AN ANALYSIS OF THE SHAPE OF THE SPINE AND TORSO IN THOSE WITH AND WITHOUT SCOLIOSIS

by

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In completing this work I have been fortunate to have the support of my supervisors, Professor Paul Pynsent and Professor Joanne Wilton. I am very grateful for all of their help and advice along the way. With the untimely passing of Professor Wilton, I am grateful to Professor Kumar who stepped in to the breech at very short notice. I am also indebted to Dr Fiona Berryman, who was the originator of the ISIS2 system and has been instrumental in all of my work, especially for her understanding and correction of my writing and grammar. I would like to acknowledge the Birmingham Orthopaedic Charity and the Royal Orthopaedic Hospital NHS Foundation Trust for their support. Finally, I would like to acknowledge my family who have been understanding of my absences over the years throughout my career.

Abstract

The shape of the spine and torso are central to the management of scoliosis, a condition with a three-dimensional rotation in the spine associated with asymmetry of the torso. It is not clear what the variability in shape and symmetry of both the spine and the torso is in a non-scoliotic cohort. Using ISIS2 surface topography, the torsos of a non-scoliotic cohort were measured yearly for seven years, allowing true longitudinal analysis. Parameters of growth and symmetry were measured and analysed to demonstrate the variability of normal shape during the adolescent growth spurt using linear mixed effect modelling and data ellipses, examining for the effects of age and sex. This demonstrated a range of normal shape and the differences between males and females. The non-scoliotic shape was then analysed alongside a group of matched pre and post-operative scoliotic subjects using data ellipses and Procrustes analysis. This showed that scoliosis increases the asymmetry of the spine and torso as an amplification of the variability in the non-scoliotic cohort. This asymmetry is reduced by surgery in nearly all parameters measured. However, some appreciable differences remain when compared to the non-scoliotic cohort.

Contents

Nomenclature.

- ACJ – Acromioclavicular joint
- AIC – Akaike information criteria
- AIS – Adolescent idiopathic scoliosis
- ATR – Angle of trunk rotation
- AxDiffHt – The difference in vertical height (y axis) of the axillary points
- AxDiffOff – The difference in horizontal distance (x axis) from the midline to the axillary points
- BMI – Body mass index
- CD – Cotrel Dubousset spinal system
- CDC – Centers for Disease Control
- CSS – Cosmetic spinal score
- CT – Computed tomography
- DAPI – Deformity in the axial plane index
- DiffHt – The difference in vertical height (y axis) between points on either side of the torso
- DiffOff – The difference in distance from the midline (x axis) between points on either side of the torso
- EQ-5D – EuroQol measure of generic health status
- GPA – Generalised Procrustes analysis
- ISIS – Integrated surface imaging system
- ISIS2 – Integrated surface imaging system 2
- L – Lumbar
- LA – Lateral asymmetry
- LOESS – Locally weighted scatterplot smoothing
- MaxDiffOff – The difference in distance from the midline (x axis) of the most prominent points on either side of the torso

- MRI – Magnetic resonance imaging
- MT – Mid thoracic
- OPA – Ordinary Procrustes analysis
- PA – Postero-anterior
- PCA – Principal component analysis
- PCs – Principal components
- POTSI – Posterior trunk asymmetry index
- PSIS – Posterior superior iliac spine
- PT – Proximal thoracic
- RCPCH – Royal College of Paediatics and Child Health
- SAQ – Spinal assessment questionnaire
- ScapDiffDepth – The difference in 3D height (z axis) between the right and left most prominent points
- ScapDiffHt – The difference in vertical height (y axis) between the right and left most prominent points
- ShDiffHt – The difference in vertical height (y axis) of the shoulder points
- SRS-22 – Scoliosis Research Society 22 question outcome questionnaire
- SRS-30 – Scoliosis Research Society 30 question outcome questionnaire
- TAPS – Trunk appearance perception scale
- TL – Thoracolumbar
- TRACE – Trunk aesthetic clinical evaluation
- VA – Volumetric asymmetry
- VP – Vertebra prominens
- UK90 – The UK growth surveys of the 1990s
- UK-WHO – The combination of the UK90 and WHO growth standards
- WaistDiffHt – The difference in vertical height (y axis) of the waist points

- WaistDiffOff – The difference in horizontal distance (x axis) from the midline to the waist points
- WHO – World Health Organisation

Parts of this work have been published as (and are found in the Appendix):

- Gardner A, Berryman F, Pynsent P. A description of three-dimensional shape of the posterior torso comparing those with and without scoliosis. Symmetry (2019). 11; 211.

- Gardner A, Berryman F, Pynsent P. The development of kyphosis and lordosis in the growing spine. Spine. (2018); 43; 19: E1109-E1115.

- Cheshire J, Gardner A, Berryman F, Pynsent P. Do the SRS-22 self-image and mental health domain scores reflect the degree of asymmetry of the back in adolescent idiopathic scoliosis? Scoliosis and Spinal Disorders. (2017); 12; 37: 1-7.

- Gardner A, Berryman F, Pynsent P. The effects of scoliosis and subsequent surgery on the shape of the torso. Scoliosis and Spinal Disorders. (2017); 12; 31: 1-12.

- Gardner A, Berryman F, Pynsent P. What is the variability in shoulder, axillae and waist position in a group of adolescents? Journal of Anatomy. (2017); 231; 2: 221-228.

- Gardner A, Price A, Berryman F, Pynsent P. Erratum to: The use of growth standards and corrective formulae to calculate the height loss caused by idiopathic scoliosis. Scoliosis and Spinal Disorders. (2016) 11; 16.

- Gardner A, Price A, Berryman F, Pynsent P. The use of growth standards and corrective formulae to calculate the height loss caused by idiopathic scoliosis. Scoliosis and Spinal Disorders. (2016) 11; 6: 1-9

Originality of this work.

