewed and commented on each draft of the protocol. All authors have approved and contributed to the final manuscript.

Funding SA is a PhD student, supported by a scholarship form University of Tabuk, Tabuk, Saudi Arabia.

Competing interests None declared.

Patient and public involvement Patients and/or the public were involved in the design, or conduct, or reporting, or dissemination plans of this research. Refer to the Methods section for further details.

Patient consent for publication Not required.

Provenance and peer review Not commissioned; externally peer reviewed.

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REFERENCES

1 Reamy BV, Slakey JB. Adolescent idiopathic scoliosis: review and current concepts. *Am Fam Physician* 2001;64:111–6.
2 Cobb JR. The problem of the primary curve. *J Bone Joint Surg Am* 1960;42-A:1413–25.
3 Konieczny MR, Senyurt H, Krauspe R. Epidemiology of adolescent idiopathic scoliosis. *J Child Orthop* 2013;7:3–9.

4 Hamad A, Ahmed EB, Tsirikos AI. Adolescent idiopathic scoliosis: a comprehensive approach to aetiology, diagnostic assessment and treatment. *Orthop Trauma* 2017;31:343–9.
5 Horne JP, Flannery R, Usman S. Adolescent idiopathic scoliosis: diagnosis and management. *Am Fam Physician* 2014;89:193–8.
6 Lonstein D. Adolescent idiopathic scoliosis. *The Lancet* 1994;344:1407–12.
7 Du C, Yu J, Zhang J, *et al*. Relevant areas of functioning in patients with adolescent idiopathic scoliosis on the International classification of functioning, disability and health: the patients' perspective. *J Rehabil Med* 2016;48:806–14.
8 Makino T, Kaito T, Kashii M, *et al*. Low back pain and patient-reported QOL outcomes in patients with adolescent idiopathic scoliosis without corrective surgery. *Springerplus* 2015;4:397.
9 Durmala J, Tomalak W, Kotwicki T. Function of the respiratory system in patients with idiopathic scoliosis: reasons for impairment and methods of evaluation. *Stud Health Technol Inform* 2008;135:237–45.
10 Leszczewska J, Czaprowski D, Pawłowska P, *et al*. Evaluation of the stress level of children with idiopathic scoliosis in relation to the method of treatment and parameters of the deformity. *ScientificWorldJournal* 2012;2012:1–5.
11 Malmqvist M, Tropp H, Lyth J, *et al*. Patients with idiopathic scoliosis run an increased risk of schizophrenia. *Spine Deform* 2019;7:262–6.
12 Taylor AM, Phillips K, Patel KV, *et al*. Assessment of physical function and participation in chronic pain clinical trials: IMMPACT/OMERACT recommendations. *Pain* 2016;157:1836–50.
13 Tomey KM, Sowers MR. Assessment of physical functioning: a conceptual model encompassing environmental factors and individual compensation strategies. *Phys Ther* 2009;89:705–14.
14 Cooper R, Kuh D, Cooper C, *et al*. Objective measures of physical capability and subsequent health: a systematic review. *Age Ageing* 2011;40:14–23.
15 Dodd S, Clarke M, Becker L, *et al*. A taxonomy has been developed for outcomes in medical research to help improve knowledge discovery. *J Clin Epidemiol* 2018;96:84–92.
16 de Kleuver M, Faraj SSA, Holewijn RM, *et al*. Defining a core outcome set for adolescent and young adult patients with a spinal deformity. *Acta Orthop* 2017;88:612–8.
17 Kibsgård T, Brox JI, Reikerås O. Physical and mental health in young adults operated on for idiopathic scoliosis. *J Orthop Sci* 2004;9:360–3.
18 Bastrom TP, Marks MC, Yaszay B, *et al*. Prevalence of postoperative pain in adolescent idiopathic scoliosis and the association with preoperative pain. *Spine* 2013;38:1848–52.
19 Seki H, Ideno S, Ishihara T, *et al*. Postoperative pain management in patients undergoing posterior spinal fusion for adolescent idiopathic scoliosis: a narrative review. *Scoliosis Spinal Disord* 2018;13:17.
20 Ward MM. Interpreting measurements of physical function in clinical trials. *Ann Rheum Dis* 2007;66 Suppl 3:iii32–4.
21 Haher TR, Gorup JM, Shin TM, *et al*. Results of the scoliosis research Society instrument for evaluation of surgical outcome in adolescent idiopathic scoliosis. A multicenter study of 244 patients. *Spine* 1999;24:1435–40.
22 Baldus C, Bridwell K, Harrast J, *et al*. The scoliosis research Society health-related quality of life (SRS-30) age-gender normative data: an analysis of 1346 adult subjects unaffected by scoliosis. *Spine* 2011;36:1154–62.
23 Chen AF, Bi W, Singhabahu D, *et al*. Converting scoliosis research Society-24 to scoliosis research Society-22r in a Surgical-Range, Medical/Interventional adolescent idiopathic scoliosis patient cohort. *Spine Deform* 2013;1:108–14.
24 Asher M, Min Lai S, Burton D, *et al*. Discrimination validity of the scoliosis research society-22 patient questionnaire: relationship to idiopathic scoliosis curve pattern and curve size. *Spine* 2003;28:74–8.
25 Asher MA, Min Lai S, Burton DC. Further development and validation of the scoliosis research Society (SRS) outcomes instrument. *Spine* 2000;25:2381–6.
26 Reiman MP, Manske RC. The assessment of function: how is it measured? a clinical perspective. *J Man Manip Ther* 2011;19:91–9.
27 Bean JF, Olveczky DD, Kiely DK, *et al*. Performance-Based versus patient-reported physical function: what are the underlying predictors? *Phys Ther* 2011;91:1804–11.
28 Mokkink LB, de Vet HCW, Prinsen CAC, *et al*. COSMIN risk of bias checklist for systematic reviews of patient-reported outcome measures. *Qual Life Res* 2018;27:1171–9.
29 Mokkink LB, Terwee CB, Patrick DL, *et al*. The COSMIN study reached international consensus on taxonomy, terminology, and definitions of measurement properties for health-related patient-reported outcomes. *J Clin Epidemiol* 2010;63:737–45.
30 Prinsen CAC, Mokkink LB, Bouter LM, *et al*. COSMIN guideline for systematic reviews of patient-reported outcome measures. *Qual Life Res* 2018;27:1147–57.
31 Moher D, Shamseer L, Clarke M, *et al*. Preferred reporting items for systematic review and meta-analysis protocols (PRISMA-P) 2015 statement. *Syst Rev* 2015;4:1.
32 Williamson PR, Altman DG, Bagley H, *et al*. The comet Handbook: version 1.0. *Trials* 2017;18:280.
33 Terwee CB, Jansma EP, Riphagen II, *et al*. Development of a methodological PubMed search filter for finding studies on measurement properties of measurement instruments. *Qual Life Res* 2009;18:1115–23.
34 Mokkink LB, Terwee CB, Patrick DL, *et al*. The COSMIN checklist for assessing the methodological quality of studies on measurement properties of health status measurement instruments: an international Delphi study. *Qual Life Res* 2010;19:539–49.
35 Balshem H, Helfand M, Schünemann HJ, *et al*. Grade guidelines: 3. rating the quality of evidence. *J Clin Epidemiol* 2011;64:401–6.

Appendix 6. Protocol , Ethical Review and STROBE Checklist for *Chapter Two*

FULL/LONG TITLE OF THE TRIAL

Assessment of Functional Outcomes of Surgically Managed Adolescents with Idiopathic Scoliosis

SHORT TRIAL TITLE / ACRONYM

Functional **O**utcomes of **A**dolescents with **I**diopathic **S**coliosis (**FOAIS)**

PROTOCOL VERSION NUMBER AND DATE

Version 1.0 July 2019

RESEARCH REFERENCE NUMBERS

IRAS Number:	**274218**
EudraCT Number:	
SPONSORS Number:	

This protocol has regard for the HRA guidance

SIGNATURE PAGE

The undersigned confirm that the following protocol has been agreed and accepted and that the Chief Investigator agrees to conduct the trial in compliance with the approved protocol and will adhere to the principles outlined in the Medicines for Human Use (Clinical Trials) Regulations 2004 (SI 2004/1031), amended regulations (SI 2006/1928) and any subsequent amendments of the clinical trial regulations, GCP guidelines, the Sponsor's (and any other relevant) SOPs, and other regulatory requirements as amended.

I agree to ensure that the confidential information contained in this document will not be used for any other purpose other than the evaluation or conduct of the clinical investigation without the prior written consent of the Sponsor

I also confirm that I will make the findings of the trial publicly available through publication or other dissemination tools without any unnecessary delay and that an honest accurate and transparent account of the trial will be given; and that any discrepancies and serious breaches of GCP from the trial as planned in this protocol will be explained.

Lead investigator Date: ...../...../.....

Signature:

Name : Samia Alamrani

Position: PhD student

Chief Investigator: Dr Nicola Heneghan

Signature: Date: ...../...../.....

Sponsor Statement:

Where the University of Birmingham takes on the sponsor role for protocol development oversight, the signing of the IRAS form by the sponsor will serve as confirmation of approval of this protocol.

TABLE OF CONTENTS

KEY STUDY CONTACTS

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Sponsor	
Joint sponsor	None
Funder(s)	None

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Committees	**Study Management Group:** Dr Nicola Heneghan (Chief Investigator) Mrs. Samia Alamrani (lead investigator) Dr Alison Rushton Prof Deborah Falla Mr. Adrian Gardner Mr. Andrew Emms **Study Steering Group:** Dr Nicola Heneghan (Chief Investigator) Mrs. Samia Alamrani (lead investigator) Dr Alison Rushton Dr Deborah Falla Mr. Adrian Gardner Mr. Andrew Emms

STUDY SUMMARY

Adolescent Idiopathic Scoliosis (AIS) is a complex three-dimensional deformity of the spine, including lateral curvature and rotation of the vertebrae [1]. AIS is the most common spinal deformity among the pediatric population, with a prevalence ranging from 2% to 3% , depending on the defined Cobb angles, age and gender [2]. AIS often diagnosed among adolescent between 10 and 16 years of age [3]. A number of health-related problems are reported among individuals as a result of AIS including; lower quality of life , back pain [4], pulmonary dysfunction [5], stress [6], and mental health disorders [7] . In addition, functional limitations can exist such as difficulty walking and maintaining body position [8]. Functioning is considered as a major component of health status and health-related quality of life [9].It can be used to identify individuals at risk of disability and to predict health and social care use [9]. The spinal fusion surgery aim to correct deformity, however it affects spinal flexibility and mobility [10], which in turns has an affect the overall quality of life [11]. The gold standard tool for assessment of quality of life for individuals with AIS, is the Scoliosis Research Society Questionnaire (SRS), as recommended by Scoliosis Research Society [12]. It consists of five domains including mental health, function, satisfaction, pain and self-image [13, 14]. Individuals with AIS usually self-report their functional abilities using functional subscale of SRS-22 questionnaire [15]. Within, the scoliosis literature little attention has been paid to the effect of the scoliosis deformity and the correction surgery on the functional abilities of individuals with AIS. Further, the association between self-reporting of function and the radiographic parameters such as curve size and shape and the clinical parameters such as pain and the type of surgery is not fully explored. The SRS-22 , radiological measurements and clinical parameters are routinely collected and stored on trust databases. Trust databases are a valuable source of evidence from which we can evaluate the functional outcomes of individuals with AIS prior and after surgery. To our knowledge, function domain of the SRS-22 and the associated factors such as curve shape and size, surgery type and pain is not well explored within the trust database.

Study Title	Assessment of Functional Outcomes of Surgically Managed Adolescents with Idiopathic Scoliosis	
Internal ref. no. (or short title)	**F**unctional **O**utcomes among **A**dolescents with **I**diopathic **S**coliosis (**FOAIS)**	
Study Design	Retrospective observational study design	
Study Participants	Patients aged between 10 and 18 years old, diagnosed with AIS with a Cobb Angle of at least 40° pre-surgery.	
	Objectives	Outcome Measures
Primary	○ To evaluate function of individuals with AIS post-surgery	SRS-22- function domain pre-surgery and post-surgery
Secondary	○ To investigate any association between function domain and the following 1. Curve angle 2. Curve Shape 3. Pain 4. Type of surgery 5. Length of instrumentation	SRS-22- function domain Cobb degrees using radiographs LENKE classification system SRS-22- Pain domain Anterior or posterior surgery and length of instrumentation Number of vertebra fused

FUNDING AND SUPPORT IN KIND

FUNDER(S) (Names and contact details of ALL organizations providing funding and/or support in kind for this trial)	**FINANCIAL AND NON FINANCIALSUPPORT GIVEN**
University of Tabuk / Tabuk- Saudi Arabia	Scholarship for the SA lead investigator and PhD student

ROLE OF TRIAL SPONSOR AND FUNDER

The sponsor is the University of Birmingham which assumes overall responsibility for the initiation and management of the study. The sponsor has no direct role in study design, conduct, data analysis and interpretation, manuscript writing, and dissemination of findings, except for their responsibility for oversight. The sponsor does not control the final decision regarding any of these study aspects. Identification of the study sponsor provides transparency and accountability.

ROLES AND RESPONSIBILITIES OF SUDY MANAGEMENT COMMITEES/GROUPS & INDIVIDUALS

The study will be coordinated by a **Study Management Group (SMG).** The **SMG** will comprise all co-investigators involved in the design and interpretation of the study and identification of the participants including (the lead investigator, (SA), PhD supervisory team (AR, DF, NH), site lead surgeon and physiotherapist (AG, AE). The SMG will meet regularly face-to-face and/or online via skype to monitor progress and address any logistical issues raised.

The **Study Steering Group (SSG)** will overview the study and analyses to inform data interpretation. The SSG will include academics from the University of Birmingham with expertise in designing and conducting observational studies who are well placed to guide the study.

The Study will be conducted in accordance with the principles of the Research Governance Framework for Health and Social Care. National Health Service (NHS) ethical approval, Health Research Authority (HRA) approval and individual site Confirmation of Capacity and Capability will be obtained for this study. The Study will follow Good Clinical Practice (GCP) principles and a pre-defined protocol. Pseudonymized participant data will be stored confidentially for 10 years in the protected Redcap data storage system in the Centre for Precision Rehabilitation of Spinal Pain (CPR Spine) at the School of Sport and Exercise, Rehabilitation Sciences, at the University of Birmingham, in accordance with GDPR and the Data Protection Act 2018 and University of Birmingham's research governance frameworks

PROTOCOL CONTRIBUTORS

Mrs. Samia Alamrani, Dr Nicola Heneghan, Dr Alison Rushton, Professor Deborah Falla, Mr Adrian Gardner

PATIENT AND PUBLIC INVOLVEMENT

This study is informed by suggestions made by healthcare professionals. It was conceived directly because of gaps identified in a systematic review of the physical functioning outcome measures among AIS individuals. The study questions and systematic review protocol were informed following discussion at a patient and public involvement meeting at the Centre of Precision Rehabilitation for Spinal Pain (CPR) at the University of Birmingham.

Key Words: Retrospective, Adolescent Idiopathic Scoliosis, Functional Outcomes, Surgery, Function

STUDY FLOW CHART

Pre-surgery data of eligible participants diagnosed with AIS (curve angle ≥ 40°) including:

- Demographic information i.e. (Gender, Age, Hight, Weight)
- SRS-22 Function domain
- SRS-22- Pain domain
- Curve angle (Cobb degrees)
- Curve shape (LENKE classification system)

Post-surgery data of participants following surgery including:

- SRS-22 Function domain
- SRS-22- Pain domain
- Type of surgery
- Length of instrumentation

Statistical analysis of the eligible data

1- Descriptive statistics of participant's characteristics including means, ranges, and standard deviations for Continuous data and medians and ranges for ordinal data.
2- Paired t-test will be used to compare difference in preoperative and postoperative variables.
3- Person's correlation will be used to test the relationship between Continuous variables and regression analysis for the association between Continuous and nominal data.

STUDY PROTOCOL

1 BACKGROUND

Scoliosis is a complex three-dimensional deformity of the spine, including lateral curvature and rotation of the vertebrae [1] and characterised by a curve angle ≥10°[17] . There are two main types of scoliosis, idiopathic and non-idiopathic with the latter arising from congenital, neuromuscular or mesenchymal causes [2]. While the etiology of idiopathic scoliosis remains unknown; genetic, hormonal, and mechanical factors are involved [18]. Adolescent Idiopathic Scoliosis (AIS) often develops between 10 and 16 years of age and represents ~85% of cases [19]. AIS is the most common spinal deformity among the pediatric population, with a prevalence ranging from 2% to 3% [20]. Nearly 80% of those affected presents with a curvature of the thoracic or thoracolumbar/lumbar region [2]. Whilst males and females are equally affected, females are reported to be at 10 times greater risk of curve progression [1] .

A number of health-related problems are reported among AIS individuals including; lower quality of life [8], back pain [4] , pulmonary dysfunction [5], stress [6], and mental health disorders [7]. A major component of health status and health-related quality of life is functioning [21], which can be used to identify individuals at risk of disability and to predict health and social care use [9]. Functioning is included as a discrete domain in the Core Outcome Set (COS) and has been used in clinical trials for many musculoskeletal conditions [22], including adolescents with spinal deformity [23]. Limitations in functioning are reported by individuals with AIS, e.g. walking and maintaining body position [8, 24, 25]. Additionally, pain is often reported in individuals with AIS which may cause functional limitations [26-28]. The spinal fusion surgery aims to correct deformity, however, depending on the length of instrumentation, the surgery has an effect on the spinal mobility and flexibility [10], which will in turns affect the quality of life. The Scoliosis Research Society-22 (SRS) Questionnaire and variants of such [29, 30] is the most widely used Patient Reported Outcome Measure for assessment of the quality of life including function of individuals with spinal deformity [30, 31]. The SRS-22 is mostly used for surgically treated AIS individuals [32]. It consists of five domains including mental health, function, satisfaction, pain and self-image [32-34]. Although SRS does not include all functional activities, it can offer insight into the effect of deformity on function. Some previous studies examined the association of radiological parameters with the SRS overall score [35, 36]. With little attention given to the functional-subscale and factors that may cause functional limitation. An a association between physical functional domain score of short-form 36 questionnaire and between the preoperative Cobb angle [11] and between the length of instrumentation and spinal mobility [10]. Further, lumbar spinal mobility were found correlated with physical function, as measured by the Physical Functioning subscale of SF-36 as well as Oswestry Disability Index [37]. While, in another a long term follow up study, no correlations found between spine mobility and total score SRS scores [38]. Thus, a study is needed to examine the effect of surgery on function as assessed by self-report of functional domain of SRS-22. Also, to examine the factors that has an effect on function, such as pain, type of surgery and radiological parameter among individuals with AIS.

2 RATIONALE

Our systematic review (manuscript under revision) examining the outcome measures of physical functioning among AIS individuals, highlighted the importance of assessment of function in individuals with AIS. A limitation in physical activity e.g. walking and moving around was reported by individuals with AIS [8]. Several studies have examined the effect of surgery on the functional outcomes. However, to our knowledge no previous studies focused on the functional domain of SRS-22 questionnaire and it is association with the clinical and radiological parameters among AIS population within the trust database. Therefore, further research is required to determine the effect of deformity on function following surgery and examine the association between different variables such as the curve type and size, surgery type and pain , surgery and length of instrumentation with function. Routinely collected and electronically stored the SRS-22 and radiological measurements are available on trust databases. This information has increased over the past decade and provides a valuable resource to enable evaluation of clinical management using key outcomes; reflecting data from real clinical practice. Trust databases are a valuable source of evidence from which we can evaluate the association between self-reporting of function by the SRS-22 questionnaire and the clinical and radiological parameters among individuals with AIS.

2.1 Assessment and management of risk

The study is retrospective evaluation of patient data collected previously, thus no risks are identified.

3 OBJECTIVES AND OUTCOME MEASURES/ENDPOINTS

Aim: To evaluate functional outcomes of individuals with AIS following surgery.

3.1 Objectives

1. To evaluate function of AIS individual's pre-surgery.
2. To evaluate function of AIS individual's post-surgery
3. To examine the association between function with
    a. Curve size
    b. Curve shape classified according to the LENKE classification system
    c. Pain
    d. Type of surgery
    e. Length of instrumentation

3. 2 Outcome Measures

- Function can be assessed using the SRS-22- function domain, which is a valid, reliable and responsive to change in the evaluation of quality of life of adolescents with idiopathic scoliosis [13, 14, 30, 32].The functional domain was refined and validated for AIS individuals [15].

- Cobb angle (degrees) measured by radiographs (AP X-rays), topography or MRI [39].

- Pain which can be assessed using the SRS-22- Pain domain. The SRS-22 is a valid, reliable and responsiveness to change tool for quality of life of adolescent with idiopathic scoliosis [13, 14, 30, 32]
- Curve shape determined according to the LENKE classification system [40]. The Lenke classification system divides idiopathic scoliosis curves into 6 types: Type 1 indicating primary thoracic scoliosis, Type 2 being double thoracic scoliosis, Type 3 representing double major scoliosis, Type 4 being triple major scoliosis, Type 5 indicating primary thoracolumbar/lumbar scoliosis with nonstructural thoracic curve, and Type 6 signifying primary thoracolumbar/lumbar scoliosis with a structural thoracic curve. Then, each type of curve is divided into lumbar spine modifiers: A, B, or C according to the preoperative relationship of the center sacral vertical line (CSVL) to the apex of the lumbar spine [40].

- Type of surgery (anterior instrumentation or posterior fusion)

- Length of instrumentation (number of vertebra fused)

3.3 Table of Outcomes

Objectives	Outcome Measures	Time point(s) of evaluation of this outcome measure (if applicable)
• To evaluate function post-surgery among AIS individuals	SRS-22- function domain	Before surgery and after surgery
• To determine the association between function with 1. Curve angle 2. Curve Shape 3. Pain 4. Type of surgery	SRS-22- function domain Cobb degrees Using Lenke classification system SRS-22- Pain domain Surgery type	Before surgery and After surgery

4 STUDY DESIGN

This is a retrospective observational cohort study design of adolescent with idiopathic scoliosis.

5 STUDY SETTING

The study will use the Royal Orthopaedic Hospital NHS Trust database at Birmingham, UK.

6 PARTICIPANT ELIGIBILITY CRITERIA

6.1 Inclusion criteria

Participant data will be included in the study analysis if they fulfil the following criteria:

- Age range between 10 to 18 years old
- Diagnosis of idiopathic scoliosis
- Cobb angle ≥ 40° pre-surgery
- Pre and post-surgery completed data including:
    - SRS-22- function domain
    - SRS-22- Pain domain

6.2 Exclusion criteria

Participant data will be excluded if:

- Incomplete data about function before and after surgery.

7 STUDY PROCEDURES

This is a retrospective observational study aiming to evaluate function among AIS individuals' pre and post-surgery. A secondary aim is to investigate association of pain, Cobb degrees, curve shape, type of surgery and length of instrumentation with function. These data are collected from patients in routine practice and stored at the Royal Orthopaedic Hospital NHS Trust. Only relevant data to the study protocol will be used and analyzed. The patient data will be anonymized and will be checked for accuracy by 2 researchers independently (SA, AG). Where missing data can be retrieved through hospital record systems, this will be agreed through standard trust procedures facilitated by trust staff who are co-investigators within this study.

7.1 Recruitment

Retrospective analysis of trust standardised outcome data and so not applicable.

7.1.1 Participants Identification

Using the Royal Orthopaedic Hospital NHS Trust database, the co-investigator who is trust staff (AG) and part from the patient clinical care team will identify the potential participants. The lead researcher (SA) will check the accuracy of data. Only data directly relevant to objective and outcome measures identified in the study protocol will be retrieved and included in the analysis.

7.2 Consent

This is a retrospective review of previously collected routine care data with no need for further interactions with the participants. There is no danger to the participants and no findings that will affect a participant's individual care. Thus consent is not required.

8 STATISTICS AND DATA ANALYSIS

8.1 Sample size calculation

We will retrieve data of eligible patient from the ROH trust database from 2007 till 2019.

8.2 Statistical analysis plan

The data will be evaluated using the Statistical Package for Social Science (SPSS) version 24.0 software. Descriptive statistics including (frequency, mean, and standard deviation (SD)) will be reported for Continuous and medians and ranges for ordinal data. A statistical significance level will set on $p < 0.05$. Paired t-test will be used to compare the changes on function before and following surgery. Person's correlation will be used to test the relationship between Continuous variables and regression analysis for the association between Continuous and nominal variable.

9 DATA MANAGEMENT

9.1 Access to Data

Direct access will be granted to authorize representatives from the Sponsor, host institution and the regulatory authorities to permit study-related monitoring, audits, and inspections.

9.2 Archiving

Archiving will be authorized by the Sponsor following submission of the end of study report. All essential documents and data will be archived in the secure Redcap system for a minimum of 10 years after completion of study. Destruction of essential documents will require authorization from the Sponsor

10 ETHICAL AND REGULATORY CONSIDERATIONS

10.1 Research Ethics Committee (REC) review& reports

Before commencing the study, a favorable opinion will be sought from a Research Ethics Committee (REC) for the study protocol and all other relevant documents. Any protocol amendments that require review by the NHS REC will not be implemented until that review is in place and other mechanism are in place to implement at the ROH. All correspondence with the REC will be retained. If required, an Annual Progress Report (APR) will be submitted to the REC within 30 days of the anniversary date on which the favorable opinion was given, and annually until the study is declared ended. If the study is ended prematurely, the Chief Investigator will notify the REC, including the reasons for the premature termination. Within one year after the end of the study, the Chief Investigator will submit a final report with the results, including any publications/abstracts, to the REC. The results will be presented to the specialists who work at, and any groups of adolescents with idiopathic scoliosis associated with The Royal Orthopaedic Hospital NHS Trust.

10.2 Peer review

Expert, independent and proportionate. The study rationale, methodology, and intended analysis have been independently peer reviewed by specialists' surgeons and physiotherapists' expert in the field. This has afforded independent, high quality and proportionate peer review.

10.3 Public and Patient Involvement

This study is informed by suggestions made by healthcare professionals. The results of this study will be shared in patient and public involvement meeting at the Centre of Precision Rehabilitation for Spinal Pain (CPR) at the University of Birmingham.

10.4 Regulatory Compliance

Protocol deviations, non-compliances, or breaches are departures from the approved protocol. Accidental protocol deviations can happen at any time. They will be adequately documented on the relevant forms and reported to the Chief Investigator and Sponsor immediately. Deviations from the protocol which are found to frequently recur are not acceptable and will necessitate immediate action and could potentially be classified as a serious breach.

10.5 Data protection and patient confidentiality

All investigators and study site staff will comply with the requirements of the GDPR and Data Protection Act 2018 with regards to the collection, storage, processing and disclosure of personal information and will uphold the Act's core principles. Patient's data will be anonymized once collected and before it goes on to the Redcap system within CPR spine. Participant data will be stored under an allocated number with no identifying features or personal data. To ensure that no participant can be identified, information emanating from this program of work will only be

presented / published in a completely unattributable format or at an aggregate level. Data can only be accessed by members of the research / supervisory team.
Data will be confidentially stored in the secure Redcap system within CPR Spine, and for data analysis purposes on password protected computers at the School of Sport, Exercise and Rehabilitation Sciences, University of Birmingham. After completion of this study, all information will be kept for 10 years before being securely destroyed.

10.6 Amendments

If the sponsor wishes to make a substantial amendment to the REC application or the supporting documents, the sponsor will submit a valid notice of amendment to the REC for consideration. The REC will provide a response regarding the amendment within 35 days of receipt of the notice. It is the sponsor's responsibility to decide whether an amendment is substantial or non-substantial for the purposes of submission to the REC. Amendments will also be notified to NHS R&D departments of participating sites to assess whether the amendment affects the NHS permission for that site. The Chief Investigator is responsible for making any amendments to this protocol when agreed with the sponsor. Substantive changes will be communicated to all relevant stakeholders (REC, both R&D). Following an amendment, the version of the protocol will be updated, and the amendment recorded.

10.7 End of study

The end of the study will occur after data analysis is complete, the study has been written up for publication, the summary of findings has been disseminated to any funder, and the results have been presented to the participating physiotherapists and individuals with AIS. This is anticipated to take approximately one year.

10.8 Access to the final study dataset

Access to the final study dataset is currently only planned for the Chief and Lead Investigators. However, direct access will be granted to authorize representatives from the Sponsor, host institution, and the regulatory authorities to permit study-related monitoring, audits and inspections. It is not envisaged that the dataset will be used for secondary analysis.

11 DISSEMINIATION POLICY

11.1 Dissemination policy

- The Investigators will rapidly disseminate (oral presentation, social media) key findings to their clinical colleagues in the NHS
- The findings will be presented in an article that will be submitted to a high impact open access journal, accessible to UK and international professionals
- Findings will be presented at conferences nationally and internationally.

11.2 Authorship eligibility guidelines and any intended use of professional writers

Samia Alamrani, Nicola Heneghan, Alison Rushton, Deborah Falla , Adrian Gardner will all be authors in the final write up of the study for publication. They will meet the following recommendations for authorship as defined by the International Committee of Medical Journal Editors

The ICMJE criteria:

- Substantial contributions to the conception or design of the work; or the acquisition, analysis, or interpretation of data for the work; AND
- Drafting the work or revising it critically for important intellectual content; AND
- Final approval of the version to be published; AND
- Agreement to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

12 REFERENCES

1. Reamy, B.V. and J.B. Slakey, *Adolescent idiopathic scoliosis: review and current concepts.* Am Fam Physician, 2001. **64**(1): p. 111-6.
2. Konieczny, M.R., H. Senyurt, and R. Krauspe, *Epidemiology of adolescent idiopathic scoliosis.* Journal of children's orthopaedics, 2013. **7**(1): p. 3-9.
3. James, J.I., *Idiopathic scoliosis; the prognosis, diagnosis, and operative indications related to curve patterns and the age at onset.* J Bone Joint Surg Br, 1954. **36-b**(1): p. 36-49.
4. Makino, T., et al., *Low back pain and patient-reported QOL outcomes in patients with adolescent idiopathic scoliosis without corrective surgery.* SpringerPlus, 2015. **4**(1).
5. Durmala, J., W. Tomalak, and T. Kotwicki, *Function of the respiratory system in patients with idiopathic scoliosis: reasons for impairment and methods of evaluation.* Stud Health Technol Inform, 2008. **135**: p. 237-45.
6. Leszczewska, J., et al., *Evaluation of the stress level of children with idiopathic scoliosis in relation to the method of treatment and parameters of the deformity.* ScientificWorldJournal, 2012. **2012**: p. 538409.
7. Malmqvist, M., et al., *Patients With Idiopathic Scoliosis Run an Increased Risk of Schizophrenia.* Spine Deform, 2019. **7**(2): p. 262-266.
8. Du, C., et al., *Relevant areas of functioning in patients with adolescent idiopathic scoliosis on the International Classification of Functioning, Disability and Health: The patients' perspective.* J Rehabil Med, 2016. **48**(9): p. 806-814.
9. Tomey, K.M. and M.R. Sowers, *Assessment of Physical Functioning: A Conceptual Model Encompassing Environmental Factors and Individual Compensation Strategies.* Physical Therapy, 2009. **89**(7): p. 705-714.
10. Fan, H., et al., *Comparison of Functional Outcome and Quality of Life in Patients With Idiopathic Scoliosis Treated by Spinal Fusion.* Medicine, 2016. **95**(19): p. e3289-e3289.
11. Padua, R., et al., *Patient outcomes after Harrington instrumentation for idiopathic scoliosis: a 15- to 28-year evaluation.* Spine (Phila Pa 1976), 2001. **26**(11): p. 1268-73.
12. !!! INVALID CITATION !!! 9.
13. Asher, M., et al., *Scoliosis research society-22 patient questionnaire: responsiveness to change associated with surgical treatment.* Spine (Phila Pa 1976), 2003. **28**(1): p. 70-3.

14. Asher, M., et al., *The reliability and concurrent validity of the scoliosis research society-22 patient questionnaire for idiopathic scoliosis.* Spine (Phila Pa 1976), 2003. **28**(1): p. 63-9.
15. Asher, M.A., et al., *Refinement of the SRS-22 Health-Related Quality of Life questionnaire Function domain.* Spine (Phila Pa 1976), 2006. **31**(5): p. 593-7.
16. Williams, M.A., et al., *Active Treatment for Idiopathic Adolescent Scoliosis (ACTIvATeS): a feasibility study.* Health technology assessment (Winchester, England), 2015. **19**(55): p. 1-242.
17. Cobb, J.R., *The Problem of the Primary Curve.* 1960. **42**(8): p. 1413-1425.
18. Hamad, A., E.B. Ahmed, and A.I. Tsirikos, *Adolescent idiopathic scoliosis: a comprehensive approach to aetiology, diagnostic assessment and treatment.* Orthopaedics and Trauma, 2017. **31**(6): p. 343-349.
19. Horne, J.P., R. Flannery, and S. Usman, *Adolescent idiopathic scoliosis: diagnosis and management.* Am Fam Physician, 2014. **89**(3): p. 193-8.
20. Lonstein, J.E., *Adolescent idiopathic scoliosis.* The Lancet, 1994. **344**(8934): p. 1407-1412.
21. Schmid, S., et al., *Quantifying spinal gait kinematics using an enhanced optical motion capture approach in adolescent idiopathic scoliosis.* Gait & Posture, 2016. **44**: p. 231-237.
22. Dodd, S., et al., *A taxonomy has been developed for outcomes in medical research to help improve knowledge discovery.* J Clin Epidemiol, 2018. **96**: p. 84-92.
23. de Kleuver, M., et al., *Defining a core outcome set for adolescent and young adult patients with a spinal deformity.* Acta Orthopaedica, 2017. **88**(6): p. 612-618.
24. Kibsgård, T., J.I. Brox, and O. Reikerås, *Physical and mental health in young adults operated on for idiopathic scoliosis.* Journal of Orthopaedic Science, 2004. **9**(4): p. 360-363.
25. Lerman, J.A., E. Sullivan, and R.J. Haynes, *The Pediatric Outcomes Data Collection Instrument (PODCI) and functional assessment in patients with adolescent or juvenile idiopathic scoliosis and congenital scoliosis or kyphosis.* Spine (Phila Pa 1976), 2002. **27**(18): p. 2052-7; discussion 2057-8.
26. Seki, H., et al., *Postoperative pain management in patients undergoing posterior spinal fusion for adolescent idiopathic scoliosis: a narrative review.* Scoliosis and spinal disorders, 2018. **13**: p. 17-17.
27. Taylor, A.M., et al., *Assessment of physical function and participation in chronic pain clinical trials: IMMPACT/OMERACT recommendations.* Pain, 2016. **157**(9): p. 1836-50.
28. Bastrom, T.P., et al., *Prevalence of postoperative pain in adolescent idiopathic scoliosis and the association with preoperative pain.* Spine (Phila Pa 1976), 2013. **38**(21): p. 1848-52.
29. Baldus, C., et al., *The Scoliosis Research Society Health-Related Quality of Life (SRS-30) age-gender normative data: an analysis of 1346 adult subjects unaffected by scoliosis.* Spine (Phila Pa 1976), 2011. **36**(14): p. 1154-62.
30. Haher, T.R., et al., *Results of the Scoliosis Research Society instrument for evaluation of surgical outcome in adolescent idiopathic scoliosis. A multicenter study of 244 patients.* Spine (Phila Pa 1976), 1999. **24**(14): p. 1435-40.
31. Negrini, S., et al., *Why do we treat adolescent idiopathic scoliosis? What we want to obtain and to avoid for our patients. SOSORT 2005 Consensus paper.* Scoliosis, 2006. **1**(1): p. 4.
32. Asher, M.A., S. Min Lai, and D.C. Burton, *Further development and validation of the Scoliosis Research Society (SRS) outcomes instrument.* Spine (Phila Pa 1976), 2000. **25**(18): p. 2381-6.
33. Asher, M., et al., *Discrimination validity of the Scoliosis Research Society-22 Patient Questionnaire: relationship to idiopathic scoliosis curve pattern and curve size...including commentary by Keller RB.* Spine (03622436), 2003. **28**(1): p. 74-78.
34. Asher, M., et al., *Scoliosis Research Society-22 Patient Questionnaire: responsiveness to change associated with surgical treatment.* Spine (03622436), 2003. **28**(1): p. 70-73.
35. D'Andrea, L.P., et al., *Do Radiographic Parameters Correlate With Clinical Outcomes in Adolescent Idiopathic Scoliosis?* Spine, 2000. **25**(14): p. 1795-1802.
36. Connolly, P.J., et al., *Adolescent idiopathic scoliosis. Long-term effect of instrumentation extending to the lumbar spine.* J Bone Joint Surg Am, 1995. **77**(8): p. 1210-6.
37. Danielsson, A.J., K. Romberg, and A.L. Nachemson, *Spinal range of motion, muscle endurance, and back pain and function at least 20 years after fusion or brace treatment for adolescent idiopathic scoliosis: a case-control study.* Spine (Phila Pa 1976), 2006. **31**(3): p. 275-83.

38. Helenius, I., et al., *Harrington and Cotrel-Dubousset Instrumentation in Adolescent Idiopathic Scoliosis: Long-Term Functional and Radiographic Outcomes.* JBJS, 2003. **85**(12): p. 2303-2309.
39. Langensiepen, S., et al., *Measuring procedures to determine the Cobb angle in idiopathic scoliosis: a systematic review.* European spine journal : official publication of the European Spine Society, the European Spinal Deformity Society, and the European Section of the Cervical Spine Research Society, 2013. **22**(11): p. 2360-2371.
40. Slattery, C. and K. Verma, *Classifications in Brief: The Lenke Classification for Adolescent Idiopathic Scoliosis.* Clinical Orthopaedics and Related Research®, 2018. **476**(11): p. 2271-2276.

13 APPENDICIES

13.1 Required documentation

Please find required documentation attached to IRAS documents at point of submission

NHS
The Royal
Orthopaedic Hospital
NHS Foundation Trust
KNOWLEDGE | HUB

Our Ref: DP/ROH20ORTH01
DATE: 5th March 2020

Mr Adrian Gardner
The Royal Orthopaedic Hospital
Bristol Road South
B31 2AP

Dear Mr Gardner,

Research Project: FOAIS - Assessment of Functional Outcomes of Adolescents with Idiopathic Scoliosis

The above research project has been reviewed by the Trust through the Research & Development Department.

This letter confirms that The Royal Orthopaedic Hospital NHS Foundation Trust has the capacity and capability to deliver the above referenced study.

Version of Documents approved for the element of the research that will be conducted at this Trust:

Document	*Version*	*Date*
Protocol	V1.0	18.12.19

1. **Research Governance**: All research in the Trust must be conducted within the DoH Research Governance Framework (3rd ed. 2017).

2. **Recruitment of First Participant:** In line with NIHR objectives, you are responsible for ensuring that the first participant is recruited within 30 days of the date of this letter. Where this is not possible you must inform the R&D Department.

3. **Amendments:** You are required to submit all amendments to the project, including the changed documents, notification of amendment form and HRA approval letter to the R&D Department for approval. New versions of documentation can only be used once you have received notification from the R&D Department.

4. **Completion of Project:** Please inform the R&D Department when your project closes to recruitment and also when the project has finished in order for the files to be archived appropriately and our database to remain up-to-date.

RESPECT COMPASSION
EXCELLENCE PRIDE
OPENNESS INNOVATION

5. **Dissemination of Research Findings:** As with all NHS research, it is expected that the results of this study will be published in a reputable journal, may be presented at various meetings and will influence decisions on best practice. I would be very grateful if you could keep me informed of any publications, presentations and how the results of this research have been implemented into practice once the research has finished.

6. **Pharmacy Green Light**: Should your study require the dispensing of drugs, please do not commence work on the project until pharmacy has issued the green light, as per MHRA requirements. The green light confirms that pharmacy has all procedures and documentation in place and can comply with the medicines management aspects of the study. The pharmacy team will email you the green light approval once the above is in place.

On behalf of the Research & Development Department, I would like to wish you every success with your research project.

Best wishes

Yours sincerely

Prof. Phil Begg
Executive Director of Strategy & Delivery

cc. Samia Alamrani, University of Birmingham

STROBE Statement—Checklist of items that should be included in reports of ***cohort studies***

	Item No	Recommendation	Page No
Title and abstract	1	(*a*) Indicate the study's design with a commonly used term in the title or the abstract (*b*) Provide in the abstract an informative and balanced summary of what was done and what was found	34
Introduction			
Background/rationale	2	Explain the scientific background and rationale for the investigation being reported	37-38
Objectives	3	State specific objectives, including any prespecified hypotheses	39
Methods			
Study design	4	Present key elements of study design early in the paper	39
Setting	5	Describe the setting, locations, and relevant dates, including periods of recruitment, exposure, follow-up, and data collection	39
Participants	6	(*a*) Give the eligibility criteria, and the sources and methods of selection of participants. Describe methods of follow-up (*b*) For matched studies, give matching criteria and number of exposed and unexposed	39-40
Variables	7	Clearly define all outcomes, exposures, predictors, potential confounders, and effect modifiers. Give diagnostic criteria, if applicable	40
Data sources/ measurement	8*	For each variable of interest, give sources of data and details of methods of assessment (measurement). Describe comparability of assessment methods if there is more than one group	41
Bias	9	Describe any efforts to address potential sources of bias	42
Study size	10	Explain how the study size was arrived at	
Quantitative variables	11	Explain how quantitative variables were handled in the analyses. If applicable, describe which groupings were chosen and why	42
Statistical methods	12	(*a*) Describe all statistical methods, including those used to control for confounding (*b*) Describe any methods used to examine subgroups and interactions (*c*) Explain how missing data were addressed (*d*) If applicable, explain how loss to follow-up was addressed (*e*) Describe any sensitivity analyses	42-43
Results			
Participants	13*	(a) Report numbers of individuals at each stage of study—e.g. numbers potentially eligible, examined for eligibility, confirmed eligible, included in the study, completing follow-up, and analysed (b) Give reasons for non-participation at each stage (c) Consider use of a flow diagram	44
Descriptive data	14*	(a) Give characteristics of study participants (e.g. demographic, clinical, social) and information on exposures and potential confounders	45

		(b) Indicate number of participants with missing data for each variable of interest (c) Summarise follow-up time (e.g., average, and total amount)	
Outcome data	15*	Report numbers of outcome events or summary measures over time	46

Main results	16	(*a*) Give unadjusted estimates and, if applicable, confounder-adjusted estimates and their precision (e.g., 95% confidence interval). Make clear which confounders were adjusted for and why they were included (*b*) Report category boundaries when continuous variables were categorized (*c*) If relevant, consider translating estimates of relative risk into absolute risk for a meaningful time period	46-55
Other analyses	17	Report other analyses done—e.g. analyses of subgroups and interactions, and sensitivity analyses	NA
Discussion			
Key results	18	Summarise key results with reference to study objectives	56
Limitations	19	Discuss limitations of the study, taking into account sources of potential bias or imprecision. Discuss both direction and magnitude of any potential bias	60
Interpretation	20	Give a cautious overall interpretation of results considering objectives, limitations, multiplicity of analyses, results from similar studies, and other relevant evidence	56-59
Generalisability	21	Discuss the generalisability (external validity) of the study results	61
Other information			
Funding	22	Give the source of funding and the role of the funders for the present study and, if applicable, for the original study on which the present article is based	NA

Chapter Eight

Chapter Nine *Give information separately for exposed and unexposed groups.

Chapter Ten

Chapter Eleven **Note:** An Explanation and Elaboration article discusses each checklist item and gives methodological background and published examples of transparent reporting. The STROBE checklist is best used in conjunction with this article (freely available on the Web sites of PLoS Medicine at http://www.plosmedicine.org/, Annals of Internal Medicine at http://www.annals.org/, and Epidemiology at http://www.epidem.com/). Information on the STROBE Initiative is available at http://www.strobe-statement.org.

Chapter Twelve

Appendix 7. Terms and keywords used in search one and search two, Risk of bias results of measurement properties studies , Interpretability and feasibility results, AMSTAR checklist for *Chapter Three*

Terms and key words used in Search one- (MEDLINE database)

1. scoliosis.mp.
2. exp Scoliosis/
3. exp Spinal Curvatures/
4. Adolescen$.mp.
5. exp Adolescent/
6. PF.mp.
7. exp Physical Functional Performance/
8. Functional activity.mp.
9. independence.mp.
10. Functional independence.mp.
11. exp Health Status/
12. exp performance status/
13. exp Health Behaviour/ behaviour
14. exp Movement/
15. mobility.mp.
16. Functional limitation.mp.
17. Activity limitation.mp.
18. exp Motor Activity/
19. Recovery of function/
20. (Recover$ adj5 function$).tw.
21. exp Motor Skills/
22. exp Disability Evaluation/
23. exp Disabled Persons/
24. exp physical examination/
25. exp "Activities of Daily Living"/
26. ((daily or domestic or house or home) adj5 (activit$ or task$ or skill$ or chore$)).t
27. (Activities of daily living or adl$ or eadl$ or iadl$).tw.
28. exp Self Care/
29. ((self or personal) adj5 (Care or manage$)).tw.
30. (Dressing or feeding or eating or toilet$ or bathing or mobil$ or driving or public
31. transport$).tw.
32. exp Lifting/
33. Bending.mp.
34. exp sitting/
35. exp Walking/
36. exp Walking Speed/
37. exp Postural Balance/
38. Standing balance.mp.
39. exp Hand Strength/ or Grip strength.mp.
40. 1-3/OR
41. 4 OR 5
42. 6-39/OR
43. 40 and 41 and 42
44. Limit 43 to humans

Search two : Measurement properties PROM (Generic)

(Ovid MEDLINE)
1. SF -36.mp.
2. short form 36.mp.
3. SF-12.mp.
4. Oswestry disability index.mp.
5. ODI.mp.
6. RMDQ.mp.
7. Roland-Morris Disability Questionnaire.mp.
8. PROMIS.mp.
9. Patient-Reported OMment Information System.mp.
10. Competence Scale of the Youth Self-Report and Profile.mp.
11. Sports Activity Questionnaire.mp.
12. Paediatric Outcomes Data Collection Instrument.mp.
13. PODI.mp.
14. Paediatric Quality of Life Inventory.mp.
15. PedsQLTM.mp.
16. Functional Rating Index.mp.
17. FRI.mp.
18. Hannover Functional Ability Questionnaire.mp.
19. HFAQ.mp.
20. Patient Specific Functional Scale.mp.
21. Paediatric Evaluation of Disability Inventory.mp.
22. PEDI.mp.
23. Brief Pain Inventory.mp.
24. BPI.mp.
25. Activity scale for kids.mp.
26. EuroQoL 5-dimension 5-level.mp.
27. EQ-5D-5L.mp.
28. Child Health Questionnaire CF87.mp.
29. Child Health Questionnaire.mp.
30. 1-29/OR
31. validity.mp.
32. exp validation studies/
33. reliability.mp.
34. exp reproducibility of results/
35. interpretability.mp.
36. internal consistency.mp.
37. sensitivity/ and Specificity.mp.
38. clinical sensitivity.mp.
39. exp psychometrics/
40. responsiveness.mp.
41. exp Evaluation studies/
42. measurement error.mp.
43. measurement properties.mp.
44. 31-43/OR
45. 30 and 44
46. Adolescen$.mp.
47. exp adolescent/
48. 46 or 47
49. scolio*.ti,ab.
50. exp Scoliosis/ or scoliois.mp
51. 49 or 50
53. 30 and 44 and 48 and 51

Search two : Measurement properties PBOM and body structure and function OM (Ovid MEDLINE)
1. validity.mp.
2. exp validation studies/
3. reliability.mp.
4. exp reproducibility of results/
5. interpretability.mp.
6. internal consistency.mp.
7. exp sensitivity/ or exp Specificity/
8. clinical sensitivity.mp.
9. exp psychometrics/
10. responsiveness.mp.
11. exp Evaluation studies/
12. measurement error.mp.
13. measurement properties.mp.
14. 1-13/OR
15. exp Scoliosis/ or Scolioisis.mp. or scolio*.ti,ab.
16. adolescen$.mp. or Exp adolescent/
17. Cobb angle.mp.
18. 6 MWT.mp.
19. Six Minutes' Walk Test.mp.
20. Shuttle Walk Test.mp.
21. incremental shuttle walk test.mp. or Walk Test/
22. (Time Up and Go test).mp.
23. "Time Up and Go test".kw.
24. ISWT.mp.
25. TUG.mp.
26. (Sit and reach test).mp.
27. Fingertip-To-Floor.mp.
28. Fingertip-To-Floor Distance.mp.
29. Dega wall test.mp.
30. Thomas test.mp.
31. Lasegue test.mp.
32. Lasegue sign.mp.
33. Suspension test.mp.
34. Romberg Test.mp.
35. tandem Romberg test.mp.
36. Limit Of Stability Test.mp.
37. Sensory Organization Test.mp.
38. Hop test.mp.
39. single leg Hop test.mp.
40. Y-Balance Test.mp.
41. Fukuda Utenberger stepping test.mp.
42. Sharpened Romberg test.mp.
43. Unipedal stance test.mp.
44. Spinal ROM.mp.
45. Spinal Range of motion.mp.
46. Spine Range Of Motion.mp.
47. Trunk-Pelvis-Hip Angle test.mp.
48. Trunk Pelvis Hip Angle test.mp.
49. Modified Schober's Test.mp.
50. Sorensen Test.mp.