This thesis adds to the literature about the management of scoliosis through the analysis of the 3D shape of the torso in scoliosis and how that is changed by scoliosis surgery referenced to a cohort of non-scoliotic children. This was performed using linear mixed effect modelling and Procrustes analysis. This thesis also establishes normative data for torso shape and growth for future work in this area.

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My role in the study.

As part of the work that has lead to this thesis, I have personally:

1. Designed and written the protocol, participant and parent information sheets and consent and assent forms used for the recruitment of the school children that formed the non-scoliotic cohort. These documents can be found as part of the appendices.

2. Applied for ethical approval for the use of ISIS2 surface topography for the normal group.

3. Recruited the children for the non-scoliotic cohort by meeting children and parents at parent evenings, speaking to class teachers and presenting at whole school gatherings.

4. Provided physical assistance in the transport and assembly of the ISIS2 equipment to the school.

5. Designed and written the protocol and applied for ethical approval to retrospectively review the ISIS2 images. The images were taken as part of routine care from the hospital. The protocol can be found in the appendix.

6. Reanalysed all of the ISIS2 photographs from both the non-scoliotic and scoliotic cohorts for the identification of the torso points alongside the data from the standard ISIS2 output.

7. Analysed all of the data, using programmes I wrote for R. These produced the plots seen in the figures and the statistical analysis seen throughout the thesis.

8. Performed, with a colleague, an assessment of inter-observer and intra-observer error.

9. Written all of the thesis.

10. Written, as first author, all of the papers (except one) listed in the appendices.

I did not take any of the ISIS2 images. This is for two linked reasons. Within the hospital where the images are taken as part of routine care, the images were either already available and were reviewed retrospectively, or were obtained as part of standard care by hospital staff employed to perform this task. The images from the school were from children within the peer group of my own children, including their friends. I felt that this would be a moral conflict of interest and also might limit recruitment. The ethics committee agreed and were also keen that a male was not present for the taking of images in the school in the presence of girls who would have to undress to take part in the study.

1 Introduction.

The management of scoliosis, particularly in teenagers' can be a clinical challenge. This is because the treating doctors need to deal with the spine, a structure of both bone and neural tissue, and also treat the effects that the spine has on the body [1, 2]. Scoliosis causes torso asymmetry [3], and scoliosis surgery attempts to equalise that asymmetry. However, surgery does not effect a complete correction of deformity [4]. On the other hand, it is unclear how much correction is enough or appropriate [4], particularly if compared to the variability of torso and back shape seen within the non-scoliotic population.

This thesis examines the changing torso shape of a group of children who do not have spinal deformity, through serial measurements of their backs over a period of seven years in a longitudinal fashion. Using this knowledge of how a 'normal' back behaves, the shape of the torso in a cohort of adolescents with scoliosis, both before and after their operation, are examined to identify the effects of scoliosis on the torso and what surgery really does in correcting abnormal torso shape.

1.1 Subtypes of scoliosis.

Scoliosis is a deformity of the spine that has been recognised in history and is described in the ancient world in Hindu texts [5] and in the teachings of Hippocrates [6]. Scoliosis is a term that describes a lateral bend in the spine when viewed in a coronal (or anteroposterior) plane [3]. In this view, the spine should be straight between the occiput superiorly and the pelvis inferiorly. Scoliosis is a condition that can affect all ages, although from different aetiologies [3]. By definition, scoliosis is diagnosed once the lateral bend or curve of the spine is greater than 10° measured using the Cobb technique [7, 8].

1.1.1 Early onset scoliosis.

The causes of scoliosis are best subdivided using a combination of age and aetiology. Scoliosis can be seen in early childhood. This type of scoliosis is now commonly referred to as 'early onset scoliosis' (EOS) and is defined as scoliosis from any cause affecting the child under the age of 10 [9]. The term EOS refers to the effect that the scoliosis has on the development of the underlying lungs and associated respiratory function [10]. A failure to maintain adequate growth of the thorax and underlying lungs, particularly before the age of 7 years is associated with a smaller number of total alveoli [11], and an earlier death [12]. The management of EOS revolves around maintaining spinal growth and as symmetrical a thorax as possible to maximise lung development [13], through external orthoses (casting and bracing) or surgically using growing spinal systems such as the MAGEC® rod (copyright NuVasive Inc).

The causes of EOS are seen in three different groups, congenital, idiopathic and associated with another neuromuscular or syndromic condition. Congenital scoliosis is caused by the presence of abnormal bony architecture within the vertebral column. This is thought to be the result of an insult of unknown origin to the developing foetus causing disturbance in the development of the centres of ossification, and the subsequent segmentation of the vertebral bodies [14]. Congenital scoliosis can be associated with anomalies in other body organs such as the heart or kidneys and can also be associated with the VACTERLS grouping of abnormalities (abnormalities in the vertebral, anorectal, cardiac, tracheo-oesphageal and renal systems, along with limb abnormalities and a single umbilical artery) [15]. The amount of deformity that a particular arrangement of congenital abnormalities might cause has been documented by McMaster et al [16, 17]. Whilst it is not possible to return a congenital scoliosis to a spine without congenital anomalies, the management is based on preventing the development of scoliosis at the site of the anomaly, and also to prevent the development of compensatory curves elsewhere in the spine, allowing an overall balanced posture of head over pelvis [18].

Infantile idiopathic scoliosis in the EOS group is a curve that develops in a child who does not have either a congenital anomaly or a medical condition which is associated with the development of scoliosis. By definition, infantile is from birth to the age of 3 years [19]. The idiopathic curve is often noted in early life before the child can sit up independently. Sometimes this type of curve will regress spontaneously, usually as the child develops trunk control and sits up [20]. There is an association of intra-dural anomalies that are thought to contribute to the development of an idiopathic curve in this age group, which is higher than the rate seen in the adolescent group (see later in the text) and so all children need to have an MRI scan of the entire neural axis [21].