51. Kraus-Weber Test.mp.
52. Lumbar trunk muscle endurance test.mp.
53. Lumbar trunk endurance test.mp.
54. non-dynamometric trunk performance tests.mp.
55. Hand Strength/ or Hand grip strength test.mp.
56. Shoulder Range of Motion.mp.
57. Temporomandibular Range of Motion.mp.
58. Short Physical Performance Battery.mp.
59. Berg Balance Scale.mp.
60. 17-59/OR
61. 14 and 15 and 16 and 60
62. limit 61 to humans

Terms and key words used in Search Two- (MEDLINE database)

Measurement properties PROM (Disease specific)
(Ovid MEDLINE) 1. validity.mp. 2. exp validation studies/ 3. reliability.mp. 4. exp reproducibility of results/ 5. interpretability.mp. 6. internal consistency.mp. 7. clinical sensitivity.mp. 8. exp psychometrics/ 9. responsiveness.mp. 10. exp Evaluation studies/ 11. measurement error.mp. 12. measurement properties.mp. 13. 1 to 12/or 14. SRS-7.mp. 15. SRS-22.mp. 16. SRS-23.mp. 17. SRS-22r.mp. 18. SRS-24.mp. 19. SRS-30.mp. 20. Scoliosis Research Society Questionnaire.mp. 21. Quality of Life Profile for Spinal Deformities.mp. 22. Quality of Life for Spinal Deformities Scale.mp. 23. QLPSD.mp. 24. Scoliosis Spine Score Questionnaire.mp. 25. Brace Questionnaire.mp. 26. 14 -25 / OR 27. Adolescen$.mp. 28. Adolescent 29. 27 or 28 30. scoliosis.mp. 31. exp Scoliosis/ 32 scolio*.ti,ab. 33. 30 or 31 or 32 34. 13 and 26 and 29 and 33

COSMIN criteria and rating system for evaluating the content validity

	SQLI development (Feise et al., 2005)	**Rating of reviewers**	**Overall ratings**	**Quality of evidence**
Relevance				
1. Are the included items relevant for the construct of interest?	+	+	/	**Very low**
2. Are the included items relevant for the target population of interest?	-	+	/	
3. Are the included items relevant for the context of use of interest?	+	+	/	
4. Are the response options appropriate?	+	+	/	
5. Is the recall period appropriate?	+	+	/	
RELEVANCE RATING	±	+	+	**Very low**
Comprehensiveness				
6. Are all key concepts included?	_	+	/	
COMPREHENSIVENESS RATING	_	+	+	**Very low**
Comprehensibility				
7. Are the PROM instructions understood by the population of interest as intended?	_	/	/	
8. Are the PROM items and response options understood by the population of interest as intended?	_	/	/	
9. Are the PROM items appropriately worded?	/	+	/	
10. Do the response options match the question?	/	+	/	
COMPREHENSIBILITY RATING	-	+	+	**Very low**
CONTENT VALIDITY RATING	±	+	+	**Very low**

SQLI indicates Scoliosis Quality of Life Index; Ratings: sufficient (+), insufficient (-), inconsistent (±), indeterminate (?), not applicable (NA), (/) empty cells.

Results of studies on measurement properties

Name of OM	**Measurement error**			**Criterion validity**			**Hypotheses testing**			**Responsiveness**		
	Study n	Meth qual	Result (rating)	Study n	Meth qual	Result (rating)	Study n	Meth qual	Result (rating)	Study n	Meth qual	Result (rating)
PODCI (Lerman et al., 2002)							102	Adequate	Result in line with 2 hypo's (2+) Result not in line with Hypo's (3-) Overall (-)			
SQLI (Feise et al., 2005)							70	Very good	Result in line with 2 hypo's (2+) Overall (+)			
PROMIS (Fedorak et al., 2019)							113	Very good	Result in line with 1 hypo's (1+) overall (+)			
SRS 30 (Lubicky et al., 2011)							356	Very good	Result in line with 1 hypo's (1+) overall (+)			
SRS-22, SRS-24 (Bastrom et al., 2015)							829	Very good	Result in line with 2 hypo's (2+) Overall (+)			

Meth qual indicates Methodology quality; n: number; PODCI, Paediatrics Outcomes Data Collection Instrument; PROMSIS, Patient-Reported Outcomes Measurement Information System; SRS, Scoliosis Research Society; SRS-22r, Scoliosis Research Society-22Revised; SQLI, Scoliosis Quality of Life Index; Ratings: + = Sufficient ; ? = Indeterminate; − = Insufficient.

Results of studies on measurement properties

Name of OM	Measurement error			Criterion validity			Hypotheses testing			Responsiveness		
	Study n	Meth qual	Result (rating)	Study n	Meth qual	Result (rating)	Study n	Meth qual	Result (rating)	Study n	Meth qual	Result (rating)
SRS-22 (Asher et al., 2003b)										58	Doubtful	Result in line with 4 hypo's (+4)
SRS-22 (Asher et al., 2003a)							119	Very good	Result not in line with 3 hypo's (-)			
SRS-22,24 (Bastrom et al., 2015)							829	Very good	Result in line with 1 hypo's (1+) result not in line with 2 hypo's (2-)			
SRS-22 (Parent et al., 2009)							153	Very good	Result in line with 1 hypo's (1+) result not in line with 1 hypo's (2-)			
SRS-22 (Carreon et al., 2010)	887	Adequate	SDC(0.41)> MIC(.08) (-)									
Pooled or summary result (overall rating)							1101		(7-) and (2+) overall (-)			

Meth qual indicates Methodology quality; MIC, Minimal Important Change; n, number ; SDC, Small Detectable Change; SRS, Scoliosis Research Society; SRS-22r, Scoliosis Research Society-22Revised; Ratings: + = Sufficient; ? = Indeterminate; − = Insufficient

Results of studies on measurement properties

Name of OM	Measurement error			Criterion validity			Hypotheses testing			Responsiveness		
	Study n	Meth qual	Result (rating)	Study n	Meth qual	Result (rating)	Study n	Meth qual	Result (rating)	Study n	Meth qual	Result (rating)
SRS-22r (Glattes et al., 2007)							70	Adequate	Pearson r=0.73 with CHQ-CF87 Result in line with 1 hypo's (2+)			
SRS-22r (Fedorak et al., 2019)							113	Very good	Pearson r=0.65 with PROMIS Result not in line with 1 hypo's(1-) Result in line with 1 hypo's (2+)			
SRS-22r (Berliner et al., 2013)							155	Very good	Result not in line with 2 hypo's (2-)			
SRS-22r (Kelly et al., 2019)	1281	Adequate	SDC (0.24) >MIC(0.08) (-)							1281	Doubtful	Result not in line with 1 hypo's (-) Overall (-)
Pooled or summary result (overall rating)							338		(2-) (4+) Overall (+)			

CHQ-CF87 indicates Child Health Questionnaire- Child Self-Report Form 87; Meth qual, Methodology quality; MIC, Minimal Important Change; n, number ; SDC, Small Detectable Change; SRS, Scoliosis Research Society; SRS-22r, Scoliosis Research Society-22Revised, Ratings: + = Sufficient,? = Indeterminate, − = Insufficient

Results of studies on measurement properties

Name of OM	Structural validity			Internal consistency			Reliability			Cross-cultural validity\ measurement invariance		
	Study n	Meth qual	Result (rating)	Study n	Meth qual	Result (rating)	Study n	Meth qual	Result (rating)	Study n	Meth qual	Result (rating)
CHQ-CF87 (Glattes et al., 2007)				70	Very good	∝=0.89 (?)	54	Adequate	ICC=0.73 (0.20– 0.85) (+)			
SQLI (Feise et al., 2005)				84	Very good	∝ = 0.82 (?)	18	Adequate	ICC= 0.46 (0.29 –0.63) (-)			
SAQ (Sarwahi et al., 2018)							75	Doubtful	Kappa k ≥ 0.70 (+)			
SRS-30 (Roberts et al., 2011)										744	Adequate	No multiple group factor analysis OR DIF analysis performed (?)
SRS-22r (Glattes et al., 2007)				70	Very good	∝ = 0.82 (?)	54	Adequate	ICC=0.76 (0.56– 0.80) (+)			
SRS-22 (Verma et al., 2014)										80	Inadequate	No multiple group factor analysis OR DIF analysis performed (?)
TPHA Test (Stępień et al., 2018)							49	Very good	ICC= 0.85 (0.95-0.98) (+)			

CHQ-CF87 indicates Child Health Questionnaire- Child Self-Report Form 87; ICC, Interclass Correlation Coefficient; Meth qual, Methodology quality; n, number; SAQ, Sport Activity Questionnaire; SRS, Scoliosis Research Society; SRS-22r, Scoliosis Research Society-22Revised; SQLI, Scoliosis Quality of Life Index; TPHA, Trunk Pelvis Hip Angle test; ∝=Cronbach alpha; Ratings: + = Sufficient; ? = Indeterminate; − = Insufficient.

Results of studies on measurement properties

Name of OM	Measurement error			Criterion validity			Hypotheses testing			Responsiveness		
	Study n	Meth qual	Result (rating)	Study n	Meth qual	Result (rating)	Study n	Meth qual	Result (rating)	Study n	Meth qual	Result (rating)
TUG (Gao et al., 2019)							30	Adequate	Result not in line with 1 hypo's(1-) Result in line with 2 hypo's (2+) overall (+)			
FTF (Hresko et al., 2006)							58	Very good	Result not in line with 1hypo's overall (-)			
C7-PSIS, MST (Hresko et al., 2006) (Eyvazov et al., 2017)				37	Doubtful	(?) Not all information for '+' reported	58	Very good	Result not in line with 2 hypo's overall (-)			
Axial rotation, LSB (Eyvazov et al., 2017)							58	Very good	Result in line with 2 hypo's (2+) overall (+)			

C7-PSIS indicates Cervical 7 to Posterior Superior Iliac Spine ; FTF, Fingertip To Floor Test; LSB, Lateral Side Bending; Meth qual, Methodology quality; MST, Modified Schober Test; n, number; TPHA, Trunk Pelvis Hip Angle test; TUG, Timed Up and Go test; Ratings: + = Sufficient,? = Indeterminate, − insufficient

Interpretability of Patient Reported Outcome Measures

PROM (Ref)	Subscale	Distribution of scores in study population	% Missing items & % Missing total scores	Floor and Ceiling effects (%)	Score and change scores available for relevant (sub) groups	MIC or MID	Information on response shift
SRS 30 **(Lubicky et al., 2011),(Roberts et al., 2011)**	Function/Activity (5) post-surgery questions (2)	NR	NR	NR	NR	NR	NR
SRS 24 **(Bastrom et al., 2015)**	Function /activity	Ceiling	NR	Ceiling pre surgery (39%) Ceiling post-surgery (46%)	NR	NR	NR
SRS 22 **(Bastrom et al., 2015)**	Function /activity	Ceiling	NR	Ceiling (34%)	NR		NR
SRS22**(Carreon et al., 2010)**	Function /activity	Ceiling	NR	NR	NR	MIC (0.08) MDC (0.41)	NR
SRS22r **(Kelly et al., 2019)**	Function /activity	Ceiling	NR		NR	MDMD ((0.24	NR
SRS22r **(Fedorak et al., 2019)**	Function /activity	Ceiling	NR	Ceiling (27.4%) Floor (1.8%)	NR	NR	NR
SRS22r **(Glattes et al., 2007)**	Function	No floor effects, Ceiling effect	None	Ceiling (47.1%) Floor (1.4%)	NR	NR	NR
SQLI **(Feise et al., 2005)**	Physical activity	Ceiling	None	Floor (0 %) Ceiling (2.9%)	NR	NR	NR
SQLI**(Parent et al., 2007)**	Physical activity	No floor effects, Ceiling (2, 3, 5)	None	Ceiling (1.1%)	>50% scored over 4 out 5 on all domains	NR	NR
CHQ-CF87**(Glattes et al., 2007)**	Physical function	No floor effects, Ceiling effect	None	Floor (1.4) Ceiling (44.8%)	NR	NR	NR
SAQ **(Sarwahi et al., 2018)**	School Gym Carry backpack Bend over Running	NR	NR	NR	NR	NR	NR
PODCI **(Lerman et al., 2002)**	Upper Extremity Functioning , Transfers& basic Mobility Sport & Physical Function Global function	NR	NR	NR	NR	NR	NR
PROMIS **(Fedorak et al., 2019)**	Mobility	Floor and ceiling effects	NR	Ceiling 20.4% Floor 0.9%	NR	NR	NR

CHQ-CF87 indicates Child Health Questionnaire- Child Self-Report Form 87; NR, Not Reported; MDC, Minimal Detectable Change; MDMD, Minimal Detectable Minimal Difference; MIC, Minimal important Change; NR,Not Reported; PODCI, Paediatrics Outcomes Data Collection Instrument; PROMIS, Patient-Reported Outcomes Measurement Information System; SAQ, Sport Activity Questionnaire; SRS, Scoliosis Research Society; SRS-22r, Scoliosis Research Society-22Revised; SQLI, Scoliosis Quality of Life Index

Feasibility of Patient Reported Outcome Measures

Feasibility aspects	SRS-24	SRS-22	SRS-22r	SRS30	SQLI	SAQ	CHQ-CF87	PODCI	PROMIS
Patient's comprehensibility	NR	NR	NR	NR	Tested among female school children (9-11) Year	NR	NR	NR	NR
Clinician's comprehensibility	NR	NR	NR	NR	NR	NR	NR	NR	NR
Type and ease of administration	Self-administration	Self-administration-easy	Self-administration-easy	NR	Self-administration-easy	NR	Self-administration-	NR	NR
Length of the instrument Completion time	3 Min	3 Min	2-3 Min	NR	2.5±1.83 Min.	NR	NR	NR	NR
Patient's required mental and physical ability level	Minimum	Minimum	Minimum	NR	Minimum	Minimum	Minimum	NR	NR
Ease of standardization	Easy	Easy	Easy	NR	Easy	NR		NR	NR
Ease of score calculation	Easy	Easy	Easy	NR	Easy	NR	Moderate	NR	NR
Copyright				NR				NR	NR
Cost of an instrument	Free	Free	Free	Free	Free	Free	Paid	NR	NR
Required equipment	Pencil & Paper	Pencil & Paper	Pencil & Paper	NR	Pencil c& paper/ computerized	Phone, electronically	Pencil & Paper	Pencil &paper	Electronic
Availability in different settings	NR	NR	NR	NR	NR	NR	Available	Available	Available
Regulatory agency's requirement for approval	NR	NR	NR	NR	NR	NR	NR	NR	NR

CHQ-CF87 indicates Child Health Questionnaire- Child Self-Report Form 87; PODCI, Pediatric Outcomes Outcome Data Collection; PROMIS, Patient Reported Outcomes Information System; SAQ, Sport Activity Questionnaire; SRS, Scoliosis Research Society; SRS-22r, Scoliosis Research Society-22Revised; SQLI, Scoliosis Quality of Life Index; Min, Minutes :NR, Not Reported.

AMSTAR 2 Results

Printer Friendly Version

Article Name: PHYSICAL FUNCTIONING IN ADOLESCENT WITH IDIOPATHIC SCOLIOSIS

You are currently logged on as Guest. You need to be logged on as a member to submit your score.
Log On

PHYSICAL FUNCTIONING IN ADOLESCENT WITH IDIOPATHIC SCOLIOSIS is a Moderate quality review

1. Did the research questions and inclusion criteria for the review include the components of PICO?	Yes Yes Yes Yes Yes
2. Did the report of the review contain an explicit statement that the review methods were established prior to the conduct of the review and did the report justify any significant deviations from the protocol?	YesYesYesYesYesYes
3. Did the review authors explain their selection of the study designs for inclusion in the review?	Yes Yes
4. Did the review authors use a comprehensive literature search strategy?	Yes Yes Yes Yes Yes Yes Yes Yes Yes
5. Did the review authors perform study selection in duplicate?	Yes Yes
6. Did the review authors perform data extraction in duplicate?	Yes Yes
7. Did the review authors provide a list of excluded studies and justify the exclusions?	Partial Yes Yes
8. Did the review authors describe the included studies in adequate detail?	Yes Yes Yes Yes Yes Yes Yes Yes Yes Yes Yes
9. Did the review authors use a satisfactory technique for assessing the risk of bias (RoB) in individual studies that were included in the review? **RCT**	Yes
NRSI	Yes
10. Did the review authors report on the sources of funding for the studies included in the review?	No
11. If meta-analysis was performed did the review authors use appropriate methods for statistical combination of results? **RCT**	0
NRSI	0
12. If meta-analysis was performed, did the review authors assess the potential impact of RoB in individual studies on the results of the meta-analysis or other evidence synthesis?	0
13. Did the review authors account for RoB in individual studies when interpreting/ discussing the results of the review?	Yes
14. Did the review authors provide a satisfactory explanation for, and discussion of, any heterogeneity observed in the results of the review?	Yes Yes
15. If they performed quantitative synthesis did the review authors carry out an adequate investigation of publication bias (small study bias) and discuss its likely impact on the results of the review?	0
16. Did the review authors report any potential sources of conflict of interest, including any funding they received for conducting the review?	Yes Yes

To cite this tool: Shea BJ, Reeves BC, Wells G, Thuku M, Hamel C, Moran J, Moher D, Tugwell P, Welch V, Kristjansson E, Henry DA. AMSTAR 2: a critical appraisal tool for systematic reviews that include randomised or non-randomised studies of healthcare interventions, or both. BMJ. 2017 Sep 21;358:j4008.

Appendix 8. Scoliosis Research Society -22 revised (SRS-22r) patient questionnaire

SRS-22r Patient Questionnaire (adapted from the SRS.org)

INSTRUCTIONS: We are carefully evaluating the condition of your back and it is **IMPORTANT THAT YOU ANSWER EACH OF THESE QUESTIONS YOURSELF.** Please **CIRCLE THE ONE BEST ANSWER TO EACH QUESTION.**

1. Which one of the following best describes the amount of pain you have experienced during the past 6 months?

    - None
    - Mild
    - Moderate
    - Moderate to severe
    - Severe

2. Which one of the following best describes the amount of pain you have experienced over the last month?

    - None
    - Mild
    - Moderate
    - Moderate to severe
    - Severe

3. During the past 6 months have you been a very nervous person?

    - None of the time
    - A little of the time
    - Some of the time
    - Most of the time
    - All of the time

4. If you had to spend the rest of your life with your back shape as it is right now, how would you feel about it?
    - Very happy
    - Somewhat happy
    - Neither happy nor unhappy Somewhat unhappy
    - Very unhappy

5. What is your current level of activity?
    - Bedridden
    - Primarily no activity
    - Light labor and light sports
    - Moderate labor and moderate sports
    - Full activities without restriction

6. How do you look in clothes?
    - Verygood
    - Good
    - Fair
    - Bad
    - Verybad

7. In the past 6 months have you felt so down in the dumps that nothing could cheer you up?
    - Very often
    - Often
    - Sometimes
    - Rarely
    - Never
8. Do you experience back pain when at rest?
    - Very often
    - Often
    - Sometimes
    - Rarely
    - Never
9. What is your current level of work/school activity?
    - 100% normal
    - 75% normal
    - 50% normal
    - 25% normal
    - 0% normal

10. Which of the following best describes the appearance of your trunk; defined as the human body except for the head and extremities?

    - Very good
    - Good
    - Fair
    - Poor
    - Very Poor

11. Which one of the following best describes your pain medication use for back pain?

    - None
    - Non-narcotics weekly or less (e.g., aspirin, Tylenol, Ibuprofen)
    - Non-narcotics daily
    - Narcotics weekly or less (e.g. Tylenol III, Lorcet, Percocet)
    - Narcotics daily

12. Does your back limit your ability to do things around the house?

- Never
- Rarely
- Sometimes
- Often
- Very Often

13. Have you felt calm and peaceful during the past 6 months?

- All of the time
- Most of the time
- Some of the time
- A little of the time
- None of the time

14. Do you feel that your back condition affects your personal relationships?

- None
- Slightly
- Mildly
- Moderately
- Severely

15. Are you and/or your family experiencing financial difficulties because of your back?

- Severely
- Moderately
- Mildly
- Slightly
- None

16. In the past 6 months have you felt down hearted and blue?

- Never
- Rarely
- Sometimes
- Often
- Very often

17. In the last 3 months have you taken any days off of work, including household work, or school because of back pain?

- 0 days
- 1 day
- 2 days

- 3 days
- 4 or more days

18. Does your back condition limit your going out with friends/family?

- Never
- Rarely
- Sometimes
- Often
- Very often

19. Do you feel attractive with your current back condition?

- Yes, very
- Yes, somewhat
- Neither attractive nor unattractive
- No, not very much
- No, not at all

20. Have you been a happy person during the past 6 months?

- None of the time
- A little of the time
- Some of the time
- Most of the time
- All of the time

21. Are you satisfied with the results of your back management?

- Very satisfied
- Satisfied
- Neither satisfied nor unsatisfied
- Unsatisfied
- Very unsatisfied

22. Would you have the same management again if you had the same condition?

- Definitely yes
- Probably yes
- Not sure
- Probably not
- Definitely not

Appendix 9. Ethical Review, Patient Information Sheets, Assent/Consent Forms, Consent to contact form , Topic guide, Topic guide after amendment, Saturation table, Themes, and subthemes with quotations and COREQ checklist for *Chapter Four*, and *Chapter Five*

Mr Adrian Gardner
Consultant Spine Surgeon
The Royal Orthopaedic Hospital NHS Foundation Trust
Bristol Road South
Northfield
Brimingham
B31 2APN/A

Email: approvals@hra.nhs.uk
HCRW.approvals@wales.nhs.uk

28 May 2021

Dear Mr Gardner,

HRA and Health and Care Research Wales (HCRW) Approval Letter

Study title:	**Content validity of the Scoliosis Research Society questionnaire-22r (SRS-22r) for Adolescents with Idiopathic Scoliosis : a qualitative interview study exploring patient's and practitioner's perspectives.**
IRAS project ID:	**289888**
Protocol number:	**V5**
REC reference:	**21/WM/0076**
Sponsor	**The Royal Orthopaedic Hospital NHS Foundation Trust**

I am pleased to confirm that **HRA and Health and Care Research Wales (HCRW) Approval** has been given for the above referenced study, on the basis described in the application form, protocol, supporting documentation and any clarifications received. You should not expect to receive anything further relating to this application.

Please now work with participating NHS organisations to confirm capacity and capability, in line with the instructions provided in the "Information to support study set up" section towards the end of this letter.

How should I work with participating NHS/HSC organisations in Northern Ireland and Scotland?
HRA and HCRW Approval does not apply to NHS/HSC organisations within Northern Ireland and Scotland.

If you indicated in your IRAS form that you do have participating organisations in either of these devolved administrations, the final document set and the study wide governance report

(including this letter) have been sent to the coordinating centre of each participating nation. The relevant national coordinating function/s will contact you as appropriate.

Please see IRAS Help for information on working with NHS/HSC organisations in Northern Ireland and Scotland.

How should I work with participating non-NHS organisations?
HRA and HCRW Approval does not apply to non-NHS organisations. You should work with your non-NHS organisations to obtain local agreement in accordance with their procedures.

What are my notification responsibilities during the study?

The standard conditions document "*After Ethical Review – guidance for sponsors and investigators*", issued with your REC favourable opinion, gives detailed guidance on reporting expectations for studies, including:

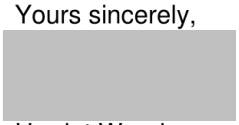
- Registration of research
- Notifying amendments
- Notifying the end of the study

The HRA website also provides guidance on these topics, and is updated in the light of changes in reporting expectations or procedures.

Who should I contact for further information?
Please do not hesitate to contact me for assistance with this application. My contact details are below.

Your IRAS project ID is **289888**. Please quote this on all correspondence.

Yours sincerely,



Harriet Wood
Approvals Specialist
Email: approvals@hra.nhs.uk

Copy to: *Ms Carolyn Langford*

List of Documents

The final document set assessed and approved by HRA and HCRW Approval is listed below.

Document	*Version*	*Date*
Interview schedules or topic guides for participants [Patient topic guide - tracked]	4.0	21 April 2021
Interview schedules or topic guides for participants [HCP topic guide - tracked]	2.0	21 April 2021
Interview schedules or topic guides for participants [Patient topic guide]	4	21 April 2021
Interview schedules or topic guides for participants [HCP topic guide]	2.0	21 April 2021
IRAS Application Form [IRAS_Form_22022021]		22 February 2021
Letter from sponsor [n/a]		12 February 2021
Other [Ethics Responses]	1.0	21 April 2021
Other [Samia Alamrani DBS cerificate]	1.0	17 July 2019
Other [Samia Alamrani Safeguarding children training certificate]	1.0	10 April 2021
Other [Ethics Response 19.05.2021]		
Participant consent form [Assent 10-15]	5.0	19 May 2021
Participant consent form [consent 16-18]	4.0	19 May 2021
Participant consent form [consent Parent]	5.0	19 May 2021
Participant consent form [consent HCP]		
Participant consent form [Assent 10-15 TC]	5	19 May 2021
Participant consent form [consent 16-18 TC]	4	19 May 2021
Participant consent form [consent Parent TC]	5	19 May 2021
Participant consent form [consent HCP TC]	3	19 May 2021
Participant information sheet (PIS) [PIS 10-12]	3.0	19 May 2021
Participant information sheet (PIS) [PIS 13-15]	3.0	19 May 2021
Participant information sheet (PIS) [PIS 16-18]	4.0	19 May 2021
Participant information sheet (PIS) [PIS HCPs]	4.0	19 May 2021
Participant information sheet (PIS) [PIS Parent]	4.0	19 May 2021
Participant information sheet (PIS) [PIS 10-12 TC]	3	19 May 2021
Participant information sheet (PIS) [PIS 13-15 TC]	3	19 May 2021
Participant information sheet (PIS) [PIS 16-18 TC]	4	19 May 2021
Participant information sheet (PIS) [PIS HCPs TC]	4	19 May 2021
Participant information sheet (PIS) [PIS Parent TC]	4	19 May 2021
Research protocol or project proposal [protocol]	6.0	19 May 2021
Research protocol or project proposal [protocol TC]	6	19 May 2021
Summary CV for Chief Investigator (CI) [CI CV]		25 November 2019
Summary CV for student [student CV]		20 January 2020
Summary CV for supervisor (student research) [supervisor CV]		21 January 2020

IRAS project ID	289888

Information to support study set up

The below provides all parties with information to support the arranging and confirming of capacity and capability with participating NHS organisations in England and Wales. This is intended to be an accurate reflection of the study at the time of issue of this letter.

Types of participating NHS organisation	Expectations related to confirmation of capacity and capability	Agreement to be used	Funding arrangements	Oversight expectations	HR Good Practice Resource Pack expectations
This is a single site study sponsored by the participating NHS organization therefore there is only one site type.	This is a single site study sponsored by the participating NHS organisation. You should work with your sponsor R&D office to make arrangements to set up the study. The sponsor R&D office will confirm to you when the study can start following issue of HRA and HCRW Approval.	As this is a single site study sponsored by the participating NHS organisation, no Organisation Information Document or Schedule of Events is required or expected. You should work with your sponsor R&D office to make arrangements to set up the study. The sponsor R&D office will confirm to you when the study can start following issue of HRA and HCRW Approval.	No external study funding has been sought.	A Principal Investigator should be appointed at study sites.	Where arrangements are not already in place, research staff not employed by the NHS host organisation undertaking any of the research activities listed in the research application would be expected to obtain a Letter of Access based on enhanced DBS checks and occupational health clearance.

Other information to aid study set-up and delivery

This details any other information that may be helpful to sponsors and participating NHS organisations in England and Wales in study set-up.
The applicant has indicated that they intend to apply for inclusion on the NIHR CRN Portfolio.

NHS
The Royal
Orthopaedic Hospital
NHS Foundation Trust

Our ref: EL/AG/ROH21ORTH04

09 February 2022

Professor Adrian Gardner
The Royal Orthopaedic Hospital NHS Foundation Trust
Bristol Road South
Birmingham
B31 2AP

Dear Adrian,

Research Project: Content validity of the Scoliosis Research Society questionnaire-22r (SRS-22r) for Adolescents with Idiopathic Scoliosis: a qualitative interview study exploring patient's and practitioner's perspectives.

R&D Project No: ROH21ORTH04

The above research project has been re-reviewed by the Trust through the Research & Development Department in light of the Non-Substantial Amendment 04 dated 08 February 2022

New Version of Documents approved:

Document	Version	Date
N/A		

Summary of Amendment (not exhaustive):

- To increase the recruitment rate, we propose to introduce remote consent to contact. The PI who is also part of the clinical care team will call up the patient and introduce the study. PIS and ICF will be sent to all participants

The original Trust approval letter has also been reviewed and the R&D Department can confirm that the above non-substantial amendment has been approved and should be read in conjunction with terms and conditions of the original Trust approval letter.

On behalf of the Research & Development Department, I would like to wish you every continued success with your research project.

Best wishes

Yours sincerely

Professor Philip Begg,
Executive Director of Strategy and Delivery
Royal Orthopaedic Hospital

cc. Samia Alamrani, UoB

UNIVERSITY OF BIRMINGHAM

Participant Information Sheet (Parents/Carer)

Study title: Content validity of the Scoliosis Research Society questionnaire-22r (SRS-22r) for Adolescents with Idiopathic Scoliosis

Invitation paragraph

We would like to invite your child to take part in our research study. This research is being conducted by the University of Birmingham and sponsored by the Royal Orthopaedic Hospital NHS Foundation Trust, who are the data controller for the project.

Before you decide, we would like you to understand why the research is being done and what it will involve.

One of our team will go through this information with you and answer any questions you may have. You can talk to others about the study if you wish.

Please ask if there is anything is not clear to you or if you would like more information. Take time to decide whether you want your child to participate.

What is the purpose of the study?

Adolescent idiopathic scoliosis is sideway bending of the spine in children aged 10-18 years old. It can affect their quality of life in many aspects. These effects should be assessed using a questionnaire that is designed for their age group.

The Scoliosis Research Society questionnaire-22r (SRS-22r) is the questionnaire that commonly used for scoliosis patients to assess their quality-of-life. To trust the results obtained from SRS-22r, it should have question and answers that are relevant to their age and their language.

To ensure that the results obtained from the SRS-22r questionnaire are accurate and trustful, it should include questions and answers that are relevant to the age and language of adolescent.

We test this by interviewing adolescents with scoliosis to ask them how scoliosis is affecting their life. Then, the information collected during interviews is analysed, compared, and matched to the content of SRS-22r to assess the suitability of the questionnaire for adolescents with idiopathic scoliosis.

Why have we been chosen?

Your child has been invited to be in this study because he/she has been diagnosed with adolescent idiopathic scoliosis and his/ her age is between 10-15 years old. We hope to have 15 children in this study.

Do we have to take part?

No. It is up to you and your child to decide to join the study. We will explain the study and go through this information sheet with you. If you agree to take part, we will then ask you to sign a consent form. If your child happy to take part, they will be asked to sign an assent form with you, if they want to.

You will be given a copy of the information sheet and your signed consent form, and your child's assent form to keep with your records.

You are free to withdraw your child at any time, without giving a reason. This will not affect the standard care that your child receives.

What will happen to my child if we agree take part?

Your child will be invited to take part in an interview. The interview will be conducted through video call (Microsoft teams or Zoom). If you don't have access to have a video call, the interview can be completed via a telephone call. The preference of which platform to use for the interview will be for you to decide.

During any telephone or video calls, we may ask your child some questions in order for us to verify his/her identity. We will only use this information to confirm who he/she is, and it will not be recorded or stored.

All interviews will be held at a convenient time for you and your child. We will audio record the content of the interview. The audio recordings will then be transcribed (audio to text) using a University approved transcription service. Your child's identity will not be identifiable in the transcripts or audio recordings. The transcripts will be forwarded to your email address so you can review it with your child to ensure that it is accurate. He/she can add any further thoughts if they like. Your child will have the opportunity to talk to the interviewer by video or telephone if required.

What will we have to do?

Interviews will be conducted by an experienced researcher and mother, Samia, who is a physiotherapist and is trained in conducting interviews. Samia will ask your child questions about how scoliosis affects his/her activities of daily living, then they will be asked to fill SRS-22r and give their thoughts and opinion on the content of SRS-22r (relevance of questions). We estimate the interview will last approximately 60 to 90 minutes.

Samia will spend time before and after the interviews chatting with your child about the study. Participation in the interview is entirely voluntary and the interview can be stopped at you or your child request at any point.

An adult family member should be present with your child for the interview, but they will not actively participate in the interview.

What are the possible disadvantages of taking part?

There are no known risks for your child taking part in this study. One potential disadvantage is that when he/she is talking about their condition or the treatment they received, and we are asking him/her to express and explain how it is affecting their daily life, it may be emotional or distressing to them.

We will make every effort to ensure your child is comfortable at all times, and he/she can ask for the interview to be paused to take a break if needed. If required, we can guide you to relevant services which may help your child if he/she get upset at all.

What are the possible benefits of taking part?

By agreeing to your child to take part in this study, we cannot promise that this study will help your child. However, talking about his/her condition and their experience can be valuable. The information in which your child provide may help individuals like him/her in the future.

Expenses and Payments

Since we collect information via the internet, this will not cost you anything.

What will happen when the research stops?

At the end of the research, any future treatment will not be affected in any way. Decisions about your child's future care will be in-line with standard procedure at the hospital.

What will happen if I don't want to carry on with the study?

If you decide you do not wish to your child to carry on with the study, you are free to withdraw him/her at any time, without giving any reason. Your decision will not affect his/her current or future health care.

What if there is a problem?

It is unlikely there will be any problems during the study. If you have a concern about any aspect of the study, for example the way your child has been approached or treated during the study, please contact the chief investigator, Samia who will do her best to answer any questions. If you remain unhappy and wish to complain formally, you can do this by following the NHS complaints procedure. You may obtain advice from the Patient Advice and Liaison Service at the hospital (contact details below).

Will my child's taking part in the research project be kept confidential?

We will need to use information on your child and his/ her interviews for this research project. "This information will include your name, contact information, your child's name and his/her medical records transcripts of his/her interview". People will use this information to do the research or to check your records to make sure that the research is being done properly. People who do not need to know who you are will not be able to see your name or contact details. Personal data will be kept/accessed after the study has ended for a period of 3-6 months so that we can send you a copy of the study findings if you wish to receive this. After that all personal data will be destroyed.

Your child's name will be replaced by a code number. We will keep all information about your child safe and secure on a password protected computer and only accessible by the research team.

Once we have finished the study, we will keep some of the data so we can check the results. We will write reports in a way that no one can identify that your child took part in that study.

The recordings of the interviews will be immediately uploaded to a password protected computer and transcripts will be stored on an encrypted database (Redcap) at the University of Birmingham for ten years following the study. After that period, all information will be destroyed.

What are your choices about how your information is used?

You can withdraw your child being part of the study at any time, without giving reason, but we will keep information about your child that we already have. We need to manage your child records in specific ways for the research to be reliable. This means that we won't be able to let you see the information we hold about your child.

Where can you find out more about how your information is used?

You can find out more about how we use your information at www.hra.nhs.uk/information-about-patients/, by asking one of the research team, contacting Samia Alamrani (details below) or University of Birmingham's data protection officer dataprotection@contacts.bham.ac.uk.

What will happen to the results of the study?

The results of this study will be used to assess the suitability of SRS-22r questionnaire to children with AIS. It will be reported in scientific journals and presentations at research conferences. Your child will not be identifiable in any report, publications, or presentations. Following completion of the study, Samia will send you a short summary of the study results.

Who is organising and funding this study?

The research study is being undertaken as an educational project. It is organized by researchers from the University of Birmingham and sponsored by The Royal Orthopaedic Hospital NHS Foundation Trust.

Who has reviewed the study?

This study has been reviewed by members of the research team with experience and expertise in musculoskeletal physiotherapy, and spinal surgery as well as members of the public.

All research which is conducted within the NHS is reviewed by an independent panel called the Research Ethics Committee. It has also been approved by the Research Department at the hospital.

Contacts for further information or any questions about this study:

Mrs. Samia Alamrani, Principle Investigator, University of Birmingham

Tel:

Email:

Mr. Adrian Gardner, Chief Investigator. The Royal Orthopaedic Hospital NHS Foundation Trust

Tel:

Email:

Dr Nicola Heneghan, Academic Supervisor, University of Birmingham

Tel:

Email:

Patient Advice and Liaison Service, The Royal Orthopaedic Hospital NHS Foundation Trust

Tel:

Email: roh-tr.pals@nhs.net

Thank you for taking the time to read this information sheet.

Participant Information Sheet (Participant aged 10-12)

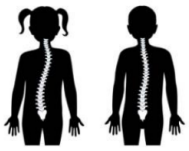
Study title: Content validity of the Scoliosis Research Society questionnaire-22r (**SRS-22r**)

Invitation paragraph

- We would like you to help us with our research study conducted by the University of Birmingham and sponsored by the Royal Orthopaedic Hospital NHS Foundation Trust who are the data controller for the study.
- Please read this information sheet carefully and talk to your parent or carer about it.
- Ask us if there is anything not clear to you, or if you want to know more.
- Take your time to decide if you want to participate. It is up to you if you want to do this.
- If you don't want then it is fine, the hospital will continue to take care of you just the same.

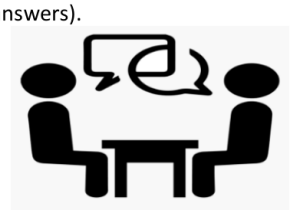
Why are we doing this research?

- Adolescent idiopathic scoliosis is a side bending of spine that happens to young people aged 10 to 18 years old.



- There is a questionnaire named "Scoliosis Research Society questionnaire-22revised", used by doctors/nurses to evaluate how scoliosis affects the quality of life.
- A questionnaire is a "list of questions used to collect information or opinion".

- So, the aim of this research study is to ask young people how scoliosis affects their life.
- Also, we want to know what they think about the content of the SRS-22r (the instructions, the questions, and the choices of answers).



Why have I been invited?

- You have been invited because you have idiopathic scoliosis, and you are between 10-18 years old.
- You would be one of the 15 young people helping us with this study.

Do I have to take part?

- No. It is your choice whether you want to participate in the study or not. You can always change your mind.
- If you would like to participate:
  - You will be asked to sign a form to say that you agree to participate (the assent form)
  - You will be given this information sheet and a copy of your signed assent form for you to keep.
  - Your parent / carer will also be asked to sign a consent form.

What will happen to me if I participate?

- You will be invited to participate in an interview at a time that is comfortable to you.
- We need to use video call by (Microsoft Teams or Zoom) or by using telephone call if you can't make a video call.

- During any video or telephone calls we may ask you some questions to check who you are, but we will not record or keep this information.
- The researcher who is called Samia, will take notes throughout the interview, and who will also audio record the interview.

- Then the recording will be transferred to a typed word format by a specialist company.
- Your name will not be connected to any notes or recordings. You will be given a code number.

- We will send the notes of your interview to the email address of your parent/carer, so you can check if it is correct. Also, you can add any further ideas if you want.

- You are free to stop participating in the research at any time without giving a reason. This will not affect your current or future care. But we need to keep information we have already collected about you.

What will I have to do?

- The interview will be done by an experienced researcher and mother, Samia, who is also a physiotherapist and is trained in asking questions.

- The interview will last approximately 60 to 90 minutes.

- It will include questions about your scoliosis and how it's affecting your daily life.

- Samia will spend time before and after the interview chatting with you about the study.

- Doing the interview is completely optional and the interview can be stopped at any time if you like.

- Your parent/carer or any adult family member should be with you for the interview, but they will not say anything, as we want to know what you think.

What are the possible disadvantages of taking part?

- There are no known dangers for participating in this study.

- One possible thing that may happen, is that while you are talking about your scoliosis or the treatment you received, and we are asking you to explain how it is affecting your life this may upset you.

- We will make every effort to ensure that you are comfortable at all times, and you can ask for the interview to be paused to take a break if you need.

- If required, we can guide you to appropriate services, which may help you if you do get upset at all.

What are the possible benefits of taking part?

- We cannot promise that this study will help you, however talking about your experiences may help people like you in the future.

Expenses and Payments

- Since we collect information via the internet, this will not cost you or your family anything.

What will happen when the research finishes?

- When the research finishes, the hospital will continue to take care of you as before.

- You should still ask your doctor if you have any questions about your back.

What will happen if I don't want to do the research anymore?

- Just tell your parent/ carer or the researcher at any time. They will not be cross with you. You will still have the same care whilst you are at hospital.

- All information collected about you for the study will not be connected to your hospital information.

What if there is a problem?

- It is unlikely there will be any problems during the study.

- In case you have a problem about any part of the study, please ask your parent/carer to contact the researcher, Samia who will do her best to answer any questions.

- If you remain unhappy and you want to complain formally, your parent/carer can do this by following the National Health Service complaints procedure. You can have advice from the Patient Advice and Liaison Service at the hospital (contact details below).

How will we use information about you?

- We will need to use information from you, your medical records, and the interview for this research project.
- This information will include your initials, name and contact details.
- People will use this information to do the research or to check your records to make sure that the research is being done properly.
- Your personal data will be kept/accessed after the study has ended for a period of 3-6 months so that we can send you a copy of the study findings if you wish to receive this. After that all personal data for the purpose of the research will be destroyed.

Will anyone else know I'm doing this?

- We will keep all information about you safe and secure. It can be accessed by the research team only.

- People will not be able to see your name or contact details. You will be given a code number instead.

- Information collected during the study will be stored on University of Birmingham on password protected computers for 10 years. After that period, all information will be destroyed.

- Once we have finished the study, we will keep some of the information so we can check the results.

- We will write out reports in a way that no one can find out that you took part in that study.

What are your choices about how your information is used?

- You can stop taking part at any time, without giving reason, but we will keep information about you that we already have.

Where can you find out more about how your information is used?

- You can find out more about how we use your information at www.hra.nhs.uk/information-about-patients/, by asking one of the research team, contacting Samia (details below) or University of Birmingham's data protection officer dataprotection@contacts.bham.ac.uk.

What will happen to the results of the study?

- When the study has finished, we will present our findings in medical magazines, websites, and conferences.

- The results will also be included as part of Samia's educational qualification. They will be anonymous, which means that you will not be able to be identified by anyone.

- Following completion of the study, Samia will provide a short summary of the study results.

Who is organising and funding this study?

- The research study is being undertaken as an educational project. It is organised by the University of Birmingham and sponsored by The Royal Orthopaedic Hospital NHS Foundation Trust.

Who has reviewed the study?

- Before the research is allowed to go ahead it has been checked by a group called the Research Ethics Committee.

Contacts for further information or any questions about this study:

Mrs. Samia Alamrani, Principal investigator, University of Birmingham
Tel:
Email:

Mr Adrian Gardner, Chief Investigator. The Royal Orthopaedic Hospital NHS Foundation Trust
Tel:
Email:

Dr Nicola Heneghan, Academic supervisor, University of Birmingham
Tel:
Email:

Patient Advice and Liaison Service, The Royal Orthopaedic Hospital NHS Foundation Trust
Tel:
Email: roh-tr.pals@nhs.net

Thank you for taking the time to read this information sheet.

<*To be printed on NHS Trust Headed paper – delete this text on completion.*

UNIVERSITY OF BIRMINGHAM

Participant Information Sheet for (Participants aged 13-15)

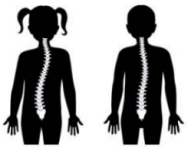
Study title: Content validity of the Scoliosis Research Society questionnaire-22r (SRS-22r)

Invitation paragraph

- We would like you to help us with our research study conducted by the University of Birmingham and sponsored by the Royal Orthopaedic Hospital NHS Foundation Trust who are the data controller for the project.
- Please read this information carefully and talk to your parent or carer about the study.
- Ask us if there is anything that is not clear or if you want to know more.
- Take time to decide if you want to take part. It is up to you if you want to do this.
- If you don't then that's fine, you'll be looked after at the hospital just the same.

Why are we doing this research?

- Adolescent idiopathic scoliosis is the side bending of spine which happens to young people aged 10 to 18 years old.



- The Scoliosis Research Society questionnaire-22r (SRS-22r), is the questionnaire used to measure how scoliosis affects the quality of life.
- Questionnaire means a list of questions used to collect information or opinion.

- The aim of this research is to ask children about how scoliosis having is affecting their life, and their opinion of the SRS-22r (the instructions, the questions, and the options for answers).

Why have I been invited?

- You have been invited because you have idiopathic scoliosis, and you are aged between 10-18 years old. You would be one of the 15 children helping us with this study.

- It will last about 60 to 90 minutes and it will include questions about your scoliosis and how it is affecting your daily life.

- Samia will spend time before and after the interviews chatting to you about the study.

- Doing the interview is completely optional and the interview can be stopped at any point if you like.

- Your parent/carer or any adult family member should be with you for the interview but will not say anything as we want to know what you think.

What are the possible disadvantages of taking part?

- There are no known dangers for taking part in this study.

- One possible thing that may happen is that while you are talking about your condition or the treatment you received, and we are asking you to express and explain how it is affecting your life this may upset you.

- We will make every effort to ensure you are comfortable at all times and can ask for the interview to be paused to take a break if you needed. If required, we can guide you to relevant services which may help you if you do get upset at all.

What are the possible benefits of taking part?

- We cannot promise that this study will help you, however talking about your condition and your experiences may help children like you in the future.

Expenses and Payments

- Since we collect information via the internet, this will not cost you or your parents anything.

What will happen when the research finishes?

- When the research finishes, the hospital will continue to look after you as before. You should still ask your doctor if you have any questions about your back.

What will happen if I don't want to do the research anymore?

- Just tell your parent/ carer or the researcher at any time. They will not be cross with you. You will still have the same care whilst you are at hospital.

- All information which is collected about you for the study is not linked to your hospital record.

What if there is a problem?

- It is unlikely there will be any problems during the study. If you have a problem about any part of the study, please ask you parent to contact the researcher, Samia will do her best to answer any questions.

- If you remain unhappy and wish to complain formally, your parent can do this by following the National Health Service complaints procedure. You may obtain advice from the Patient Advice and Liaison Service at the hospital (contact details below).

How will we use information about you?

- We will need to use information from you, from your medical records and transcripts of your interview for this research project.
- This information will include your initials, name, and contact details.
- People will use this information to do the research or to check your records to make sure that the research is being done properly.
- Once the study has ended, your personal information will be kept for a period of 3-6 months so that we can send you a copy of the study findings if you wish to receive this. After that, all your personal data will be destroyed.

Will anyone else know I'm doing this?

- We will keep all information about you safe and secure. It can only be accessed by the research team.
- People will not be able to see your name or contact details. You will be given a code number which will be used instead.
- Information collected during the study will be stored on University of Birmingham on password protected computers for ten years. After that period, all information will be destroyed.
- Once we have finished the study, we will keep some of the information so we can check the results.
- We will write out reports in a way that no one can find out that you took part in that study.

What are your choices about how your information is used?

- You can stop taking part at any time, without giving reason, but we will keep information about you that we already have.
- People who do not need to know who you are will not be able to see your name or contact details.
- We need to manage your records in specific ways for the research to be reliable. This means that we won't be able to let you see or change the data we hold about you.

Where can you find out more about how your information is used?

- You can find out more about how we use your information at www.hra.nhs.uk/information-about-patients/, by asking one of the research team, contacting Samia (details below) or University of Birmingham's data protection officer dataprotection@contacts.bham.ac.uk.

What will happen to the results of the study?

- When the study has finished, we will present our findings in medical magazines, websites, and conferences.
- The results will also be included as part of Samia's educational qualification. They will be anonymous, which means that you will not be able to be identified from them.
- Following completion of the study, Samia will provide a short summary of the study results.

Who is organising and funding this study?

- The research study is being undertaken as an educational project. It is organized by the University of Birmingham and sponsored by The Royal Orthopaedic Hospital NHS Foundation Trust.

Who has reviewed the study?

- Before the research is allowed to go ahead it has been checked by a group called the Research Ethics Committee.