Syndromic and neuromuscular conditions of childhood can be associated with the development of a scoliosis. Common examples of defined syndromes in this group are neurofibromatosis type 1 and spinal muscular atrophy. In these groups, management is again to maximise spinal growth and lung development but is tempered by the underlying diagnosis, other co-morbidities and life expectancy [22, 23].

EOS is now classified using the C-EOS system, popularised by Vitale et al [24]. This classification is based on age, progression, aetiology and overall kyphosis of the spine. The most commonly used outcome tool for EOS is the 24 question early-onset scoliosis questionnaire (EOSQ) [25] which asks questions around the health and activity of the child along with the effect on the other members of the family.

1.1.2 Adolescent scoliosis.

Adolescent scoliosis is defined as occurring between the ages of 10 and 18 years [19]. Again, in a similar way to the EOS group, adolescent scoliosis is either idiopathic and seen in otherwise normal children, or secondary to an underlying cause. Adolescent idiopathic scoliosis (AIS) is the most commonly seen form of childhood spinal deformity

[3]. The exact cause of AIS is still unclear, with a number of candidate genes identified [26]. For reasons that are unknown, AIS is more commonly seen in females [3]. Reasons for this may include differences in the age of the growth spurt between males and females along with differences in the sagittal profile of the spine (the amount of kyphosis and lordosis and how that changes with age) [27, 28, 29]

The issues of AIS can relate to external body shape and asymmetry. This is particularly seen in an unequal shoulder height and axillary height along with waist crease asymmetry. When bending forwards a rib or loin hump is seen. What is clear is that adolescents with scoliosis seek medical attention, not because they know that their spine is not as it should be, but because they are aware that their overall shape is not as they would wish [30, 31, 32].

In clinical practice, AIS is investigated through a combination of physical examination, radiographs of the spine and more specialist imaging of the spine, thorax and abdomen. Examination by a clinician documents the visual appearance qualitatively, a dynamic process associated with assessment in different positions and during movement. When the patient is viewed in a posture of spinal flexion, either standing or sitting, the asymmetries of the posterior aspect of the torso become more apparent. This forms the basis of the Adams forward bend test [33]. Radiographs are taken in the upright, weight bearing position. This maximises the size of any deformity as gravity is an active force on the body and means the radiograph is a true representation of the spinal shape in the upright position. The whole spine has radiographs taken in both a back to front (postero-anterior or PA) view and a lateral view. These are static two-dimensional (2D) images. The size of the curves are measured using the Cobb angle [7] although alternative methods are described such as the Ferguson angle [34] amongst others [35]. The Cobb angle is the angle subtended between two lines drawn parallel to the endplates of the most angulated vertebrae relative to the horizontal at either end of the curve (the points of inflection). The Cobb angle is the 'gold standard' for the measurement of the size of a scoliosis. Radiographs are repeated on an intermittent basis, usually twice a year during the growing period of the child to monitor changes in the shape of the spine. The use of specialist imaging in AIS includes both Magnetic Resonance Imaging (MRI) and Computed Tomography (CT). An MRI scan is used to assess the neural structures from the base of the brain to the end of the cauda equina in the sacral cul-de-sac. The MRI is performed to investigate the potential presence of neural axis abnormalities that can be the cause of the scoliosis. This is of clinical relevance as, if an abnormality is found, it may require treatment in its own right and can also change the risks of spinal injury and paralysis during surgical intervention. An MRI scan is usually only performed once unless there are symptoms that develop indicating irritation of the neural structures. A CT scan is required if there is a need to see the 3D architecture of the vertebral bodies. This may be required for the planning of spinal instrumentation as part of the preparations for surgical intervention. Both an MRI and CT scan are usually performed in a supine position. Like a radiograph, both are static images. Assessment of the size of the curve from either an MRI or CT scan is less useful to the clinician, as the supine position eliminates the effects of gravity and the curve will reduce in size. The supine position also flattens the rib hump because of the weight of the body on it, this affects the shape and size of the spinal curve and the rib hump compared to the shape when standing [36]. Radiographs and CT scans use ionising radiation. There will be a cumulative radiation dose to the child over the time of their treatment and this is associated with an increased risk of malignancy in later life [37, 38].

AIS is a condition where surgery is not mandatory. AIS is not associated with the respiratory problems of EOS and is a far more benign condition [39]. Surgical intervention is really only considered for curves that reach a Cobb angle of 50° or more.

The management of AIS in curves smaller than 50° is aimed at preventing the curve from getting larger towards the surgical range. This is often through the use of bracing [40]. Bracing is the wearing of an external orthosis that applies forces to the spine, correcting spinal shape and preventing the development of further scoliosis. There are many different types of brace, the most common based on the original Boston type [41]. The best evidence for the efficacy of bracing is the BRAIST study [40]. The BRAIST study compared full time brace wear against observation for curves between 20° and 40°, where there is remaining skeletal growth. Success of either treatment was defined by a curve of less than 50° at skeletal maturity. BRAIST demonstrated that bracing reduced the number of children who developed a scoliosis large enough to consider surgery in 75% of those braced compared to 42% of those observed. Wear for longer than 13 hours per day led to a better result. Unfortunately, there is resistance to prolonged brace wear from adolescents and this leads to compliance issues [42]. There is also a growing literature that reports on the use of scoliosis specific exercises, as popularised by Schroth [43, 44]. In the UK, the National Institute for Health Research Health Technology Assessment (NIHR HTA) funded ACTIVATES trial (Active Treatment for Idiopathic Adolescent Scoliosis (ACTIvATeS): a feasibility study) [45] carried out a feasibility study to assess whether a full randomised controlled trial of scoliosis specific exercises in the management of AIS would be warranted in the future. They concluded that there was scope for a full RCT. A recent systematic review by the same group has noted that the quality of the evidence so far published on scoliosis specific exercises is poor [46], and this is confirmed by Day et al [47].