Contacts for further information or any questions about this study:

Mrs. Samia Alamrani, Principla Investigator, University of Birmingham
Tel: 07846732191
Email: sxa1258@student.bham.ac.uk

Mr. Adrian Gardner, Chief Investigator. The Royal Orthopaedic Hospital NHS Foundation Trust
Tel:
Email:

Dr Nicola Heneghan, Academic Supervisor, University of Birmingham
Tel:
Email:

Patient Advice and Liaison Service, The Royal Orthopaedic Hospital NHS Foundation Trust
Tel:
Email: roh-tr.pals@nhs.net

Thank you for taking the time to read this information sheet.

Participant Information Sheet for (Participants aged 16-18)

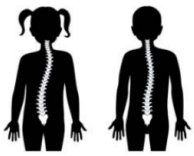
Study title: Content validity of the **S**coliosis **R**esearch **S**ociety questionnaire-22 **r**evised (SRS-22r) for Adolescents with Idiopathic Scoliosis

Invitation paragraph

- We would like to you to help us with our research study conducted by the University of Birmingham and sponsored by the Royal Orthopaedic Hospital NHS Foundation Trust who are the data controller for the project.
- Please read this information carefully and talk to others if you wish.
- Ask us if there is anything that is not clear to you or if you want to know more.
- Take time to decide if you want to participate. It is up to you if you want to do this.
- If you don't then that's fine, you'll be looked after at the hospital just the same.

What is the purpose of the study?

- Adolescent idiopathic scoliosis is a sideway bending of the spine which happen in young people aged 10-18 years old.
- The Scoliosis Research Society questionnaire-22r (SRS-22r) is the questionnaire used to measure how scoliosis affects the quality of life.
- Questionnaire means a list of questions used to collect information or opinion.
- The aim of this research is to ask about how scoliosis is affecting your life, and your opinion of the SRS-22r (the instructions, the questions, and the choices for answers).



Why have I been invited?

- You have been invited to participate because you have idiopathic scoliosis, and you are aged between 10-18 years old. You would be one of the 15 young people helping us with this study.

Do I have to take part?

- No. It is your choice whether you want to be part of the study or not. You can always change your mind, and your treatment will remain the same.

If you do decide to participate

1. You will be asked to sign a form to say that you agree to participate (consent form)
2. You will be given this information sheet and a copy of your signed consent form to keep.

What will happen to me if I take part?

- You will be invited to participate in an interview at your convenience, using video call (Microsoft teams or Zoom) or a telephone call if you cannot make a video call.

- During any telephone or video calls, we may ask you some questions to check who you are, and we will not record or store this information.

- The researcher who is called Samia, will take notes throughout the interview, and who will also audio record the interview.
- The audio recordings will be turned in to typed text by a specialist company.
- Your details will not be recorded in the audio recordings or any notes taken.
- We will forward the notes of your interview to your email address so you can check if it is correct, and you can add any further ideas if needed.
- You are free to stop participating at any time without giving a reason. However, in that situation we need to keep information we already collected about you.

What will I have to do?

- The interview will be with an experienced researcher and mother, Samia, who is a physiotherapist, and she trained in conduct interviews.
- It will last about 60 to 90 minutes, and it will include questions about how idiopathic scoliosis affects your daily life activities.
- Samia will spend time before and after the interviews chatting with you about the study.
- Doing the interview is completely optional, and interview can be stopped at any point if you want.

What are the possible disadvantages of taking part?

- There are no known dangers from participation in this study.
- One possible thing that may happen is that while you are discussing your condition and treatment you have received, or when we ask you to express and explain how it affects your life, this may upset you.
- We will make every effort to ensure you are comfortable at all times, and you can ask for the interview to paused to take a break if needed. If required, we can guide you to relevant services which may help you if you get upset at all.

What are the possible benefits of taking part?

- We cannot promise that this study will help you, however talking about your condition and your experience may help young person/people like you in the future.

Expenses and Payments

- Since we collect information via the internet, this will not cost you anything.

What will happen when the research stops?

- When the research finishes, the hospital will continue to look after you as before.
  You should still ask your doctor if you have any questions about your back.
- All information which is collected about you for the study is not connected to your hospital record.

What will happen if I don't want to carry on with the study?

- If you decide you do not wish to continue in the study, you are free to withdraw at any time, without having to give a reason. You will still have the same care when you are at hospital.

What if there is a problem?

- It is unlikely there will be any problems during the study. If you have a worry about any part of the study, please contact the chief investigator, Samia who will do her best to answer any questions.
- If you remain unhappy and wish to complain formally, you can do this by following the National Health Service complaints procedure. You may obtain advice from the Patient Advice and Liaison Service at the hospital (contact details below).

How will we use information about you?

- We will need to use information from you, from your medical records and transcripts of your interview for this research project.
- This information will include your initials, name and contact details.
- People will use this information to do the research or to check your records to make sure that the research is being done properly.
- Your personal data will be kept/accessed after the study has ended for a period of 3-6 months so that we can send you a copy of the study findings if you wish to receive this. After that all personal data will be destroyed.

Will anyone else know I'm doing this?

- We will keep all information about you safe and secure.
- Your information will be accessed by the research team only.
- You will be given a code number which will be used instead.

- People who do not need to know who you are will not be able to see your name or contact details.
- Information collected during the study will be stored on University of Birmingham on password protected computers for ten years. After that period, all information will be destroyed.
- Once we have finished the study, we will keep some of the information so we can check the results.
- We will write reports in a way that no one can find out that you participated in that study.

What are your choices about how your information is used?

- You can stop participating in a study at any time, without giving reason, but we need to keep information about you that we already have.
- We need to manage your records in specific ways for the research to be reliable. This means that we won't be able to let you see or change the data we hold about you.

Where can you find out more about how your information is used?

- You can find out more about how we use your information at www.hra.nhs.uk/information-about-patients/, by asking one of the research team, contacting Samia (details below) or University of Birmingham's data protection officer dataprotection@contacts.bham.ac.uk.

What will happen to the results of the study?

- When the study has finished, we will present our findings in medical magazines, websites, and conferences.
- The results will also be included as part of Samia's educational qualification. They will be anonymous, which means that you will not be able to be identified from them.
- Following completion of the study, Samia will provide you with a short summary of the study results.

Who is organising and funding this study?

- This research study is being undertaken as an educational project. It is organized by the University of Birmingham and sponsored by The Royal Orthopaedic Hospital NHS Foundation Trust.

Who has reviewed the study?

- Before the research is allowed to go ahead it has been checked by a group called the Research Ethics Committee.

Contacts for further information or any questions about this study:

Mrs. Samia Alamrani, Principle investigator, University of Birmingham
Tel:
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Mr Adrian Gardner, Chief Investigator. The Royal Orthopaedic Hospital NHS Foundation Trust
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Email:

Dr Nicola Heneghan, Academic supervisor, University of Birmingham
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Thank you for taking the time to read this information sheet.

<*To be printed on NHS Trust Headed paper – delete this text on completion.*

UNIVERSITY OF BIRMINGHAM

PARENT CONSENT FORM

Title of Project: Content validity of the Scoliosis Research Society questionnaire-22r (SRS-22r) for Adolescents with Idiopathic Scoliosis

Participant's Study Identification Number: _____

Please initial

1.	I confirm that I have read and understand the Participant Information Sheet, dated 10th May 2021 (version 4), for the above study. I have had the opportunity to consider the study information, ask questions and have had these answered satisfactory.	☐
2.	I choose voluntary to allow my child to participate in the study. I understand that he/she is free to withdraw at any time, without giving any reason, without his/her medical care or legal rights being affected. I understand that if he/she withdraws from the study, his/her data up to the point of withdrawal will be used in the data analysis.	☐
3.	I agree to the interviews being video recorded. I understand that the audio recording will be shared with a transcription service provider with whom contractual agreements are in place. I agree to my child data being shared securely with the transcription services for the purpose of this study.	☐
4.	I understand that relevant sections of my child medical records may be looked at by responsible individuals the Royal Orthopaedic Hospital, where it is relevant to my child participation in this research. I give permission for these individuals to have access to my child records.	☐
5.	I understand that my child research data will be kept confidential and anonymised and be securely stored on servers at the University of Birmingham for a period of ten years, which will only be accessible to designated researchers.	☐
6.	I understand that my child personal information will be kept confidential.	☐
7.	I confirm that I agree for the use of anonymised direct quotes of my child in publications.	☐
8.	I confirm that I agree for my child to participate in this study.	☐
9.	Optional: I wish to receive a summary of the study results.	☐

_____	_____	_____
Name of Parent of participant	Date	Signature
_____	_____	_____
Name of person taking consent	Date	Signature

When completed: 1 copy for Participant
1 copy for medical notes / care record
1 (original) copy for participating site investigators site file

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ASSENT FORM (Participant aged 10-15 years)

Title of Project: Content validity of the Scoliosis Research Society questionnaire-22r (SRS-22r)

Please put a circle around **'Yes'** for all the things that you agree with and put a circle around **'No'** for all the things you do not agree with.

1. Have you read about the study on the content validity of SRS-22r (or it has been read aloud to you)?
   Yes / No

2. Has someone explained to you what the project is about?
   Yes / No

3. Do you understand what the project is about?
   Yes / No

4. Have you been given a chance to ask questions and asked all the questions you want to ask?
   Yes/No

5. Has someone answered all your questions?
   Yes/No

6. Did you understand the answers?
   Yes/No

7. Do you understand that you can stop taking part in the project at any time if you want to, and that you will not have to say why you don't want to carry on?
   Yes/No

8. Do you agree to us recording what you say during the interview?
   Yes/No

9. Do you understand that your personal information will be kept confidential, and only be looked at by authorised persons in the University of Birmingham and Royal Orthopaedic Hospital?
   Yes/No

10. Do you agree that your research information will be kept confidential and anonymised and securely stored at the University of Birmingham for 10 years?
    Yes/No

11. Would you like to join this study?
    Yes/No

12. Would you like to join this study?
    Yes/No

13. Optional: I wish to be sent a summary of the study results
    Yes/No

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Write your name here if you would like to join this study and you have circled yes to all the questions.

_____	_____	_____
Name of Participant	Date	Signature
_____	_____	_____
Name of Parent/carer	Date	Signature
_____	_____	_____
Name of person taking consent	Date	Signature

When completed: 1 Copy for Participant
1 (original) copy for participating site investigators site file

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CONSENT TO CONTACT FORM

Title of Project: Content validity of the Scoliosis Research Society questionnaire-22r (SRS-22r) for Adolescents with Idiopathic Scoliosis

		Please initial
1.	I confirm that my name, date of birth, diagnosis, phone number and address will be shared with a member of the research team located at the University of Birmingham.	
2.	I understand that the above mentioned details will be shared through NHS secured email.	
3.	I understand that the member of the research team will contact me with more information about this study, after which I can decide whether I would like to participate.	

_____	_____	_____
Name of Parent of participant	Date	Signature
_____	_____	_____
Name of person taking consent	Date	Signature

When completed: 1 copy for Participant
1 copy for medical notes / care record
1 (original) copy for participating site investigators site file

Topic guide (Individuals with AIS)

Research Aim	Content validity of the Scoliosis research society questionnaire		
Interview Section	Questions/Content	Prompts	Aims
Ethics Statement	Firstly, I would like to thank you for agreeing to take part in this interview. "My name is [Samia]. I am a student in University of Birmingham. I am also a mother and one of my kids is the same age as you. Today, I will be chatting with you about your back/ "curve". Just a reminder that this interview will be audio/video-recorded, but all information will be kept confidential and anonymised, so you are not identifiable from the questions/answers you give during the recording. If you change your mind at any point, just tell me that you want to stop the interview; it is absolutely fine to do that. This will not affect your current and future health care. You also have the right not to answer a question if you do not wish to. There are no right or wrong answers. I am interested in your own experiences AND thoughts. The aim of today is to understand your experiences with living with scoliosis. If I ask you a question and you don't know what I mean, please just say 'I don't get it' or 'I don't know what you mean,' and I'll try to ask it another way. Just to reassure you- none of the answers or information that you give me will be associated with you and your name will be replaced by	• *Can I confirm that you have read and understand the information sheet and signed the consent/assent form?* • *Is this okay for you?"*	• To ensure full understanding of what is expected of the participant during this interview. • Make sure the participant is comfortable and ready to begin.

	code number when we report the findings. Once the information from the interview has been used, the recordings will be deleted. Before we start, is there anything you'd like to ask?		
Introductory Questions	• Can you tell me a bit about yourself? • What do you like to do for fun or in your spare time? • Do you like to go for a walk, or other activities like exercise? • Do you like exercising?	• *How old are you ? in which year at school you are? Do you have any brothers and sisters?* • *What is your favourite subject?* • *Which type of exercise is your favourite? Basketball, ballet, swimming?*	• Make participant relax and feel comfortable with talking and opening up. • Build rapport.
Transition Questions	• Talking about your curve/back, what do you call your back condition when you talk to family and friends? • When did you first became aware of your curve/back?	• *Can you tell me how you have been diagnosed ?* • *What was your thoughts about it ?* • *How did you feel ?*	• Start to guide the interview towards experiences of patient with scoliosis • To get an idea of their thoughts of having scoliosis. • Explore feeling and perceptions of having scoliosis.
Background of the content validity of SRS-22r questionnaire	As you have seen in the information sheet the SRS-22r is a questionnaire that assesses quality of life in people with scoliosis. To remind you QOL is measure of health, comfort, and ability to participate or enjoy life events. As part of my research we want to see if the SRS-22r is suitable for young people like you. To do this we want to understand your experiences with curve/back, Also, we want to see if the questionnaire is clear, covers all important areas –i.e. is it suitable. Today, our interview will be in two parts. In first part we would like to hear how you feel having curve/back, and how you feel it affects your life. In the second part	• *Do you have any questions?* • *If you are unsure at any point regarding the study, please ask.*	• Inform the participant of the background of the study • Ensure the participant knows they can ask questions if they are unsure.

		• *Do you think it is limiting your ability to do things? How ?* • *Do you think it is affecting your relationship with other?*	
	• Can you tell me what management/treatment that you have received for your back?	• *What was the treatment you received for your back?* • *How do you feel/think about it ?* • *Do you have an idea about other options of treatments you might receive for your back ? what is it ?* • *How do you feel if you have the same treatment again?*	• To explore the effect of scoliosis on satisfaction
	• Now, we have reached the end of first part of this interview	• *Do you need a break?*	
Cognitive debriefing	• For the second part of our discussion I want to see if the SRS-22r questionnaire is suitable for young people like you • I will read the question one by one to you. Then please tell me, what do you understand the question is asking?		• To test the relevance of SRS-22r items to individuals with AIS • To assess if the SRS-22r items are understandable by individuals with AIS. • To assess if the is SRS-22r is comprehensive.
	1. Which one of the following best describes the amount of pain you have experienced during the past 6 months? -Response options are: None, Mild, Moderate, Moderate to severe, Severe.	• *Can you tell me in your words, what this question is asking you about?* • *Do you think this question is Important to you ? why?* • *Is there any word that is not clear to you ? what is it?* • *Can you understand the answers? Is there any word that is not clear to you? what is it?*	
	2. Which one of the following best describes the amount of pain you have experienced over the last month? -Response options are: None, Mild, Moderate, Moderate to severe, Severe	• *Can you tell me in your words, what this question is asking you about?* • *Do you think this question is Important to you ? why?* • *Is there any word that is not clear to you ? what is it?* • *Can you understand the answers? Easy to choose the answer?*	
	3. During the past 6 months have you been a very nervous person? - Response options are: None of the time, A little of the time, Some of the time, Most of the time, All of the time.	• *Do you think this question is relevant to you? or Important to you ? why?* • *Is there any word that is not clear to you ? what is it?* • *Was it easy or difficult to choose that particular answer? Why?*	

	4. If you had to spend the rest of your life with your back shape as it is right now, how would you feel about it? - Response options are : Very happy, Somewhat happy, Neither happy nor unhappy, Somewhat unhappy, Very unhappy	• *What do you think the question is asking you about?* • *Is it Important to you ?why?* • *Was it easy or difficult to choose your answer? Why?* • *Do you find this question too personal/ or embarrassing? Why?*	
	5. What is your current level of activity? Response options are : Bedridden, Primarily no activity, Light labour and light sports, Moderate labour and moderate sports, Full activities without restriction.	• *Do you understand the question? is it important to you?* • *Are there any words that are not clear to you? What is your thought about these words "labour", "Bedridden", "restrictions"?* • *Was it easy or difficult to choose your answer? Why?*	
	6. How do you look in clothes? - Response options are : Very good, Good, Fair, Bad, Very bad	• *Do you understand the question? is it important to you?* • *Was it easy or difficult to choose your answer? Why?*	
	7. In the past 6 months have you felt so down in the dumps that nothing could cheer you up? -Response options are :Very often, Often, Sometimes Rarely, Never	• *Do you think this question is relevant to you?* • *Are there any words that are not clear to you? Do you understand word "dump "? What does it mean?* • *Do you find this question too personal/ or embarrassing? Why?* • *Was it easy or difficult to choose your answer? Why?*	
	8. Do you experience back pain when at rest? - Response options are : Very often, Often, Sometimes, Rarely, Never	• *Do you think this question is important to you?* • *Was it easy or difficult to choose your answer? Why?*	
	9. What is your current level of work/school activity? - Response options are : 100% normal, 75%normal 50% normal, 25% normal, 0% normal	• *What went on in your mind when you were asked the question?* • *is there any word that you don't understand?* • *What is your answer? Would you explain to me how you come out with that answer ?*	
	10. Which of the following best describes the appearance of your trunk; defined as the human body except for the head and extremities? -Response options are : Very	• *What do you think the question is asking you about?* • *Is there any word that you don't understand?* • *Was it easy or difficult to choose your answer? Why?*	

	good, Good, Fair, Poor, Very Poor		
	11. Which one of the following best describes your pain medication use for back pain? -Response options are : None, Non-narcotics, weekly or less (e.g., aspirin, Tylenol, Ibuprofen) , Non-narcotics daily, Narcotics weekly or less (e.g. Tylenol III, Lorcet, Percocet), Narcotics daily.	• *What went on in your mind when you read the question?* • *Is there any word that is not clear to you ? what is it?* • *What does the word "* Non-narcotics" *means to you?* • *What is your answer? Would you explain to me how you came out with that answer ?*	
	12. Does your back limit your ability to do things around the house? - Response options are: Never, Rarely, Sometimes Often, Very Often	• *What do you think about this question? Is it important to you ?* • *What about the answers? Was it easy or difficult to choose your answer? Why?*	
	13. Have you felt calm and peaceful during the past 6 months? -Response options are: All of the time, Most of the time, Some of the time, A little of the time, None of the time.	• *What do you think the question is asking you about?* • *Is there any word that is not clear to you ? what is it?* • *Was it easy or difficult to choose your answer? Why?*	
	14. Do you feel that your back condition affects your personal relationships? - Response options are: None, Slightly, Mildly, Moderately, Severely	• *Do you think this question is important to you?* • *Is there any word that you do not understand?* • *Was it easy or difficult to choose your answer? Why?*	
	15. Are you and/or your family experiencing financial difficulties because of your back? -Response options are: Severely, Moderately, Mildly Slightly, None.	• *What went on in your mind when you read the question?* • *Do you find this question too personal/ or embarrassing? Why?* • *Was it easy or difficult to choose your answer? Why?*	
	16. In the past 6 months have you felt down hearted and blue? -Response options are: Never Rarely, Sometimes, Often, Very often.	• *What do you think the question is asking you about?* • *Is there any word that you do not understand? what about "down hearted', "blue"?* • *Do you find this question too personal/ or embarrassing? Why?* • *Was it easy or difficult to choose your answer? Why?*	

	17. In the last 3 months have you taken any days off of work, including household work, or school because of back pain? -Response options are : 0 days 1 day,2 days,3 days, 4 or more days.	• *Is this question important/relevant to you?* • *What is your answer? Was it easy or difficult to choose your answer?*	
	18. Does your back condition limit your going out with friends/family? -Response options are : Never, Rarely, Sometimes, Often, Very often	• *Is this question important/relevant to you?* • *What is your answer? Was it easy or difficult to choose your answer?*	
	19. Do you feel attractive with your current back condition? -Response options are : Yes, very Yes, somewhat, Neither attractive nor unattractive No, not very much No, not at all	• *Is this question important/relevant to you?* • *Is there any word that you do not understand?* • *Do you find this question too personal/ or embarrassing? Why?* • *What is your answer? Was it easy or difficult to choose your answer?*	
	20. Have you been a happy person during the past 6 months? -Response options are : None of the time, A little of the time, Some of the time, Most of the time, All of the time	• *Is this question important/relevant to you?* • *Do you find this question too personal/ or embarrassing? Why?* • *What is your answer? Was it easy or difficult to choose your answer?*	
	21. Are you satisfied with the results of your back management? -Response options are : Very satisfied, Satisfied Neither satisfied nor unsatisfied, Unsatisfied, Very unsatisfied	• *What do you think the question is asking you about?* • *Is this question important/relevant to you?* • *Are there any words that you don't understand?* • *What is your answer? Was it easy or difficult to choose your answer?*	
	22. Would you have the same management again if you had the same condition? -Response options are : Definitely yes, Probably yes, Not sure, Probably not, Definitely not	• *Do you think this question is important to you?* • *What is your answer? Was it easy or difficult to choose your answer?*	
	• Do you think the questionnaire covers all areas which are important to you ?	• *Can you think of other areas that should be included in the questionnaire?*	• To assess if there are any additional areas need to be covered.