In the planning of surgical management, AIS was historically classified using the King-Moe classification system [48]. This system was a coronal only classification that recognised certain patterns of scoliotic curve morphology. This then helped surgeons to decide on the correct surgical management, assuming the use of the posteriorly based distraction rod of Harrington. The Harrington rod was developed by Paul Harrington from Houston, and reported in 1962 [49], initially for the management of the so called paralytic scoliosis caused by polio but subsequently used in other spinal deformities. The Harrington rod was successful in the correction of the coronal deformity at the expense of the sagittal plane, and was associated with a loss of both kyphosis and lordosis and the development of the 'flat back' [50]. Attempts to recreate the sagittal profile whilst effecting a coronal correction of scoliosis led to the development of segmental correction using multiple fixation points through the use of sublaminar wires [51] and subsequently the Cotrel-Dubousset (CD) system [52]. The development of multi-level pedicle screw fixation as popularised by Suk [53] in the mid 1990s, allowed greater correction of any scoliosis. This ability, along with the recognition that AIS is a condition associated with, and thought possibly to be caused by, a change in sagittal profile when compared to those without scoliosis [54], made the King-Moe classification redundant and there was a need for a more comprehensive system. This came from the group led by Lenke [55] in 2001. The Lenke classification system makes assessments of the coronal and sagittal shapes of the spine, along with the size of any compensatory curves (the lumbar modifier). The Lenke classification is based on both the major and minor curves. The major curve is always the largest curve in terms of Cobb angle. The major curve is the curve that defines the coronal subtype as the first part of the classification. The most commonly seen coronal subtype is the convex to the right thoracic curve. More recently, acting to improve on the Lenke system, further three dimensional (3D) classification systems have been developed. These include the top down Da Vinci system [56] and others based on mathematical principles [57, 58].

The surgical management of AIS is different to that of EOS. in AIS, it is not essential to be able to allow continued spinal growth, and a bone fusion needs to be created. The surgical alteration in spinal shape is maintained through the use of implants whilst bone union occurs. There was a period of anterior spinal fixation for scoliosis, popularised in the UK from Birmingham, using the Kaneda system [59, 60], amongst other systems [61]. However, with the advent of more reliable posterior pedicle screw systems with more corrective power, the majority of surgery is now performed from a posterior approach [62]. Modern surgical techniques are recognised to correct approximately 65% of any deformity [4].

Scoliosis is also seen in adolescents with an underlying medical cause (neuromuscular or syndromic). This type of scoliosis develops during adolescence or is already present as the child reaches adolescence. The commonly seen circumstances when this occurs is when there is a diagnosis of conditions such as cerebral palsy (CP) or Duchenne muscular dystrophy (DMD) [63]. The difference in this type of curve when compared to AIS is in curve location, as CP and DMD curves are predominantly seen in the lower thoracic and thoracolumbar spines and can be associated with pelvic obliquity [63]. The aim of the management of this type of scoliosis is to maintain the best quality of life for the child with minimal pain whilst preventing progression of the curve such that medical harm and/or a loss of function is prevented. In severe cases with children diagnosed with total body CP, then the aim is to allow comfortable sitting in their wheelchair, allowing continued use of the upper limbs and prevention of costoiliac impingement [64]. For DMD, comfortable sitting in the chair and the relief of costoiliac impingement are the key outcomes of surgery in the knowledge of the natural history of continued deterioration in shape, function and health if the spine is left without intervention [65].

1.1.3 Adult scoliosis.

Scoliosis in adults comprises two different groups. Those who are adults with AIS from earlier in life and those who develop adult degenerative scoliosis (ADS) [66]. ADS is a consequence of asymmetric degenerative change in the spine which typically causes a thoracolumbar curve with associated listhesis of the vertebral bodies [66]. The difference between those with ADS and those with AIS, is that ADS becomes symptomatic. Symptoms are because of pain from the degenerative change within the vertebral column, nerve root irritation and radicular pain from the listhesis and from a loss of the ability to maintain upright posture [66]. The failure to maintain an upright posture is known as sagittal imbalance and is caused by a loss of lumbar lordosis and a mismatch between the balance of the spine and the pelvis [67].

The management of an adult scoliosis of either type is, in the main, conservative. The lack of spinal growth eliminates the rapid increase in curve size that is seen in children and adolescents. Surgery is indicated for significant deformities with pain that cannot be managed non-operatively in those medically fit enough for the surgery [68, 69]. The aim of surgery is broadly around recreating the amount of lumbar lordosis that each individual patient requires, which is defined by their individual pelvic anatomy [70, 71]. This is done through techniques including both spinal osteotomy and vertebral column resection, both of which involve the removal of pieces of bone, ranging from a small amount of facet joint posteriorly, to the removal of one or more entire vertebral bodies, to allow the creation of the correct spinal shape [50, 72].

1.2 Issues with some aspects of the management of adolescent idiopathic scoliosis.

The external appearance of an adolescent with scoliosis plays a large part in their desire for treatment; they want to reduce the size of the visible deformity [73]. As noted previously, surgical intervention in AIS removes approximately 65% of the asymmetries [4]. What is less clear is the variability in shape of the back and torso in those adolescents without spinal deformity. To correct a deformity back to 'normal', a surgeon needs to know what would be described as 'within the normal range'. The measurement of growth of different parts of the body, and of the body as a whole has been investigated in various different circumstances for hundreds, if not thousands of years [74]. Initially seen as part of the burial practices of the Egyptian pharaohs, measuring growth was a feature of the slave trade, military service and policing (particularly in the pre-finger print era) [74]. A greater use of scientific principles and an understanding of the effects of health on growth, particularly in children, led to large scale growth studies in the 20th century exemplified by Tanner et al [75] with the Harpenden growth study. Not only the amount of growth, but also the mechanisms of growth have also been studied, particularly by D'Arcy Thompson in his book 'On Growth and Form' [76], where he applied mathematical principles to the growth seen across species, demonstrating a common underlying pattern.