Topic guide (Individuals with AIS)

Research Aim	Content validity of the Scoliosis research society questionnaire		
Interview Section	Questions/Content	Prompts	Aims
Ethics Statement	Firstly, I would like to thank you for agreeing to take part in this interview. "My name is [Samia]. I am a student in University of Birmingham. I am also a mother and one of my kids is the same age as you. Today, I will be chatting with you about your back. Just a reminder that this interview will be audio/video-recorded, but all information will be kept confidential and anonymised, so you are not identifiable from the questions/answers you give during the recording. If you change your mind at any point, just tell me that you want to stop the interview; it is absolutely fine to do that. This will not affect your current and future health care. You also have the right not to answer a question if you do not wish to. There are no right or wrong answers. I am interested in your own experiences AND thoughts. If I ask you a question and you don't know what I mean, please just say 'I don't get it' or 'I don't know what you mean,' and I'll try to ask it another way. Just to reassure you- none of the answers or information that you give me will be associated with you and your name will be replaced by code number when we report the findings.	• *Is this okay for you?"*	• To ensure full understanding of what is expected of the participant during this interview. • Make sure the participant is comfortable and ready to begin.

concept elicitation	• Thinking about your back, do you or have you ever had any difficulties? • Do you ever take any painkillers or medicine for your back?	• *Are there any further issues that you can think of?* • *(For example, your body is hurting you; pain at your joints; You feel pain during or after doing exercise/PE; chest pain; difficult breathing, decrease in the range of your movement? How is that?* • *Can you think of other parts of your body that are affected because of your back? How do you feel it is affected? why?* • *How many times do you take pain medications? Is it effective?* • *What do you do to ease pain other than taking medications?* • *If we want to measure the pain that you usually have. How much would you give it, in a scale from 0 to 10 ?*	• To explore effects of scoliosis on participant's body functions. • To explore the effect of scoliosis on body structures.
	• Can you tell a bit more about any difficulties you have with your back? Which aspects of your daily life does it affects? • Can you explain for me if and how your back affects you at school and activities linked to school?	• *Walking to school/catching the bus, climbing stairs, carrying bag.* • *Sitting, standing, bending, kneeling.* • *Studying/doing your homework, working in lab/art/music class.* • *Do you participate in PE/school sports? How do you feel about doing exercise or sports with your classmates?* • *Have you ever had days off school due to your back issue? Why?*	• To explore the effect of scoliosis on activities and participation (daily life activities). o Mobility o General task demand o Major life areas
	• Does your back affect your ability to do activities with family or friends?	• *How is it affect your visiting relatives ? or friends?* • *Does it affect your ability to go out with family? or friends?* • *Does your back affect your ability to do things around home? For example : playing sport, Joining a game ?*	• To explore the effect of scoliosis on activities and participation (social life). o Interpersonal interactions and relationships o Community, social and civic life

	• Does your curve/back affect your ability to do things at home at all?	• *How your back affect you when taking care of yourself e.g., eating, bathing, toileting, dressing?* • *Tidying up your room, helping family in preparing meals.* • *Assisting others* • *Taking care of pets/plants.*	• To explore effect of scoliosis on activities and participation o Self-care. o Domestic life
	• Can you describe your feelings when you have an issue with your back, how does this make you feel?	• *Does it worry you at all?* • *Do you know what it is specifically that worries you? (Fear of pain, difficulty preforming your homework, your scores in school less than usual, limited in activities that you can do).* • *Does this affect your energy, (you feel tired)?* • *Does your back condition ever make you feel down or discouraged, afraid, angry, or anxious ?* • *Does is it affect your ability to sleep?*	• To explore the effect of scoliosis on mental function o Emotional function o Energy level o Sleep function o Personality function
	• How do you feel about the shape of your back?	• *Do you think your back shape is affecting your life? How is that ?* • *Do you think it is limiting your ability to do things? How ?* • *Do you think about your back when getting ready for sports / changing into uniform or sports kit at school / away from home or with others?* • *Do you think it is affecting your relationship with other?*	• To explore the effect of scoliosis on mental function (body image)? • Explore participants perception on their back shape.
	• Can you tell me what management/treatment that you have received for your back? Can you describe the help that you received during treatment/management?	• *What was the treatment you received for your back?* • *How do you think/feel about it ?* • *Do you have an idea about other options of treatments you might receive for your back ? what is it ?* • *How do you feel if you have the same treatment again?* • *Which type of help/support have you received ? from whom? (Support from family, friends, peers, health professionals (doctors,*	• To explore the experience of participants with their management and their satisfaction. • To explore the effect of environmental factors on scoliosis

		nurses, physiotherapists, other healthcare providers? • *What do you think about it?* • *Have you ever seen a physiotherapist or other therapist (e.g., osteopath/chiropractor)? What did you do with them? Did it help?* • *Did you feel able to continue being active after seeing the physiotherapist?* • *Is there anything you feel would improve your experience of living with scoliosis? what is it?*	
	• Now, we have reached the end of first part of this interview	• *Do you need a break?*	
Cognitive debriefing	• For the second part of our discussion, I want to see if the SRS-22r questionnaire is suitable for young people like you • I will read the question one by one to you. Then please tell me, what do you understand the question is asking?	• *Have you completed this questionnaire before?*	• To test the relevance of SRS-22r items to individuals with AIS • To assess if the SRS-22r items are understandable by individuals with AIS. • To assess if the is SRS-22r is comprehensive.
	1. Which one of the following best describes the amount of pain you have experienced during the past 6 months? -Response options are: None, Mild, Moderate, Moderate to severe, Severe.	• *Can you tell me in your words, what this question is asking you about?* • *Do you think this question is Important to you ? why?* • *Is there any word that is not clear to you ? what is it?* • *Can you understand the answers? Is there any word that is not clear to you? what is it?*	
	2. Which one of the following best describes the amount of pain you have experienced over the last month? -Response options are: None, Mild, Moderate, Moderate to severe, Severe	• *Can you tell me in your words, what this question is asking you about?* • *Do you think this question is Important to you ? why?* • *Is there any word that is not clear to you ? what is it?* • *Can you understand the answers? Easy to choose the answer?*	

	3. During the past 6 months have you been a very nervous person? - Response options are: None of the time, A little of the time, Some of the time, Most of the time, All of the time.	• *Do you think this question is relevant to you? or Important to you ? why?* • *Is there any word that is not clear to you ? what is it?* • *Was it easy or difficult to choose that particular answer? Why?*	
	4. If you had to spend the rest of your life with your back shape as it is right now, how would you feel about it? - Response options are : Very happy, Somewhat happy, Neither happy nor unhappy, Somewhat unhappy, Very unhappy	• *What do you think the question is asking you about?* • *Is it Important to you ?why?* • *Was it easy or difficult to choose your answer? Why?* • *Do you find this question too personal/ or embarrassing? Why?*	
	5. What is your current level of activity? Response options are : Bedridden, Primarily no activity, Light labour and light sports, Moderate labour and moderate sports, Full activities without restriction.	• *Do you understand the question? is it important to you?* • *Are there any words that are not clear to you? What is your thought about these words "labour", "Bedridden", "restrictions"?* • *Was it easy or difficult to choose your answer? Why?*	
	6. How do you look in clothes? - Response options are : Very good, Good, Fair, Bad, Very bad	• *Do you understand the question? is it important to you?* • *Was it easy or difficult to choose your answer? Why?*	
	7. In the past 6 months have you felt so down in the dumps that nothing could cheer you up? -Response options are :Very often, Often, Sometimes Rarely, Never	• *Do you think this question is relevant to you?* • *Are there any words that are not clear to you? Do you understand word "dump "? What does it mean?* • *Do you find this question too personal/ or embarrassing? Why?* • *Was it easy or difficult to choose your answer? Why?*	
	8. Do you experience back pain when at rest? - Response options are : Very often, Often, Sometimes, Rarely, Never	• *Do you think this question is important to you?* • *Was it easy or difficult to choose your answer? Why?*	

	9. What is your current level of work/school activity? - Response options are : 100% normal, 75%normal 50% normal, 25% normal, 0% normal	• *What went on in your mind when you were asked the question?* • *is there any word that you don't understand?* • *What is your answer? Would you explain to me how you come out with that answer ?*	
	10. Which of the following best describes the appearance of your trunk; defined as the human body except for the head and extremities? -Response options are : Very good, Good, Fair, Poor, Very Poor	• *What do you think the question is asking you about?* • *Is there any word that you don't understand?* • *Was it easy or difficult to choose your answer? Why?*	
	11. Which one of the following best describes your pain medication use for back pain? -Response options are : None, Non-narcotics, weekly or less (e.g., aspirin, Tylenol, Ibuprofen) , Non-narcotics daily, Narcotics weekly or less (e.g. Tylenol III, Lorcet, Percocet), Narcotics daily.	• *What went on in your mind when you read the question?* • *Is there any word that is not clear to you ? what is it?* • *What does the word "* Non-narcotics*" means to you?* • *What is your answer? Would you explain to me how you came out with that answer ?*	
	12. Does your back limit your ability to do things around the house? - Response options are: Never, Rarely, Sometimes Often, Very Often	• *What do you think about this question? Is it important to you ?* • *What about the answers? Was it easy or difficult to choose your answer? Why?*	
	13. Have you felt calm and peaceful during the past 6 months? -Response options are: All of the time, Most of the time, Some of the time, A little of the time, None of the time.	• *What do you think the question is asking you about?* • *Is there any word that is not clear to you ? what is it?* • *Was it easy or difficult to choose your answer? Why?*	
	14. Do you feel that your back condition affects your personal relationships? - Response options are: None, Slightly, Mildly, Moderately, Severely	• *Do you think this question is important to you?* • *Is there any word that you do not understand?* • *Was it easy or difficult to choose your answer? Why?*	
	15. Are you and/or your family experiencing financial difficulties because of your back?	• *What went on in your mind when you read the question?*	

	-Response options are: Severely, Moderately, Mildly Slightly, None.	• *Do you find this question too personal/ or embarrassing? Why?* • *Was it easy or difficult to choose your answer? Why?*	
	16. In the past 6 months have you felt down hearted and blue? -Response options are: Never Rarely, Sometimes, Often, Very often.	• *What do you think the question is asking you about?* • *Is there any word that you do not understand? what about "down hearted', "blue"?* • *Do you find this question too personal/ or embarrassing? Why?* • *Was it easy or difficult to choose your answer? Why?*	
	17. In the last 3 months have you taken any days off of work, including household work, or school because of back pain? -Response options are : 0 days 1 day,2 days,3 days, 4 or more days.	• *Is this question important/relevant to you?* • *What is your answer? Was it easy or difficult to choose your answer?*	
	18. Does your back condition limit your going out with friends/family? -Response options are : Never, Rarely, Sometimes, Often, Very often	• *Is this question important/relevant to you?* • *What is your answer? Was it easy or difficult to choose your answer?*	
	19. Do you feel attractive with your current back condition? -Response options are : Yes, very Yes, somewhat, Neither attractive nor unattractive No, not very much No, not at all	• *Is this question important/relevant to you?* • *Is there any word that you do not understand?* • *Do you find this question too personal/ or embarrassing? Why?* • *What is your answer? Was it easy or difficult to choose your answer?*	
	20. Have you been a happy person during the past 6 months? -Response options are : None of the time, A little of the time, Some of the time, Most of the time, All of the time	• *Is this question important/relevant to you?* • *Do you find this question too personal/ or embarrassing? Why?* • *What is your answer? Was it easy or difficult to choose your answer?*	

	21. Are you satisfied with the results of your back management? -Response options are : Very satisfied, Satisfied Neither satisfied nor unsatisfied, Unsatisfied, Very unsatisfied	• *What do you think the question is asking you about?* • *Is this question important/relevant to you?* • *Are there any words that you don't understand?* • *What is your answer? Was it easy or difficult to choose your answer?*	
	22. Would you have the same management again if you had the same condition? -Response options are : Definitely yes, Probably yes, Not sure, Probably not, Definitely not	• *Do you think this question is important to you?* • *What is your answer? Was it easy or difficult to choose your answer?*	
	• Do you think the questionnaire covers all areas which are important to you ?	• *Can you think of other areas that should be included in the questionnaire?*	• To assess if there are any additional areas need to be covered.
Conclusion	• That's all the questions, is there anything else you would like to tell me about your back or the questionnaire. •	• • *Is there anything you would like to ask regarding the analysis of the data or the next steps of the process?*	• Ensure the participant is comfortable with what has been discussed.
	• Thank you for participating in this study, I really appreciate your time and input.		

(Chapter 5) Saturation table showing that concept saturation was achieved												
Themes & subthemes	**Subtheme**	**Interview number**										
		1	**2**	**3**	**4**	**5**	**6**	**7**	**8**	**9**	**10**	**11**
Physical effects												
Physical Symptoms	**1a. Back hurt at rest**		✓									
	1b. Back hurt with brace			✓								
	1c. Back hurt when exercising	✓										
	1d. Back hurt because of surgery		✓									
	Hips hurt						✓					
	Nerve pain, leg ache/weak							✓				
	Shoulder aches	✓										
	Feel stiffened/ inflexibility	✓										
	Feel tired	✓										
	Difficult breathing							✓				
Body asymmetry	**Uneven Shoulders**	✓										
	Uneven hips/waist		✓									
	Leaning to one side		✓									
	Rips stick out							✓				
Activity-related effects												
School-related activities	**Time off school**		✓									
	Focus during lessons	✓										
Mobility	Jumping/jog	✓										
	Hinder balance/walking straight	✓										
	Bending	✓										
	Carrying bags/things	✓										
	Going up stairs							✓				
	Walking long distances		✓									
	Sitting for long time	✓										
	Side-lying	✓										
	Stand for too long	✓										
Self-care	Ache with dressing	✓										
Psychological related effects												
Emotional effects	Annoyed-irritated- frustrated	✓										
	Sad	✓										
	Disappointed	✓										

	Nervous-worry-anxious	✓										
	Afraid				✓							
Body-image	Feel insecure to change in front of others	✓										
	Hate body shape							✓				
	Limit wearing certain clothes					✓						
	Hide back condition					✓						
Mental effects	Pain coping strategies	✓										
	Sleep quality		✓									
	Know about scoliosis	✓										
	Know about options of treatment	✓										
Social effects												
Support	From school			✓								
	From friends		✓									
	Mental health support		✓									
Participation	In sport/physical activities/physical education	✓										
	Join games/social activities with family/friends	✓										

Satisfaction	Satisfaction about given treatment (surgery, physiotherapy, brace)	✓										
No. of new codes appearing in each interview		**25**	**9**	**2**	**1**	**2**	**1**	**5**	**0**	**0**	**0**	**0**
% Of total new codes (Total =45)		**55.5**	**20**	**4.4**	**2.2**	**4.4**	**2.2**	**11.1**	**0**	**0**	**0**	**0**

Theme, subthemes, codes (Chapter 5)		
Theme	**Subtheme**	**Codes "Participants quotes"**
Physical effects		
Physical Symptoms	1a. Back hurt at rest	'It really depends, to be honest. There's nothing in particular which brings it on. It just happens. It can just happen. I might be just at rest, sat in a chair for too long, and it can hurt' (P2)
	1b. Back hurt with brace	'The only pain I get is because of my brace' (P3)
	1c. Back hurt when exercising	'Sometimes it hurts my back when I do some exercises …. Like at times, when I play football, it hurts my back and if I do too many exercises and stuff, it starts to make my back hurt.' (P11)
	1d. Back hurt because of surgery	'I wouldn't have been able to exercise or walk to help the pain before surgery, because that was what was causing my pain, whereas now if I do get pain, it's because of the surgery' (P8)
	Hips hurt	'My hips, I guess, my hips start hurting a bit but not as noticeable as my back' (P6)
	leg ache/nerve pain	'I leg-ache a lot, but we don't what that is caused by' (P8)
	Shoulder aches	'I think just my shoulders and my hips sometimes because it's kind of like disjointed everything. They ache most of the time and no matter how many medications I can take for it, it'll always be there because it just reoccurs' (P1) 'I think, when I wake up in the morning, they'll be really tight, and they'll be aching.' (P6)
	Feel stiffened/inflexible	'I'm less flexible; I've never really been that flexible. I used to be really; I could do anything. But now it feels kind of stiffened and I've definitely lost quite a lot of flexibility and sometimes mobility' (P1)
	Difficult breathing	'It makes my lungs hurt from running, so then it makes it difficult breathing.' (P7)
	Feel tired	'I feel more tired than other people after I do a lot of things and I have to take more breaks than most of my friend's when they do the same things. So usually, I'm just way more tired.' (P1)
Body asymmetry	Uneven Shoulders	"She'd noticed that my shoulders were uneven, and she tried to like straighten them but they wouldn't straighten" (P10)
	Uneven hips/waist	'I was 13, but for a year I had noticed that one side of my waist was straight and the other side was going in like that.' (P8)
	Leaning to one side	"I was constantly in a state of leaning to the one side. It was quite apparent as soon as I was diagnosed" (P2)
	Rips stick out	"that's when my back started getting worse and I noticed my rib would stick out and it progressively got a lot worse, pain and looks-wise, and I just wasn't happy with my back at all. I was very upset" (P8)
Activity related effects		
School-related activities	Time off school	'I had a lot of time off. I was off for five months.' 'Before the surgery I was off every day pretty much. I would go to school for an hour or two and the pain would get so bad that I'd have to call Mum or Dad to come and pick me up.' (P8)
	Focus during lessons	'Sometimes it's definitely difficult to focus when my back is hurting that much and I'm trying to get on with the lesson. It's worse in science because we have these chairs, they don't have a back on and so I have nothing to lean against. It makes me struggle because I'm trying to focus, but then trying to make sure that my back isn't bending or hunching over because it would hurt more.' (P1)
Self-care	Ache with dressing	'Sometimes dressing, sometimes it's – not painful, but really aching and I just can't be bothered. It's just quite a challenge every morning.' (P1)
Mobility	Jumping/jog	'Running. Football, I've played a bit of football since, it's a bit difficult. Anything kind of above a jog, it'll sometimes start hurting.' (P6) 'I started to jump, I'd been many times before, but this was the only time that it actually started to hurt.' (P1)
	Hinder balance/walking straight	'I do think it hinders my balance as well, so maybe that's why I can't stand up for very long periods of time without feeling dizzy.' 'Walking in straight... I can't walk in like... well, I feel like I'm not walking in a straight line' (P7)

	Bending	'Bending over, that kind of thing; I tend to avoid bending over. If I have to tie my shoes or something like that, I'll put my leg up on a chair. I don't tend to bend down as such' (P2) 'It's because I can't bend my back, like how you would bend to stretch your toes, I can't bend like that with the brace on ' (P3)
	Carrying bags/things	'I had like a handbag for school, so obviously there's more pressure on one side and I used to have to get my friends to carry them because my back wasn't strong enough' (P5) 'I think sometimes if I had to, let's say, carry a heavy basket of washing or something, I'd potentially struggle with that' (P2)
	Going up stairs	'Pain mostly. Walking in straight... I can't walk in like... well, I feel like I'm not walking in a straight line. But then... And then walking far distances and going up stairs.' (P7)
	Walking long distances	'And then walking far distances.' (P7) 'I guess walking for like a while, it'll be noticeable, but short distance is not too bad' (P6).
	Sitting for long time	'When doing sports activities, it causes pain in my back and sitting down for long periods of time also affects it' (P11).
	Side lying	'At the start it used to be really bad, every single time I lay in a certain position, and it started to really ache.' (P1)
	Stand for too long	'If I have to stand around for too long, it starts to really hurt and then I have to immediately sit down.' (P1) 'Before, not really. But since, it hurts to sit down or stand up for too long, I guess. (P6)
Psychological effects		
Emotional effects	Annoyed-irritated-frustrated	"It makes me feel annoyed because most people don't have this, but what can you do?' (P3) 'I'm quite moody. Sometimes I'm stroppy about it and I'm annoyed…oh irritated" (P7)
	Bad/sad/bothering	"It makes me sad because it reminds me of being in pain a lot before the surgery, because the biggest I wanted out of the surgery was to help my pain, even if it was just a little bit" (P8)
	Disappointed	"I feel quite disappointed that there can't be anything done. …. Yeah, I think just disappointment is the biggest one" (P1)
	Nervous-worry-anxious	'I'm nervous about having the surgery'. 'I feel a bit worried, like how it will affect me in the future and how it's going to impact on my life. Just a bit nervous about if it will get worse in the future or not' (P11).

	Afraid from surgery/future with scoliosis	'I didn't really want it to get bad like my sister' (P4)
Body image	Feel insecure to change in front of others	'I wouldn't get changed in front of others. I'd go in the toilet cubicle when we were getting ready for PE. I wouldn't get dressed with all the other girls. I'd go somewhere separately. I'd have to wear a jacket with everything, or a jumper, and even if I was covered up, I was still very insecure.' (P8)
	Hate body shape	'I hate it. It's given me the worst body dysmorphia' (P10)
	Limit wearing certain clothes	'I'd rather not swim, I guess. I don't hate swimming but just wearing shorts and my back is like… I'd rather not show it.' (P6)
Mental effects	Pain coping	'It's usually just a case of I just have to lie down for a minute. ..that's literally the way to quickly cure it, is lie down.' (P5) 'If I feel the pain coming on, I've got a routine I get on to, "Okay, this is coming on; before it gets worse, let me just rest for 10 minutes and then see how I feel.' (P2)
	Know about scoliosis	'I think it was just the fact that, in the early, early days, I just did not have a clue how that was going to affect my life.' (P2)
	Know about options of treatment	"I remember the first time I went to the appointment, the person basically said, you're eligible for surgery now. And I was like, what? Obviously, we didn't even know what it was" (P5-Post-surgery).
	Sleep	'I think my sleep affects me mentally, from anxiety more than it does pain. It's more the mental side effects from the scoliosis than the physical.' (P2)
Social effects		
Support	From school	'So, I have a time out card at school, meaning I can have a walk outside the classroom for like five minutes. And then I also have a five-minute early pass, so I leave five minutes early from lessons, so the other lesson.' (P7)
	From friends	'I look back on it, I would have definitely told my friends. Because when I did tell them eventually, literally when I got told that I was having it and they were all fine and supportive and things like that. When I look back on it, I think why you not just told them' (P5)

	Mental health support	‘Probably would have been good for them to think about the mental effects as well, because obviously, spines and surgery, that kind of thing, they are just focused on the metalwork and whether the actual back is okay. But I don’t feel like anybody really bothered about how I was feeling…And I think, definitely, the treatment you get post-op; you don’t get any mental support.’ (P2)
Participation	In sport/physical activities/physical education	‘It definitely stops a lot of things to do with PE as well because there’s a lot of physical activities, mainly like yoga and things, because they always say bending in all these different directions. But I’m not that flexible and I can’t do that.’ (P1) ‘I guess I can’t really do like sports. The amount of sports I’m able to do comfortably has kind of narrowed.’ (P6)
	Join games /social activities with family friends	‘Sometimes it affects it, like say you want to go on like walks or something for a long period of time, then it would hurt’ (P11) ‘I couldn’t go out with my friends or anything, if they asked me to go out or sleep over. It just restricted me to stay at home because I physically couldn’t do anything I was in that much pain and I was very, very deformed’. (P8)
Satisfaction	Satisfaction about given treatment	‘I would comply with what they say, but I would feel quite disappointed because I would have to do it all over again and have to go through the same process and feel quite uncomfortable while doing it.’ (P1) ‘They obviously don’t give you a brace because they’re like, “Your curve is so big it’s not going to work.” They basically give up on you and say, “You’ve got to wait for surgery,” (P10) ‘Much better about my back shape, because it was getting to a point before my operation, and the curve was getting worse and worse, and I was getting shorter and shorter. So now I feel a lot better. I feel a lot more upright, like I stand a lot taller.’ (P2)

Consolidated criteria for reporting qualitative studies (COREQ): 32-item checklist (Chapter 4)

No.	Item	Guide questions/description	Page number
Domain 1: Research team and reflexivity			
Personal Characteristics			
1.	Interviewer/facilitator	Which author/s conducted the interview or focus group?	177
2.	Credentials	What were the researcher's credentials? *E.g., PhD, MD*	177
3.	Occupation	What was their occupation at the time of the study?	177
4.	Gender	Was the researcher male or female?	177
5.	Experience and training	What experience or training did the researcher have?	177
Relationship with participants			
6.	Relationship established	Was a relationship established prior to study commencement?	177
7.	Participant knowledge of the interviewer	What did the participants know about the researcher? e*.g., personal goals, reasons for doing the research*	177
8.	Interviewer characteristics	What characteristics were reported about the interviewer/facilitator? e.g., Bias, assumptions, reasons, and interests in the research topic	177
Domain 2: study design			
Theoretical framework			
9.	Methodological orientation and Theory	What methodological orientation was stated to underpin the study? *e.g., grounded theory, discourse analysis, ethnography, phenomenology, content analysis*	174
Participant selection			
10.	Sampling	How were participants selected? *e.g., purposive, convenience, consecutive, snowball*	174

11.	Method of approach	How were participants approached? e.*g., face-to-face, telephone, mail, email*	175
12.	Sample size	How many participants were in the study?	179
13.	Non-participation	How many people refused to participate or dropped out? Reasons?	179
Setting			
14.	Setting of data collection	Where was the data collected? e.*g., home, clinic, workplace*	175
15.	Presence of non-participants	Was anyone else present besides the participants and researchers?	177
16.	Description of sample	What are the important characteristics of the sample? *e.g., demographic data, date*	180-181
Data collection			
17.	Interview guide	Were questions, prompts, guides provided by the authors? Was it pilot tested?	175
18.	Repeat interviews	Were repeat interviews carried out? If yes, how many?	No
19.	Audio/visual recording	Did the research use audio or visual recording to collect the data?	177
20.	Field notes	Were field notes made during and/or after the interview or focus group?	177
21.	Duration	What was the duration of the interviews or focus group?	177
22.	Data saturation	Was data saturation discussed?	179
23.	Transcripts returned	Were transcripts returned to participants for comment and/or correction?	177

Domain 3: analysis and findings			
Data analysis			
24.	Number of data coders	How many data coders coded the data?	179
25.	Description of the coding tree	Did authors provide a description of the coding tree?	182
26.	Derivation of themes	Were themes identified in advance or derived from the data?	182
27.	Software	What software, if applicable, was used to manage the data?	NA
28.	Participant checking	Did participants provide feedback on the findings?	178
Reporting			
29.	Quotations presented	Were participant quotations presented to illustrate the themes / findings? Was each quotation identified? e.*g., participant number*	183
30.	Data and findings consistent	Was there consistency between the data presented and the findings?	183-193
31.	Clarity of major themes	Were major themes clearly presented in the findings?	183-193
32.	Clarity of minor themes	Is there a description of diverse cases or discussion of minor themes?	183-193

Tong A, Sainsbury P, Craig J. (2007) Consolidated criteria for reporting qualitative research (COREQ): a32--item checklist for interviews and focus groups. International Journal for Quality in Healthcare: 19:349 –357

Appendix 9. Ethical Review after amendment, Participants Information Sheets, Consent Forms, consent to contact form, Topic guide, amendment, Saturation table, and COREQ checklist for *Chapter six*

NHS
The Royal
Orthopaedic Hospital
NHS Foundation Trust

Our ref: EL/PB/ROH21ORTH04

20 July 2022

Prof. Adrian Gardner
Orthopaedic Spinal Consultant
The Royal Orthopaedic Hospital NHS Foundation Trust
Bristol Road South
Northfield
B31 2AP

Dear Adrian,

Research Project: Content validity of the Scoliosis Research Society questionnaire-22r (SRS-22r) for Adolescents with Idiopathic Scoliosis: a qualitative interview study exploring patient's and practitioner's perspectives.