To identify what is abnormal requires that all of the parameters by which someone with scoliosis is assessed, are benchmarked against the variability in that parameter seen in children without scoliosis. To be able to achieve this, that variability must be measured and documented. To make matters more complicated, the shape of children continues to change until they reach late adolescence and early adulthood. Up to that point they are growing and developing and so any assessment of normality needs to take into account the age of the subject. Added to this, is the difference between the sexes. Males and females develop in different ways during adolescence which will affect what is classed as 'normal'. What is normal in the assessment of scoliosis is not clearly defined in the literature.

A question that occurs in any discussion around the appreciation of the external shape of the torso in the management of scoliosis is 'how to get a measure of the problem?'. As noted previously, those with scoliosis come to seek medical attention because of what they perceive they look like to themselves and to others. This is not assessed well by radiographs. Radiographs will show a 2D shape of the spine.

Clinical photographs can be used to show the external shape of the body but obtaining quantitative measures requires the use of different techniques. Surface topography is a radiation free technique that documents the 3D shape of a surface. Surface topography is a method used for measurement of scoliosis since the 1970s [77]. Current systems allow a measure of the shape of both the spine and torso and are used in clinical practice [78, 79]. Surface topography allows measures of the shape of the spine and torso to be made examining the effects of growth and sex on shape in those with a straight spine without subjecting them to invasive radiation. Measures can also be made in those with scoliosis to examine the effects of scoliosis on torso shape, and then make a comparison to the shape of those without scoliosis. Finally, the shape of those with scoliosis who undergo surgery can be cmeasured in both the pre-operative and post-operative stages and then compared to the non-scoliotic cohort.

1.3 The aims of this thesis.

This thesis answers three questions. What is the shape of the torso of the non-scoliotic adolescent and how is that affected by growth, including those differences caused by the differential growth between males and females? What does a diagnosis of AIS do to the 3D shape of the spine and torso? Finally, how does the surgical intervention performed for scoliosis alter that 3D shape and does it return it to that of the non-scoliotic?

These questions are important as they address the underlying issues that face adolescents with, and doctors treating AIS. By examining these issues, it is possible to establish what the variability of normal shape of the torso around perfect symmetry is in a cohort of children without spinal deformity. Following on from that, the shape of the torso in non-scoliotic children can then be followed through adolescent growth, and be separately examined for the any changes that occur between males and females. Once this range of normality is established, it is then possible to examine what the differences in shape caused by AIS are on the shape of the torso, particularly looking at external anatomical features that are concerns of the person with AIS.

The end treatment of AIS is surgical intervention, aimed to prevent progression and to minimise all deformities of the torso. The effects of surgery are not well documented and measuring how surgery changes the shape of the torso will address this shprtcoming. Whilst beyond the scope of the thesis, the ultimate goal would be to be able to produce a personalised prediction of the results of an operation for AIS, individualised for each adolescent, rather than the rather generic statement of a '65% improvement' [4]. This thesis takes the current knowledge along the path to that goal.

2 Literature review.

2.1 Introduction.

This literature review is written to cover the pertinent work that underpins this thesis and to 'set the scene' for the chapters that follow. It will cover the development of growth standards, the measurement of cosmesis, methods of measuring surface shape with devices, normal torso asymmetry, measures of kyphosis and lordosis and the use of surface topography in the management of scoliosis.

The search strategy used to find relevant literature was based on a thorough search for work that involved the key terms seen in each topic of interest. Given that the literature is varied and covers a long time period (decades), a degree of detective work was required to identify key papers through reference lists and conference papers, alongside internet search engines including Pubmed and Medline. Of particular, more historical interest, were papers from the International Research Society of Spinal Deformities (IRSSD) where work of this type has been preferentially presented and discussed.

The literature presented here has been critically appraised around quality, methodological issues, potential biases and any valid conclusions that can be drawn, both within the paper as it is presented but also in the framework of this thesis. However, the literature review is in the form of a narrative rather than a systematic review due to the small number and quality of the papers that were available for review, particularly around the shape of the non-scoliotic child, and the surface topographical effects of scoliosis surgery.

2.2 Growth standards.

2.2.1 Description.

Growth of height and weight has been studied extensively in the past. Standards for growth and the development of standing height, weight and body mass index (BMI) have been published by the World Health Organisation (WHO) [80], the Centers for Disease Control (CDC) [81], and the Royal College of Paediatrics and Child Health (RCPCH defining the UK-WHO) [82]. Furthermore, standards for sitting height, sitting to standing height ratio and leg length have been published for a Dutch population [83]. The CDC standards were derived as a composite of five surveys of children within the USA between 1963 and 1994 and are shown from birth to 36 months and then from 2 to 20 years [81]. The WHO standards are a composite of data collected from the National Center for Health Statistics in 1977 [84] and more recent data for the early years (birth to 4 years of age) [80]. The early years data are from surveys of children from Ghana, India, Norway, Oman and the USA. As these two data collections are not temporally related there is a discontinuity between the standards for the younger child and the older child in the WHO system. The WHO standards cover from birth to the age of 19 years of age. When the WHO and CDC growth standards are compared, it is shown that the CDC standards represent a shorter, heavier cohort and this is affected by whether the child was breast or formula fed in the early years [85]. The UK standards take the birth to 4 year data from the WHO and combine it with UK growth data from 4 to 18 years of age collated from the UK90 growth surveys 1995 and 1996 [86] (Figures 1 to 3). The WHO standards are presented as the median value and then $\pm$ 1, 2 and 3 z-scores as standard deviations away from the median. The CDC and UK-WHO standards are presented as the median and centiles from the median. The CDC and UK-WHO standards are standing height, weight and BMI across all ages and for both males and females. The WHO standards chart standing height, weight and BMI to the age of 10 years but only standing height and BMI beyond this, stating that due to the variation in the timing of the adolescent growth spurt, weight on its own becomes unreliable at this age and BMI is a better measure [80].