R&D Project No: ROH21ORTH04

The above research project has been re-reviewed by the Trust through the Research & Development Department in light of the Non-substantial Amendment 06 dated 12 July 2022.

All documents have been received the confirmation of amendment assessment email from the Health Research Authority.

New Version of Documents approved:

Document	Version	Date
Topic Guide	V3	11 July 2022
PIS HCP	V5	11 July 2022
Consent Form HCP	V4	11 July 2022

Summary of Amendment (not exhaustive):

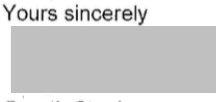
- The content validity study for the SRS-22r was planned to include both semi structured interviews with individuals with AIS and Focus group discussion with health care professionals (HCPs) who are dealing with AIS. The aim of the focus group is to understand the perspective of HCPs to the barriers and facilitators to use OM as well as assess the content validity of the SRS-22r from their HCPs perspectives. The researcher tried her best to plan and recruit HCPs including surgeons, physiotherapists and nurses to focus group discussion. However, it was challenging and had poor responses to find a convenient time that suits all of professions. Thus, we are requesting an amendment to change the methodology of collecting data from HCPs to be individual interview rather than Focus group discussion

The original Trust approval letter has also been reviewed and the R&D Department can confirm that the above non-substantial amendment has been approved and should be read in conjunction with terms and conditions of the original Trust approval letter.

On behalf of the Research & Development Department, I would like to wish you every cohtinued success with your research project.

Best wishes

Yours sincerely



Gareth Stephens
Head of Research, Audit and Development
Royal Orthopaedic Hospital

cc. Samia Alamrani, UoB

<*To be printed on NHS Trust Headed paper – delete this text on completion.*

UNIVERSITY OF BIRMINGHAM

Participant Information Sheet – Health Care Professionals

Study title: Content validity of the Scoliosis Research Society questionnaire-22r (SRS-22r) for Adolescents with Idiopathic Scoliosis: a qualitative interview study exploring patient's and practitioner's perspectives

Invitation paragraph
We would like to invite you to take part in a research study being conducted by the University of Birmingham and sponsored by the Royal Orthopaedic Hospital NHS Foundation Trust, who are the data controller for the project.

Before you decide, it is important for you to understand why the research is being conducted and what it would involve for you. One of the research team will go through the information sheet with you and answer any questions that you have. Please ask if anything is not clear or if you would like more information.

What is the purpose of the study?
Adolescent idiopathic scoliosis (AIS) is deformity of the spine in children age 10-18 years old. It can affect children quality of life in different aspects. These effects should be evaluated using a questionnaire that it's designed to this unique age group.

The Scoliosis Research Society questionnaire-22r (SRS-22r), is the widely used questionnaire for scoliosis patients. To ensure that the results obtained from the SRS-22r questionnaire are accurate and can be trusted, it should have adequate measurement properties such as content validity. Content validity means that the questionnaire items is measuring what is supposed to measure and its relevant to the age and language of targeted population. This can be assessed by interviewing both individuals with scoliosis and health professionals to understand their perspective on the questionnaire items. This information will be analysed, compared and matched to the content of SRS-22r to assess it suitability to AIS.

Furthermore, the results of our systematic review indicated that a there is a tendency in literature to use Patient Reported Outcome Measure (PROM) rather than Performance-Based Outcome Measure (PBOM). Understanding a health professional's barriers and facilitators to the use of specific outcome measures (OM) for AIS, is important to enhance evidence-based practice. So, the purpose of this study is to test the content validity of SRS-22r for AIS. Also, to explore health professionals' perceptions of using different types of OM, such as PBOM, in AIS.

Why have I been invited?
You have been invited to take part because you are a health care professional with an experience in scoliosis research or treatment working in the Royal Orthopaedic Hospital, Birmingham, United Kingdom.

Do I have to take part?
It is up to you to decide whether you would like to take part or not. Any decision regarding your participation will be confidential between you and the research team. If you do decide to take part, you will be given this information sheet to keep and will be asked to sign a consent form.

What will happen to me if I take part?
You need to attend one focus groups which will be arranged at a convenient date and time. It will be conducted virtually via Zoom/Microsoft teams. If you agree to take part, you would be involved in a focus group discussion with other health care professionals (physiotherapists, spinal surgeons, and nurses), facilitated by a member of the research team. The focus group will include a short introduction to the research team and how the focus group will be conducted.
The main discussion will last around 60-90 minutes, and it will be consisting of 2 parts. The first part is around the content validity of the SRS-22r, in which you will discuss with the group and give your feedback about the contents of SRS-22r including the instructions, questions, and response options and recall periods.
The second part in which you will chat with the group about the OM used in individuals with AIS and the barriers and facilitators to select OM in AIS.
During the focus group you will be encouraged to talk freely and openly with colleagues about your understanding as health care professional of using OM and dealing with individuals with AIS.
The focus group will be video recorded. The audio recordings will then be transcribed using a University approved external transcription service. You will not be identifiable in the transcripts or audio recordings.

What are the possible disadvantages of taking part?
There are no known risks in taking part in this study. Sometimes a focus group discussion can be challenging, however all discussions within the focus group are confidential and all participants are reminded of this prior to the focus group commencing.

What are the possible benefits of taking part?
We hope that taking part in the focus group is interesting and provide an evidence on the content validity of the SRS-22r as well as enhance the knowledge about using outcome measure in AIS. You will have the opportunity to receive feedback of the focus groups in the form of a short report which can be requested via the chief investigator (Samia Alamrani).

Expenses and Payments
Since we are collecting data via remote means, there are no expected costs to the participants.

What will happen when the research stops?
After the focus groups have ended, all information collected during the study will be stored in an electronic format on a secure web-based system called Redcap. Once the transcriptions of the interviews have been reviewed, the video/audio recordings will be destroyed. Data will be stored at the University of Birmingham for ten years following the study. After that period, all information will be destroyed.

What will happen if I don't want to carry on with the study?
If you decide you do not wish to carry on with the study, you are free to withdraw at any time prior to the focus group commencing, without having to give a reason. After the focus group takes place, you will be given the opportunity to review the transcript and make small amendments where appropriate for accuracy.
After this point, data analysis will commence, and removal of individual data will not be possible.

What if there is a problem?
It is unlikely there will be any problems during the study. If you have a concern about any aspect of the study, please contact the chief investigator, Samia Alamrani who will do her best to answer any questions. Alternatively, you can contact the Sponsor's point of contact Carolyn Langford, R&D Manager at the Royal Orthopaedic Hospital, as an independent representative.

How will we use information about you?
We will need to use information from you and the focus group for this research project. This information will include transcripts of the focus groups, your name and contact details to organise the focus groups. People will use this information to do the research or to check your records to make sure that the research is being done properly. People who do not need to know who you are will not be able to see your name or contact details. We will keep all information about you safe and secure.

All information that is collected about you in this study will be kept confidential in accordance with the Data Protection Act 2018 and General Data Protection Regulation (GDPR). Your contact details will be kept on a password protected computer and only accessible by the research team. Your data will have a code number instead. Personal data will be kept/accessed after the study has ended for a period of 3-6 months so that we can send you a copy of the study findings if you wish to receive this. After that all personal data will be destroyed.

Once we have finished the study, we will keep some of the data so we can check the results. We will write out reports in a way that no-one can work out that you took part in that study.

What are your choices about how your information is used?
After the focus group completed, we will not be able to remove individual data. We need to manage your records in specific ways for the research to be reliable. This means that we will not be able to let you see the data or change the data we hold about you.

Where can you find out more about how your information is used?
You can find out more about how we use your information at www.hra.nhs.uk/information-about-patients/, by asking one of the research team, contacting Samia Alamrani (details below) or University of Birmingham's data protection officer dataprotection@contacts.bham.ac.uk.

What will happen to the results of the study?
The results of this study will provide evidence on the content validity of the SRS-22r. Also, it will inform the feasibility of using different types of outcome measure in individuals with AIS.

The results will be disseminated in scientific journals and presentations at research conferences. Following completion of the study, Samia will send you a short summary of the study results.

Who is organising and funding this study?
The research study is being undertaken as an educational project. It is organized by researchers from the University of Birmingham and sponsored by The Royal Orthopaedic Hospital NHS Foundation Trust

Who has reviewed the study?
This study has been reviewed by members of the research team with experience and expertise in musculoskeletal physiotherapy and spinal surgery as well as members of the public. All research

which is conducted within the NHS is reviewed by an independent panel by the Research Ethics Committee, along with the Independent Scientific Advisory Committee of the Royal Orthopaedic Hospital.

Contacts for further information or any questions about this study:

Mrs. Samia Alamrani, Principle Investigator, University of Birmingham
Tel:
Email:

Mr. Adrian Gardner, Chief Investigator. The Royal Orthopaedic Hospital NHS Foundation Trust
Tel:
Email:

Dr Nicola Heneghan, Academic Supervisor, University of Birmingham
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Thank you for taking the time to read this information sheet.

<To be printed on NHS Trust headed paper – delete this text on completion>

UNIVERSITY OF BIRMINGHAM

CONSENT FORM HEALTH CARE PROFESSIONAL

Title of Project: Content validity of the Scoliosis Research Society questionnaire-22r (SRS-22r) for Adolescents with Idiopathic Scoliosis : a qualitative interview study exploring patient's and practitioner's perspectives

Participant's Name: _____

Please initial

1.	I confirm that I have read and understand the Participant Information Sheet, dated 21st April 2021 (version 3), for the above study. I have had the opportunity to consider the study information, ask questions and have had these answered to my satisfaction.	☐
2.	I understand that my participation is voluntary and that I am free to withdraw prior to the focus group taking place without giving any reason.	☐
3.	I agree to the focus group being video recorded. I understand that audio recording will be shared with a transcription service provider with whom contractual agreements are in place. I agree to my data being shared securely with the transcription services for the purpose of this study.	☐
4.	I understand that all research data will be kept confidential and anonymised, and securely stored on servers at the University of Birmingham for a period of 10 years, which will only be accessible to designated researchers.	☐
5.	I understand that my personal data will be kept confidential on servers at the University of Birmingham, which will only be accessible to designated researchers in the University of Birmingham and the Royal Orthopaedic Hospital.	☐
6.	I understand that my identifiable data may be looked at by authorised representatives of the Sponsor organisation, NHS Trust or regulatory bodies where appropriate to check the conduct of the study. I agree to these individuals accessing my data.	☐
7.	I agree to the use of anonymised quotes made by me in future publications from this research study.	☐
8.	I agree to take part in the above study.	☐
9.	Optional: I agree to receive a summary of the study results.	☐

_____	_____	_____
Name of Participant	Date	Signature
_____	_____	_____
Name of person taking consent	Date	Signature

When completed: 1 copy for Participant
1 (original) copy for participating site investigators site file

Topic guide (Health Care Professionals)

Research Aim	Content validity of the Scoliosis Research Society Questionnaire		
Interview Section	Questions/Content	Prompts	Aims
Ethics Statement and ground rules	• Welcome to this focus group discussion, my name is Nicola Heneghan and assisting me is Samia. We're both from the University of Birmingham. • We realize you are busy, and we appreciate your time coming to this focus group. • This focus group is structured in two parts. First, we want to explore clinical use outcome measure (OM) for Adolescent with Idiopathic Scoliosis, including any thoughts on barriers and enablers. Then, in the second part we want to assess the content validity of the SRS-22r. • You were selected because you have a clinical or research experience in managing individuals with AIS. • Please feel free to express your opinion and views clearly and in detail one person at a time. • There are no right or wrong answers; we are all here to share. • Just a reminder that this session is being audio/video recorded. • All information shared will be kept confidential and anonymised. Your names will not be used for the analysis of the discussion. • Before we start is any of you have any further questions?	• *Can I confirm that each one of you have read and understand the information sheet and signed the consent form?* • *Is this okay for everyone?"*	• To ensure full understanding of what is expected of the participant during this focus group. • Make sure each participant is comfortable and ready to begin.

Introductory Questions	• Would everybody please take turns to introduce themselves by giving your name, and briefly share with the group your clinical/research experiences.	• *Years of experiences with AIS, clinical posts, setting.* • *Frequency of dealing/treating individuals with AIS.*	• Make participants relax and feel comfortable with talking and opening up. • Build rapport. • To have an insight into each participant's background.
Transition Questions	• Talking about outcome measures for individuals with AIS, could you each share with the group which questionnaire or OM you use in practice for AIS?	• *What type of OM you are using? Why?* • *How often and when do you use OM?*	• Start to guide the interview towards experiences of HCPs with OM. • To get an idea of HCPs approach and preferences to use OM for AIS.
Background of the study	• As you have seen in the participants information sheet, the aim of this group discussion is first to discuss the barriers and facilitators to use outcome measure (OM) for adolescents with idiopathic scoliosis and the second part is the cognitive debriefing for the SRS-22r to test its content validity. • This study is part from Samia's PhD study. Her Systematic Review on the physical functioning OM for AIS was published on Spine. • We performed two searched-strategy on all types of OM used for physical functioning assessment for AIS. • We identified a variety of OM, however, most of studies of measurement properties were evaluating PROMs, with paucity of information on PBOM, and body structure and function measures. Construct validity, reliability and responsiveness of most PROMs is established in AIS, but not content validity or internal consistency. • So we have chosen the SRS-22r to test its content validity and we want you to give us your thoughts about the content of the SRS-22r. If it is clear, relevant and comprehensive.	• *Do you have any questions?* • *If you are unsure at any point regarding the study, please ask.*	• Inform the participant of the background of the study. • Ensure the participant knows they can ask questions if they are unsure.

1st part feasibility	• To start with, could everyone tell us what influences your choice of an OM when managing individuals with AIS?	• *When you decide to choose OM/questionnaire what do you look for ?* • *What about the characteristics of OM?* • *How the characteristics of individuals with AIS influences your choice of OM?* • *Could everyone perhaps tell us what you think is the most important factor that influence you to select OM for individuals with AIS?* • *Thinking about the time constrains? How it affects your choice of OM?*	• To explore factors that affect the selection of OM for individuals with AIS from HCPs perspective.
	• From your point of view, how do different types of OMs other than PROMs be developed to enable its use in OM?	• *Could you think of any example of OM? Consider performance and patient reported measures separately* • *Have you use any experience to use performance-based OM?* • *How do you think performance-based measures are different from other OMs such as PROMs?* • *What could be done to enhance the use of performance-based measures in AIS?*	• To understand HCPs thoughts about using of performance based measures in AIS. • To explore the perceptions and thoughts of HCPs of facilitators to enhance the use OM in AIS
	• What do you think are the limitations/barriers to using different types of OM in individuals with AIS?	• *Did the time constrain has an effect on your choice of OM? how?* • *Could you think of other barriers? Recourses available?*	• To explore the perceptions and thoughts of HCPs of barriers to use OM in AIS.
	• Finally, is there anything connected to OMs used in AIS we haven't talked about and you think it's useful to bring it up now.		
2nd part Cognitive debriefing	• This is the second part from our focus group discussion. We want to discuss if you think the SRS-22r questionnaire (question style, language, topic) is suitable for AIS. • I will read SRS-22r questions one by one to you. Then, please could you tell me, what do you think about the question.		• To test the relevance of SRS-22r items to individuals with AIS • To assess if the is SRS-22r is comprehensive. • To test if the SRS-22r response options is clear and appropriate. • To test if the recall period is appropriate.

	1. Which one of the following best describes the amount of pain you have experienced during the past 6 months? -Response options are: None, Mild, Moderate, Moderate to severe, Severe.	• *Do you think this question is relevant and Important to AIS? why?* • *What is your thought about the response options ? are they appropriate to AIS?* • *Do you think the recall period is appropriate?*	
	2. Which one of the following best describes the amount of pain you have experienced over the last month? -Response options are: None, Mild, Moderate, Moderate to severe, Severe	• *Do you think this question is relevant and Important to AIS? why?* • *What is your thought about the response options ? are they appropriate to AIS? what about the word "severe" ? is it appropriate to AIS?* • *Do you think the recall period is appropriate?*	
	3. During the past 6 months have you been a very nervous person? -Response options are: None of the time, A little of the time, Some of the time, Most of the time, All of the time.	• *Is this question relevant and Important to AIS? why?* • *What do you think about the response options ? are they appropriate to AIS?* • *What about the recall period is appropriate?*	
	4. If you had to spend the rest of your life with your back shape as it is right now, how would you feel about it? - Response options are : Very happy, Somewhat happy Neither happy nor unhappy Somewhat unhappy, Very unhappy	• *Is this question relevant and appropriate to AIS? why?* • *What do you think about the response options ? are they appropriate to AIS?*	
	5. What is your current level of activity? Response options are : Bedridden, Primarily no activity, Light labor and light sports, Moderate labor and moderate sports, Full activities without restriction.	• *Is this question relevant and Important to AIS? why?* • *What do you think about the response options ?* • *What are your thoughts about these words "labor", "Bedridden", "restrictions"? are they clear and appropriate to AIS?*	
	6. How do you look in clothes? - Response options are : Very good, Good, Fair, Bad, Very bad	• *Do you think this question is relevant and Important to AIS? why?* • *What is your thought about the response options ? are they appropriate to AIS?*	

	7. In the past 6 months have you felt so down in the dumps that nothing could cheer you up? -Response options are :Very often, Often, Sometimes Rarely, Never	• *Do you think this question is relevant and Important to AIS? why?* • *What is your thought about (so down in dumps) is it appropriate to AIS?* • *What is your thought about the response options ? are they appropriate to AIS?* • *Do you think the recall period is appropriate?*	
	8. Do you experience back pain when at rest? - Response options are : Very often, Often, Sometimes, Rarely, Never	• *Do you think this question is relevant and Important to AIS? why?* • *What is your thought about the response options ? are they appropriate to AIS?*	
	9. What is your current level of work/school activity? - Response options are : 100% normal, 75%normal 50% normal, 25% normal, 0% normal	• *Do you think this question is relevant and Important to AIS? why?* • *What is your thought about the response options ? are they appropriate to AIS?*	
	10. Which of the following best describes the appearance of your trunk; defined as the human body except for the head and extremities? - Response options are : Very good, Good, Fair, Poor, Very Poor	• *Do you think this question is relevant and Important to AIS? why?* • *What is your thought about the response options ? are they appropriate to AIS?*	
	11. Which one of the following best describes your pain medication use for back pain? -Response options are : None, Non-narcotics, weekly or less (e.g., Aspirin, Paracetamol , Ibuprofen) , Non-narcotics daily, Narcotics weekly or less (e.g., Co-codamol), Narcotics daily.	• *Do you think this question is relevant and Important to AIS? why?* • *What is your thought about the response options ? are they appropriate to AIS?* • *What is your thought about the word "Non-narcotics" ?*	
	12. Does your back limit your ability to do things around the house? -Response options are: Never, Rarely, Sometimes Often, Very Often	• *Do you think this question is relevant and Important to AIS? why?* • *What is your thought about the response options ? are they appropriate to AIS?*	

	13. Have you felt calm and peaceful during the past 6 months? -Response options are: All of the time, Most of the time, Some of the time, A little of the time, None of the time.	• *Do you think this question is relevant and Important to AIS? why?* • *What is your thought about the response options ? are they appropriate to AIS?* • *Do you think the recall period is appropriate?*	
	14. Do you feel that your back condition affects your personal relationships? -Response options are: None, Slightly, Mildly, Moderately, Severely	• *Is this question relevant and Important to AIS? why?* • *What do you think about the response options ?*	
	15. Are you and/or your family experiencing financial difficulties because of your back? - Response options are: Severely, Moderately, Mildly Slightly, None.	• *Is this question relevant and Important to AIS? why?* • *Are response options appropriate to AIS?*	
	16. In the past 6 months have you felt down hearted and blue? -Response options are: Never Rarely, Sometimes, Often, Very often	• *Is this question relevant and Important to AIS? why?* • *Are response options appropriate to AIS? what about word , "down hearted', "blue"?* • *Do you think the recall period is appropriate?*	
	17. In the last 3 months have you taken any days off of work, including household work, or school because of back pain? -Response options are : 0 days 1 day,2 days,3 days, 4 or more days.	• *Is this question relevant and Important to AIS? why?* • *Are response options appropriate to AIS?* • *Do you think the recall period is appropriate?*	
	18. Does your back condition limit your going out with friends/family? -Response options are : Never, Rarely, Sometimes, Often, Very often	• *Is this question relevant and Important to AIS? why?* • *Are response options appropriate to AIS?*	
	19. Do you feel attractive with your current back condition? -Response options are : Yes, very Yes, somewhat, Neither attractive nor unattractive No, not very much No, not at all	• *Is this question relevant and Important to AIS? why?* • *Are response options appropriate to AIS?*	

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UNIVERSITY OF BIRMINGHAM

Participant Information Sheet – Health Care Professionals

Study title: Content validity of the Scoliosis Research Society questionnaire-22r (SRS-22r) for Adolescents with Idiopathic Scoliosis: a qualitative interview study exploring patient's and practitioner's perspectives

Invitation paragraph
We would like to invite you to take part in a research study being conducted by the University of Birmingham and sponsored by the Royal Orthopaedic Hospital NHS Foundation Trust, who are the data controller for the project.

Before you decide, it is important for you to understand why the research is being conducted and what it would involve for you. One of the research team will go through the information sheet with you and answer any questions that you have. Please ask if anything is not clear or if you would like more information.

What is the purpose of the study?
Adolescent idiopathic scoliosis (AIS) is deformity of the spine in children age 10-18 years old. It can affect children quality of life in different aspects. These effects should be evaluated using a questionnaire that it's designed to this unique age group.

The Scoliosis Research Society questionnaire-22r (SRS-22r), is the widely used questionnaire for scoliosis patients. To ensure that the results obtained from the SRS-22r questionnaire are accurate and can be trusted, it should have adequate measurement properties such as content validity. Content validity means that the questionnaire items is measuring what is supposed to measure and its relevant to the age and language of targeted population. This can be assessed by interviewing both individuals with scoliosis and health professionals to understand their perspective on the questionnaire items. This information will be analysed, compared and matched to the content of SRS-22r to assess it suitability to AIS.

Furthermore, the results of our systematic review indicated that a there is a tendency in literature to use Patient Reported Outcome Measure (PROM) rather than Performance-Based Outcome Measure (PBOM). Understanding a health professional's barriers and facilitators to the use of specific outcome measures (OM) for AIS, is important to enhance evidence-based practice. So, the purpose of this study is to test the content validity of SRS-22r for AIS. Also, to explore health professionals' perceptions of using different types of OM, such as PBOM, in AIS.

Why have I been invited?
You have been invited to take part because you are a health care professional with an experience in scoliosis research or treatment working in the Royal Orthopaedic Hospital, Birmingham, United Kingdom.

Do I have to take part?
It is up to you to decide whether you would like to take part or not. Any decision regarding your participation will be confidential between you and the research team. If you do decide to take part, you will be given this information sheet to keep and will be asked to sign a consent form.

What will happen to me if I take part?

You need to attend one semi-structured interview which will be arranged at a convenient date and time. It will be conducted virtually via Zoom/Microsoft teams. If you agree to take part, you would be involved in a 60–90-minute interview with the researcher. The interview will include a short introduction to the research and how the interview will be conducted.

The main discussion will last around 60-90 minutes, and it will be consisting of 2 parts. The first part is around the content validity of the SRS-22r, in which you will discuss and give your feedback about the contents of SRS-22r including the instructions, questions, and response options and recall periods. The second part in which you will chat about the OM used in individuals with AIS and the barriers and facilitators to select OM in AIS.

During the interview you will be encouraged to talk freely and openly about your understanding as health care professional of using OM and dealing with individuals with AIS.

The interview will be video recorded. The audio recordings will then be transcribed using a University approved external transcription service. You will not be identifiable in the transcripts or audio recordings.

What are the possible disadvantages of taking part?

There are no known risks in taking part in this study. Sometimes a focus group discussion can be challenging, however all discussions within the focus group are confidential and all participants are reminded of this prior to the focus group commencing.

What are the possible benefits of taking part?

We hope that taking part in the interview is interesting and provide evidence on the content validity of the SRS-22r as well as enhance the knowledge about using outcome measure in AIS. You will have the opportunity to receive feedback of the interview in the form of a short report which can be requested via the chief investigator (Samia Alamrani).

Expenses and Payments

Since we are collecting data via remote means, there are no expected costs to the participants.

What will happen when the research stops?

After the focus groups have ended, all information collected during the study will be stored in an electronic format on a secure web-based system called Redcap. Once the transcriptions of the interviews have been reviewed, the video/audio recordings will be destroyed. Data will be stored at the University of Birmingham for ten years following the study. After that period, all information will be destroyed.

What will happen if I don't want to carry on with the study?

If you decide you do not wish to carry on with the study, you are free to withdraw at any time prior to the focus group commencing, without having to give a reason. After the focus group takes place, you will be given the opportunity to review the transcript and make small amendments where appropriate for accuracy.

After this point, data analysis will commence, and removal of individual data will not be possible.

What if there is a problem?

It is unlikely there will be any problems during the study. If you have a concern about any aspect of the study, please contact the chief investigator, Samia Alamrani who will do her best to answer any questions. Alternatively, you can contact the Sponsor's point of contact Carolyn Langford, R&D Manager at the Royal Orthopaedic Hospital, as an independent representative.

How will we use information about you?
We will need to use information from you and the focus group for this research project. This information will include transcripts of the focus groups, your name and contact details to organise the focus groups. People will use this information to do the research or to check your records to make sure that the research is being done properly. People who do not need to know who you are will not be able to see your name or contact details. We will keep all information about you safe and secure.

All information that is collected about you in this study will be kept confidential in accordance with the Data Protection Act 2018 and General Data Protection Regulation (GDPR). Your contact details will be kept on a password protected computer and only accessible by the research team. Your data will have a code number instead. Personal data will be kept/accessed after the study has ended for a period of 3-6 months so that we can send you a copy of the study findings if you wish to receive this. After that all personal data will be destroyed.

Once we have finished the study, we will keep some of the data so we can check the results. We will write out reports in a way that no-one can work out that you took part in that study.

What are your choices about how your information is used?
After the focus group completed, we will not be able to remove individual data. We need to manage your records in specific ways for the research to be reliable. This means that we will not be able to let you see the data or change the data we hold about you.

Where can you find out more about how your information is used?
You can find out more about how we use your information at www.hra.nhs.uk/information-about-patients/, by asking one of the research team, contacting Samia Alamrani (details below) or University of Birmingham's data protection officer dataprotection@contacts.bham.ac.uk.

What will happen to the results of the study?
The results of this study will provide evidence on the content validity of the SRS-22r. Also, it will inform the feasibility of using different types of outcome measure in individuals with AIS.

The results will be disseminated in scientific journals and presentations at research conferences. Following completion of the study, Samia will send you a short summary of the study results.

Who is organising and funding this study?
The research study is being undertaken as an educational project. It is organized by researchers from the University of Birmingham and sponsored by The Royal Orthopaedic Hospital NHS Foundation Trust

Who has reviewed the study?
This study has been reviewed by members of the research team with experience and expertise in musculoskeletal physiotherapy and spinal surgery as well as members of the public. All research which is conducted within the NHS is reviewed by an independent panel by the Research Ethics

Committee, along with the Independent Scientific Advisory Committee of the Royal Orthopaedic Hospital.

Contacts for further information or any questions about this study:

Mrs. Samia Alamrani, Principle Investigator, University of Birmingham
Tel:
Email:

Mr. Adrian Gardner, Chief Investigator. The Royal Orthopaedic Hospital NHS Foundation Trust
Tel:
Email:

Dr Nicola Heneghan, Academic Supervisor, University of Birmingham
Tel:
Email:

Patient Advice and Liaison Service, The Royal Orthopaedic Hospital NHS Foundation Trust
Tel:
Email: roh-tr.pals@nhs.net

Thank you for taking the time to read this information sheet.

<*To be printed on NHS Trust Headed paper – delete this text on completion.*

UNIVERSITY OF BIRMINGHAM

CONSENT TO CONTACT FORM

Title of Project: Content validity of the Scoliosis Research Society questionnaire-22r (SRS-22r) for Adolescents with Idiopathic Scoliosis

		Please initial
1.	I confirm that my name, phone number and address will be shared with a member of the research team located at the University of Birmingham.	
2.	I understand that the above-mentioned details will be shared through NHS secured email.	
3.	I understand that the member of the research team will contact me with more information about this study, after which I can decide whether I would like to participate.	

_____ _____ _____

Name of person taking consent | Date | Signature

When completed
1 copy for participant
1 original copy of participating site investigators site files

<To be printed on NHS Trust headed paper – delete this text on completion>

UNIVERSITY OF BIRMINGHAM

CONSENT FORM HEALTH CARE PROFESSIONAL

Title of Project: Content validity of the Scoliosis Research Society questionnaire-22r (SRS-22r) for Adolescents with Idiopathic Scoliosis : a qualitative interview study exploring patient's and practitioner's perspectives

Participant's Name: _____

Please initial

1. I confirm that I have read and understand the Participant Information Sheet, dated 11 July 2022 (version 3), for the above study. I have had the opportunity to consider the study information, ask questions and have had these answered to my satisfaction. ☐
2. I understand that my participation is voluntary and that I am free to withdraw prior to the interview taking place without giving any reason. ☐
3. I agree to the interview being video recorded. I understand that audio recording will be shared with a transcription service provider with whom contractual agreements are in place. I agree to my data being shared securely with the transcription services for the purpose of this study. ☐
4. I understand that all research data will be kept confidential and anonymised, and securely stored on servers at the University of Birmingham for a period of 10 years, which will only be accessible to designated researchers. ☐
5. I understand that my personal data will be kept confidential on servers at the University of Birmingham, which will only be accessible to designated researchers in the University of Birmingham and the Royal Orthopaedic Hospital. ☐
6. I understand that my identifiable data may be looked at by authorised representatives of the Sponsor organisation, NHS Trust or regulatory bodies where appropriate to check the conduct of the study. I agree to these individuals accessing my data. ☐
7. I agree to the use of anonymised quotes made by me in future publications from this research study. ☐
8. I agree to take part in the above study. ☐
9. Optional: I agree to receive a summary of the study results. ☐

_____	_____	_____
Name of Participant	Date	Signature
_____	_____	_____
Name of person taking consent	Date	Signature

When completed: 1 copy for Participant
1 (original) copy for participating site investigators site file

Topic guide (Health Care Professionals)

Research Aim	Content validity of the Scoliosis Research Society Questionnaire		
Interview Section	Questions/Content	Prompts	Aims
Ethics Statement and ground rules	• Firstly, I would like to thank you for participating in this interview. My name is {Samia}, a PhD student from University of Birmingham. • I realize that you are busy, and I appreciate your time to participate in this study. • This interview is structured in two parts. First, we want to explore clinical use outcome measure (OM) for Adolescent with Idiopathic Scoliosis, including any thoughts on barriers and enablers. Then, in the second part we want to assess the content validity of the SRS-22r. • You were selected because you have a clinical or research experience in managing individuals with AIS. • Please feel free to express your opinion and views clearly and in detail. • There are no right or wrong answers; I am interested in your thoughts and experience. • Just a reminder that this interview is being audio/video recorded. • All information shared will be kept confidential and anonymised. Your name will not be used for the analysis and will be replaced by a code number. • Before we start do you have any questions?	• *Can I confirm that you have read and understand the information sheet and signed the consent form?* • *Is this okay for you?"*	• To ensure full understanding of what is expected of the participant during interview • Make sure that participant is comfortable and ready to begin.

Introductory Questions	• Would you please share with me your clinical/research experiences.	• *Years of experiences with AIS, clinical posts, setting.* • *Frequency of dealing/treating individuals with AIS.*	• Make participants relax and feel comfortable with talking and opening up. • Build rapport. • To have an insight into each participant's background.
Transition Questions	• Talking about outcome measures for individuals with AIS, could you please tell which questionnaire or OM you use in practice for AIS?	• *What type of OM you are using? Why?* • *How often you use OM?* • *When do you use OM?*	• Start to guide the interview towards experiences of HCPs with OM. • To get an idea of HCPs approach and preferences to use OM for AIS.
Background of the study	• As you have seen in the participants information sheet, the aim of this interview is first to discuss the barriers and facilitators to use outcome measure (OM) for adolescents with idiopathic scoliosis and the second part is the cognitive debriefing for the SRS-22r to test its content validity. • This study is part from my PhD study. Our systematic review on the physical functioning OM for AIS was published on Spine. • We performed two searched-strategy on all types of OM used for physical functioning assessment for AIS. • We identified a variety of OM, however, most of studies of measurement properties were evaluating PROMs, with paucity of information on PBOM, and body structure and function measures. Construct validity, reliability and responsiveness of most PROMs is established in AIS, but not content validity or internal consistency. • So we have chosen the SRS-22r to test its content validity and we want you to give us your thoughts about the content of the SRS-22r. If it is clear, relevant and comprehensive.	• *Do you have any questions?* • *If you are unsure at any point regarding the study, please ask.*	• Inform the participant of the background of the study. • Ensure the participant knows they can ask questions if they are unsure.

1st part feasibility	• To start with, could you tell me what influences your choice of an OM when managing individuals with AIS?	• *When you decide to choose OM/questionnaire what do you look for ?* • *What about the characteristics of OM?* • *How the characteristics of individuals with AIS influences your choice of OM?* • *Could you perhaps tell me what you think is the most important factor that influence you to select OM for individuals with AIS?* • *Thinking about the time constrains? How it affects your choice of OM?*	• To explore factors that affect the selection of OM for individuals with AIS from HCPs perspective.
	• From your point of view, how do different types of OMs other than PROMs be developed to enable its use in OM?	• *Could you think of any example of OM? Consider performance and patient reported measures separately* • *Have you use any experience to use performance-based OM?* • *How do you think performance-based measures are different from other OMs such as PROMs?* • *What could be done to enhance the use of performance-based measures in AIS?*	• To understand HCPs thoughts about using of performance based measures in AIS. • To explore the perceptions and thoughts of HCPs of facilitators to enhance the use OM in AIS
	• What do you think are the limitations/barriers to using different types of OM in individuals with AIS?	• *How the time constrain has an effect on your choice of OM?* • *Could you think of other barriers?* • *Recourses available?*	• To explore the perceptions and thoughts of HCPs of barriers to use OM in AIS.
	• Finally, is there anything connected to OMs used in AIS we haven't talked about and you think it's useful to bring it up now.		
2nd part Cognitive debriefing	• This is the second part from our focus group discussion. We want to discuss if you think the SRS-22r questionnaire (question style, language, topic) is suitable for AIS. • I will read SRS-22r questions one by one to you. Then, please could you tell me, what do you think about the question.		• To test the relevance of SRS-22r items to individuals with AIS • To assess if the is SRS-22r is comprehensive. • To test if the SRS-22r response options is clear and appropriate. • To test if the recall period is appropriate.

	1. Which one of the following best describes the amount of pain you have experienced during the past 6 months? -Response options are: None, Mild, Moderate, Moderate to severe, Severe.	• *Do you think this question is relevant and Important to AIS? why?* • *What is your thought about the response options ? are they appropriate to AIS?* • *Do you think the recall period is appropriate?*	
	2. Which one of the following best describes the amount of pain you have experienced over the last month? -Response options are: None, Mild, Moderate, Moderate to severe, Severe	• *Do you think this question is relevant and Important to AIS? why?* • *What is your thought about the response options ? are they appropriate to AIS? what about the word "severe" ? is it appropriate to AIS?* • *Do you think the recall period is appropriate?*	
	3. During the past 6 months have you been a very nervous person? -Response options are: None of the time, A little of the time, Some of the time, Most of the time, All of the time.	• *Is this question relevant and Important to AIS? why?* • *What do you think about the response options ? are they appropriate to AIS?* • *What about the recall period is appropriate?*	
	4. If you had to spend the rest of your life with your back shape as it is right now, how would you feel about it? - Response options are : Very happy, Somewhat happy Neither happy nor unhappy Somewhat unhappy, Very unhappy	• *Is this question relevant and appropriate to AIS? why?* • *What do you think about the response options ? are they appropriate to AIS?*	
	5. What is your current level of activity? Response options are : Bedridden, Primarily no activity, Light labor and light sports, Moderate labor and moderate sports, Full activities without restriction.	• *Is this question relevant and Important to AIS? why?* • *What do you think about the response options ?* • *What are your thoughts about these words "labor", "Bedridden", "restrictions"? are they clear and appropriate to AIS?*	
	6. How do you look in clothes? - Response options are : Very good, Good, Fair, Bad, Very bad	• *Do you think this question is relevant and Important to AIS? why?* • *What is your thought about the response options ? are they appropriate to AIS?*	

	7. In the past 6 months have you felt so down in the dumps that nothing could cheer you up? -Response options are :Very often, Often, Sometimes Rarely, Never	• *Do you think this question is relevant and Important to AIS? why?* • *What is your thought about (so down in dumps) is it appropriate to AIS?* • *What is your thought about the response options ? are they appropriate to AIS?* • *Do you think the recall period is appropriate?*	
	8. Do you experience back pain when at rest? - Response options are : Very often, Often, Sometimes, Rarely, Never	• *Do you think this question is relevant and Important to AIS? why?* • *What is your thought about the response options ? are they appropriate to AIS?*	
	9. What is your current level of work/school activity? - Response options are : 100% normal, 75%normal 50% normal, 25% normal, 0% normal	• *Do you think this question is relevant and Important to AIS? why?* • *What is your thought about the response options ? are they appropriate to AIS?*	
	10. Which of the following best describes the appearance of your trunk; defined as the human body except for the head and extremities? - Response options are : Very good, Good, Fair, Poor, Very Poor	• *Do you think this question is relevant and Important to AIS? why?* • *What is your thought about the response options ? are they appropriate to AIS?*	
	11. Which one of the following best describes your pain medication use for back pain? -Response options are : None, Non-narcotics, weekly or less (e.g., Aspirin, Paracetamol , Ibuprofen) , Non-narcotics daily, Narcotics weekly or less (e.g., Co-codamol), Narcotics daily.	• *Do you think this question is relevant and Important to AIS? why?* • *What is your thought about the response options ? are they appropriate to AIS?* • *What is your thought about the word "Non-narcotics" ?*	
	12. Does your back limit your ability to do things around the house? -Response options are: Never, Rarely, Sometimes Often, Very Often	• *Do you think this question is relevant and Important to AIS? why?* • *What is your thought about the response options ? are they appropriate to AIS?*	

	13. Have you felt calm and peaceful during the past 6 months? -Response options are: All of the time, Most of the time, Some of the time, A little of the time, None of the time.	• *Do you think this question is relevant and Important to AIS? why?* • *What is your thought about the response options ? are they appropriate to AIS?* • *Do you think the recall period is appropriate?*	
	14. Do you feel that your back condition affects your personal relationships? -Response options are: None, Slightly, Mildly, Moderately, Severely	• *Is this question relevant and Important to AIS? why?* • *What do you think about the response options ?*	
	15. Are you and/or your family experiencing financial difficulties because of your back? - Response options are: Severely, Moderately, Mildly Slightly, None.	• *Is this question relevant and Important to AIS? why?* • *Are response options appropriate to AIS?*	
	16. In the past 6 months have you felt down hearted and blue? -Response options are: Never Rarely, Sometimes, Often, Very often	• *Is this question relevant and Important to AIS? why?* • *Are response options appropriate to AIS? what about word , "down hearted', "blue"?* • *Do you think the recall period is appropriate?*	
	17. In the last 3 months have you taken any days off of work, including household work, or school because of back pain? -Response options are : 0 days 1 day,2 days,3 days, 4 or more days.	• *Is this question relevant and Important to AIS? why?* • *Are response options appropriate to AIS?* • *Do you think the recall period is appropriate?*	
	18. Does your back condition limit your going out with friends/family? -Response options are : Never, Rarely, Sometimes, Often, Very often	• *Is this question relevant and Important to AIS? why?* • *Are response options appropriate to AIS?*	
	19. Do you feel attractive with your current back condition? -Response options are : Yes, very Yes, somewhat, Neither attractive nor unattractive No, not very much No, not at all	• *Is this question relevant and Important to AIS? why?* • *Are response options appropriate to AIS?*	

	20. Have you been a happy person during the past 6 months? -Response options are : None of the time, A little of the time, Some of the time, Most of the time, All of the time	• *Is this question relevant and Important to AIS? why?* • *Are response options appropriate to AIS?* • *Do you think the recall period is appropriate?*	
	21. Are you satisfied with the results of your back management? -Response options are : Very satisfied, Satisfied Neither satisfied nor unsatisfied Unsatisfied, Very unsatisfied	• *Is this question relevant and Important to AIS? why?* • *Are response options appropriate to AIS?*	
	22. Would you have the same management again if you had the same condition? -Response options are : Definitely yes, Probably yes, Not sure Probably not, Definitely not	• *Is this question relevant and Important to AIS? why?* • *Are response options appropriate to AIS?*	
	Finally, Do you think the questionnaire covers all the relevant and important areas to those with AIS ?	• *can you think of another important areas that should be included in the questionnaire?*	• To assess if there are any additional areas need to be covered.
Conclusion	• Thank you for participating. This has been a very useful discussion. • Your opinions will be a valuable addition to the study and my research . • I would like to remind you that any comments featuring in this report will be anonymous	• *Is there anything you would like to ask regarding the analysis of the data or the next steps of the process?*	• Ensure the participant is comfortable with what has been discussed.

Consolidated criteria for reporting qualitative studies (COREQ): 32-item checklist (Chapter 6)

No.	Item	Guide questions/description	Page number
Domain 1: Research team and reflexivity			
Personal Characteristics			
1.	Interviewer/facilitator	Which author/s conducted the interview or focus group?	198
2.	Credentials	What were the researcher's credentials? *E.g., PhD, MD*	198
3.	Occupation	What was their occupation at the time of the study?	198
4.	Gender	Was the researcher male or female?	198
5.	Experience and training	What experience or training did the researcher have?	198
Relationship with participants			
6.	Relationship established	Was a relationship established prior to study commencement?	198
7.	Participant knowledge of the interviewer	What did the participants know about the researcher? e.*g., personal goals, reasons for doing the research*	198
8.	Interviewer characteristics	What characteristics were reported about the interviewer/facilitator? e.g., Bias, assumptions, reasons, and interests in the research topic	198
Domain 2: study design			
Theoretical framework			
9.	Methodological orientation and Theory	What methodological orientation was stated to underpin the study? *e.g., grounded theory, discourse analysis, ethnography, phenomenology, content analysis*	196
Participant selection			
10.	Sampling	How were participants selected? *e.g., purposive, convenience, consecutive, snowball*	195

11.	Method of approach	How were participants approached? e.*g., face-to-face, telephone, mail, email*	195
12.	Sample size	How many participants were in the study?	198
13.	Non-participation	How many people refused to participate or dropped out? Reasons?	198
Setting			
14.	Setting of data collection	Where was the data collected? e.*g., home, clinic, workplace*	195
15.	Presence of non-participants	Was anyone else present besides the participants and researchers?	195
16.	Description of sample	What are the important characteristics of the sample? *e.g., demographic data, date*	198
Data collection			
17.	Interview guide	Were questions, prompts, guides provided by the authors? Was it pilot tested?	195
18.	Repeat interviews	Were repeat interviews carried out? If yes, how many?	NA
19.	Audio/visual recording	Did the research use audio or visual recording to collect the data?	195
20.	Field notes	Were field notes made during and/or after the interview or focus group?	195
21.	Duration	What was the duration of the interviews or focus group?	195
22.	Data saturation	Was data saturation discussed?	197
23.	Transcripts returned	Were transcripts returned to participants for comment and/or correction?	195

Domain 3: analysis and findings			
Data analysis			
24.	Number of data coders	How many data coders coded the data?	196
25.	Description of the coding tree	Did authors provide a description of the coding tree?	NA
26.	Derivation of themes	Were themes identified in advance or derived from the data?	198
27.	Software	What software, if applicable, was used to manage the data?	NA
28.	Participant checking	Did participants provide feedback on the findings?	198
Reporting			
29.	Quotations presented	Were participant quotations presented to illustrate the themes / findings? Was each quotation identified? e.*g., participant number*	198-213
30.	Data and findings consistent	Was there consistency between the data presented and the findings?	198-213
31.	Clarity of major themes	Were major themes clearly presented in the findings?	198-213
32.	Clarity of minor themes	Is there a description of diverse cases or discussion of minor themes?	198-213

Tong A, Sainsbury P, Craig J. (2007) Consolidated criteria for reporting qualitative research (COREQ): a32--item checklist for interviews and focus groups. International Journal for Quality in Healthcare: 19:349 –357

Personal History (Emily Russell)

I was 13 when I was first diagnosed with scoliosis.

Up until this point, I hadn't been aware that there was anything "wrong" with me, and the news came as a bit of a shock to me and my parents. Having addressed my initial concerns (Is it dangerous? And do I have to stop dancing?), we went through the rather clinical process of x-rays, angle measurements, examinations, assessments of the degree of deformity and my "rib hump", before my very kind surgeon sat down and explained to me his recommendations. At this point, as I had two curves which were both under 40° and compensated for each other, the recommendation was not to have surgery but rather to watch and wait. If the curves progressed beyond 40° then a new discussion could happen.

So now I was 13 years old, very aware and rather well informed about how my body was different from "normal". I also hadn't had any intervention to fix this problem, and at the moment there wasn't any intervention that was recommended. I also happened to go to an all girls school, which I imagine didn't help with my insecurity. A school friend referred to my back as "freaky" when we played leapfrog and my rib hump was visible. I started wearing baggy tops and jumpers to cover myself up.

Adolescence is a tricky time for most people, not made easier by an awareness of how you don't fit in. Whilst I had a supportive group of friends I could laugh and joke about my "wonky back" with, outside of this comfort zone I lacked self-esteem. I remember one nurse (who was a complete stranger to me) making a comment to me about how I mustn't slouch – I hadn't been slouching at the time, and small comments like this stuck with me for much longer than they otherwise might have. I now recognise this sort of behaviour as a problem with the person rather than with me, but as a young woman it could make me feel quite upset. Regardless of this, I still made it through adolescence fairly successfully, and I'd like to think reasonably well – adjusted. It is perhaps interesting then, given that I

The symptoms, in the presence of normal scans, examinations, and nerve studies, implied that there was nothing physically wrong with my brain, spine, or nerves. He then explained to me that I was experiencing somatisation.

It will not be a surprise to anyone when I say that emotions can change how the body functions physically.

When we are upset, our eyes spontaneously water.

When we are anxious, our heart pounds and our hands become sweaty.

These are well accepted by the general population as normal responses to emotion.
However, sometimes emotions can't express themselves that way. This has become associated with an undeserved stigma, and the idea that it must be "all in your head" and that people must be making their symptoms up.

Essentially, after the surgery had happened, with my recovery going so well, I was very anxious to tune into my body to know early on if anything went wrong. Just as you had forgotten you had socks on until I reminded you of it here, my body hadn't been telling me about occasional pins and needles until I started paying very close attention to when they happened. every time I had a symptom that I could relate my spine, I would pay attention to it, and focus on it, and so my brain would now automatically draw my attention to it where previously it had not done.

My neurologist was brave enough to suggest that if I acknowledged my symptoms when I experienced them, but then gave their presence no weight, they would go away. He also validated my experience of the symptoms, explaining that he understood that for me the symptoms were very real,

even though there was no underlying nerve damage to explain them. He never made me feel like they were just in my head.

Four months later I was completely better.

I have had other colleagues praise me on being so open about the fact I have experienced somatisation, which surprises me every time, because the vast majority of us will have done, and this should not be stigmatised, and I should not have to be brave to admit it. Many people experience headaches when they are stressed, or tummy pain that no one can explain when they are anxious. The problem with somatisation, is that it takes a very confident doctor to tell you that there is no organic underlying disease. As a result, many people have lots of investigations for their symptoms, many of which are invasive, without any cause being found.

Whilst the lack of a life-threatening course should be good news, the hunt for an underlying diagnosis becomes forefront in the mind, and a source of anxiety for patients. When it then sounds like doctors are implying they are making their symptoms up, this is clearly then very frustrating and alienating.

The result of my personal experience with scoliosis, and how I feel it affected my mood and body image, means I am entirely in support of the suggestion that in order to holistically care for a teenager with scoliosis, there needs to be an appreciation of the potential for underlying impacts on mental health, and provision for support of this within orthopaedic clinics. Whilst all the surgeons I met were clearly brilliant men, I did not feel as a young woman able to ask them all the questions I had without feeling vulnerable: in particular questions around body image, future pregnancy and childbirth. This situation is becoming easier and I know that support exists through my doctors, friends and family, colleagues and support groups such as SAUK.