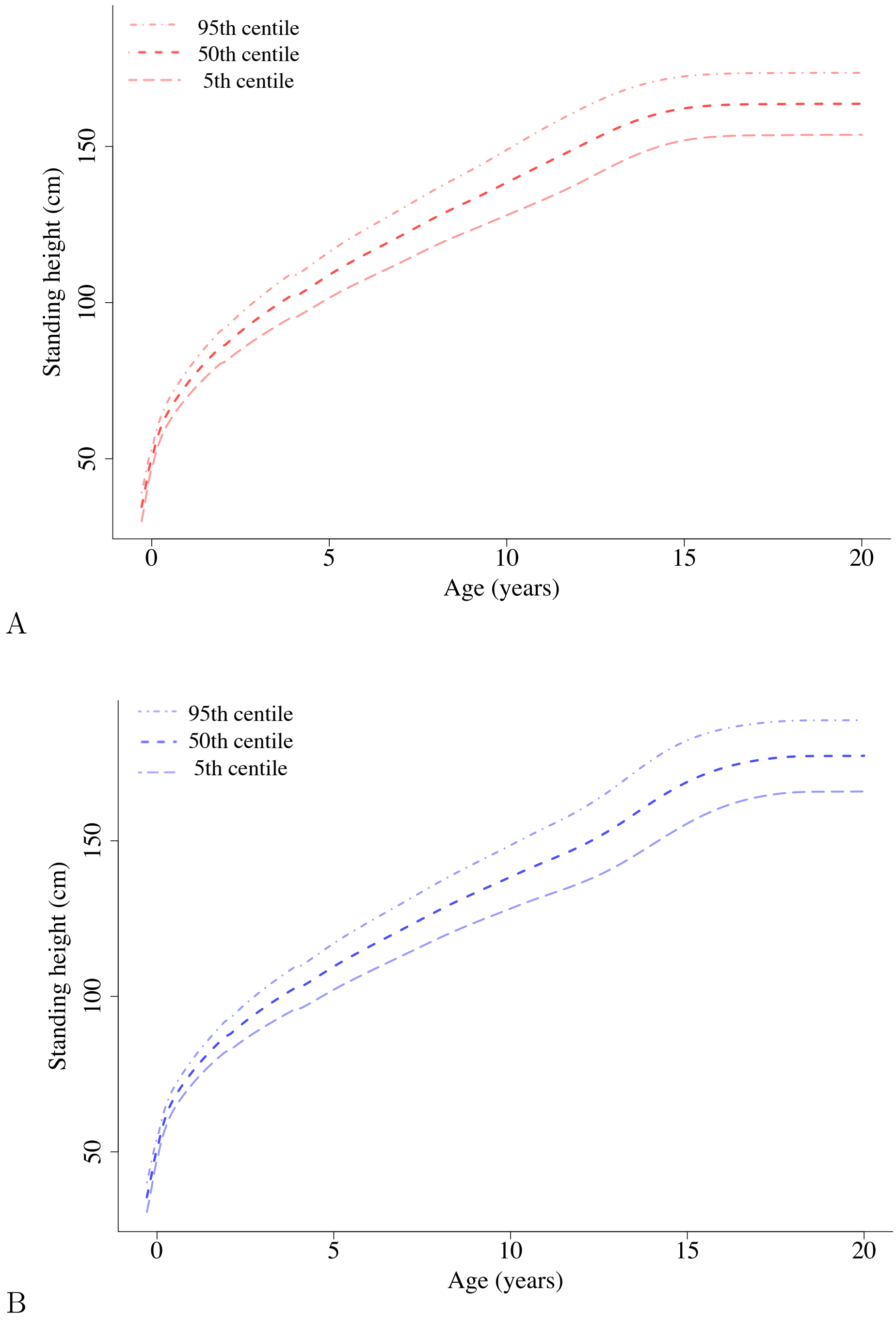
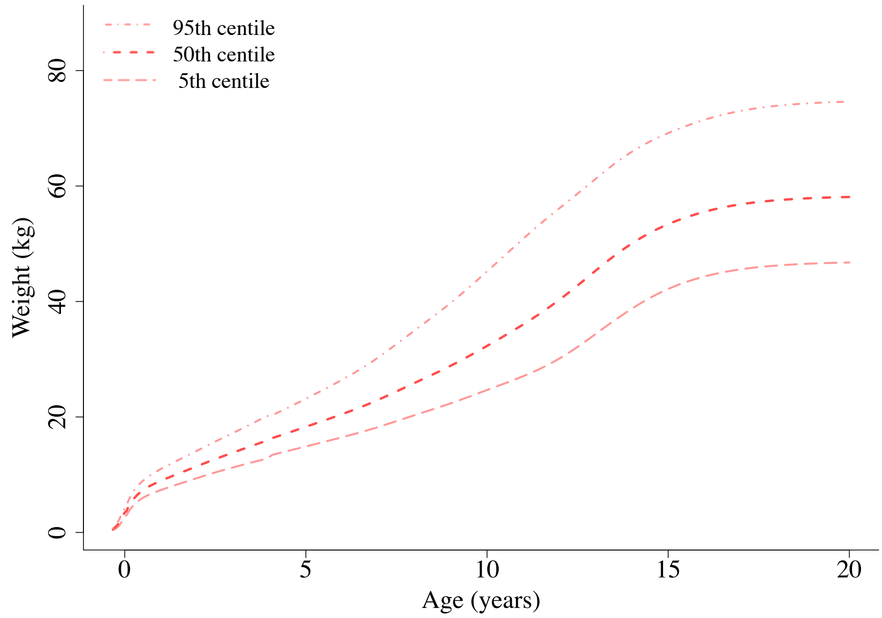


Figure 1: Standing height (cm) against age (years) for the growth standard of females (A) and males (B). The lines represent the median along with the 5th and 95th centiles for the growth standard. The growth standard was redrawn from data provided by RCPCH [82, 87].



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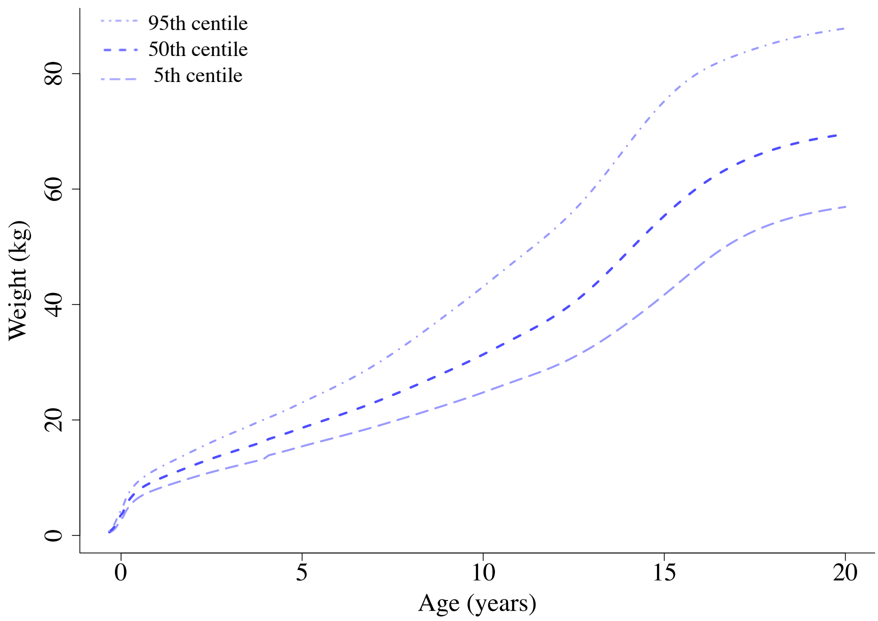
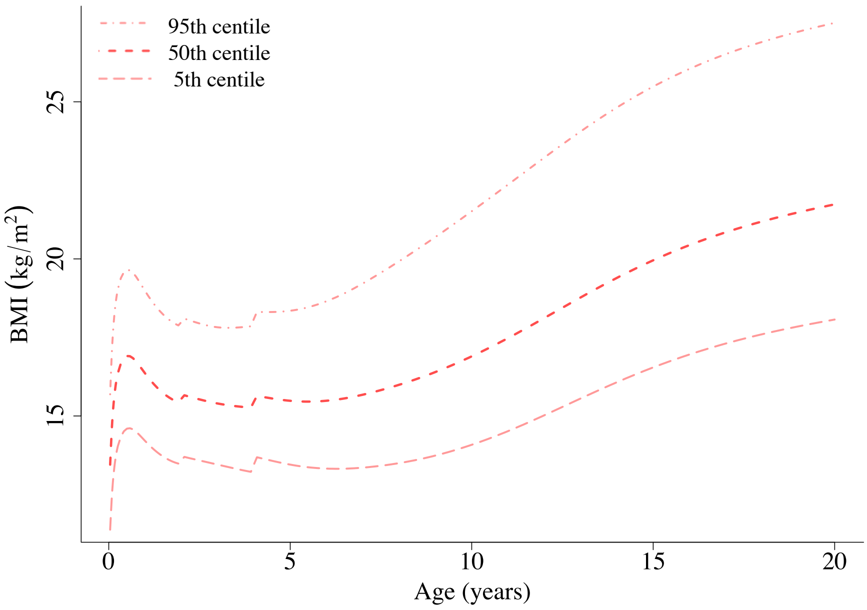


B

Figure 2: Weight (kg) against age (years) for the growth standard of females (A) and males (B). The lines represent the median along with the 5th and 95th centiles for the growth standard. The growth standard was redrawn from data provided by RCPCH [82, 87].



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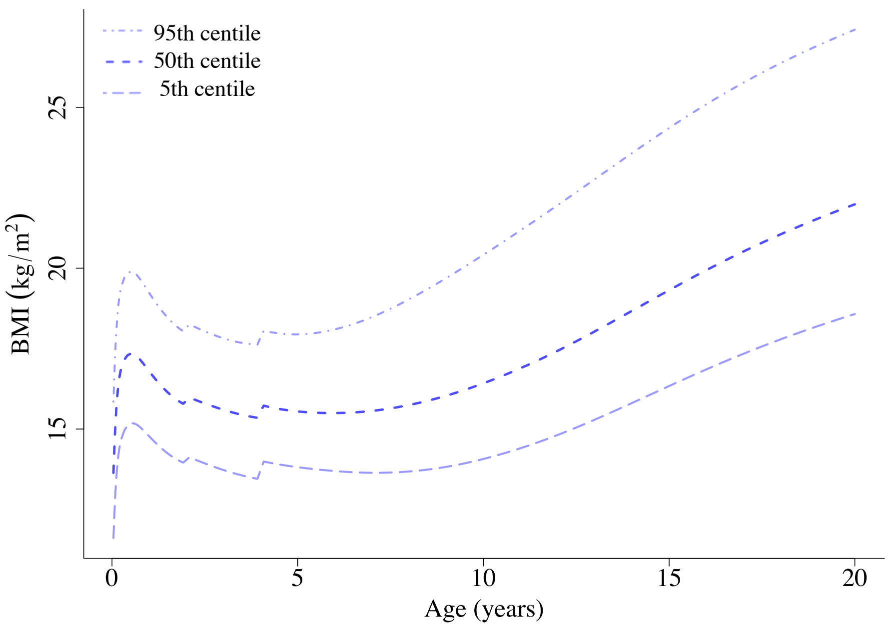


B

Figure 3: BMI (kg/m^2) against age (years) for the growth standard of females (A) and males (B). The lines represent the median along with the 5th and 95th centiles for the growth standard. The growth standard was redrawn from data provided by RCPCH [82, 87].

Whilst all of these standards have slight differences, when they are all plotted together using the z scores of the WHO standards and the centiles of the CDC and UK-WHO standards, there is little difference between them (Figure 4 showing standing height mean and ± 2 z-scores or 5th and 95th centiles against age for WHO, CDC and UK-WHO standards).

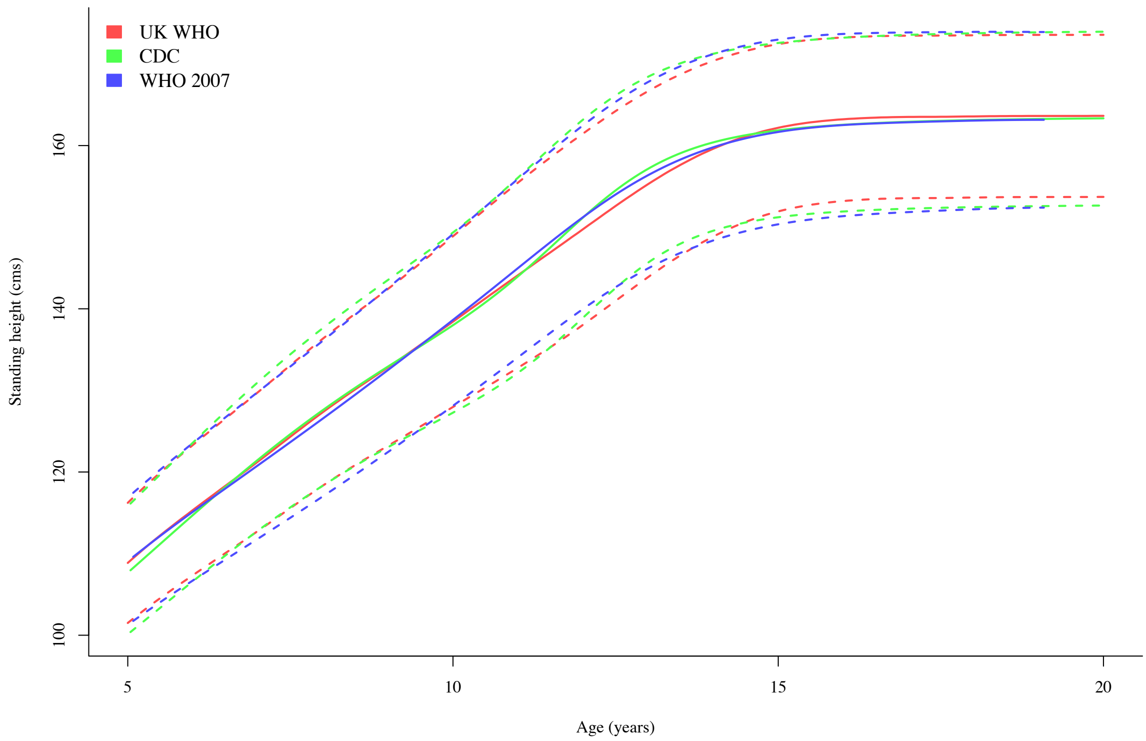
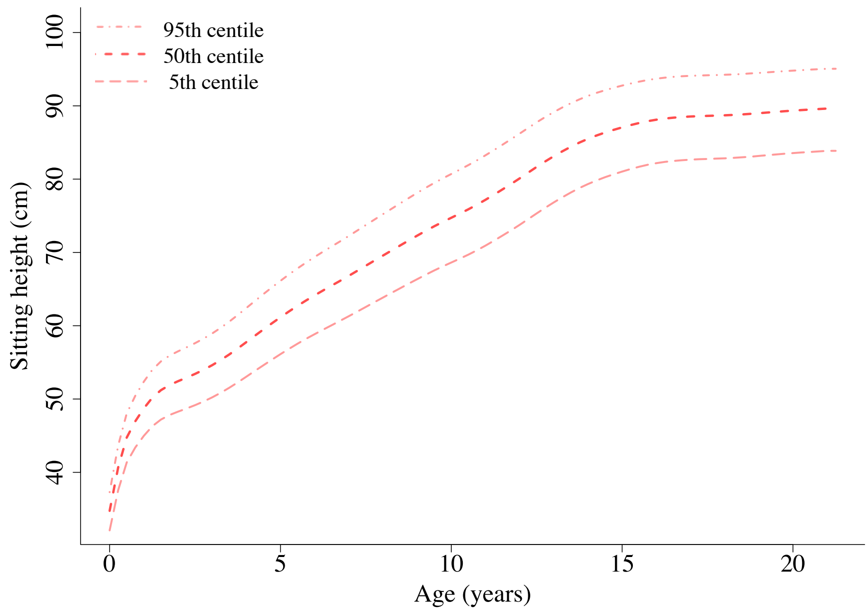


Figure 4: WHO, CDC and UK-WHO growth standards for standing height (cm) versus age (years) in females with median, and ± 2 z-scores (WHO) or 5th and 95th centiles (CDC and UK-WHO). Redrawn [87] from de Onis et al [80], Kuczmarski et al [81] and RCPCH [82].

The sitting height standards of the Dutch population are also presented as median and centiles [83] (Figures 5 to 6). It is of note that Northern Europeans are one of the tallest populations in the world [88] and comparison of such data with other populations may be problematic. The use of the sitting to standing height ratio eliminates this problem, assuming that the distribution of trunk heights and leg lengths are similar between the Dutch and any other population studied. Thus the sitting height to standing height ratio is applicable to the UK population, especially given the similar historical and genetic background of the Dutch and UK populations [89]. Measures of the width of the torso have also been published in a normal population as the biacromial distance (shoulders) and bicristal distance (hips) [90]. Of note, there is a difference between males and females here during the adolescent growth spurt for the biacromial distance but not for the bicristal distance.



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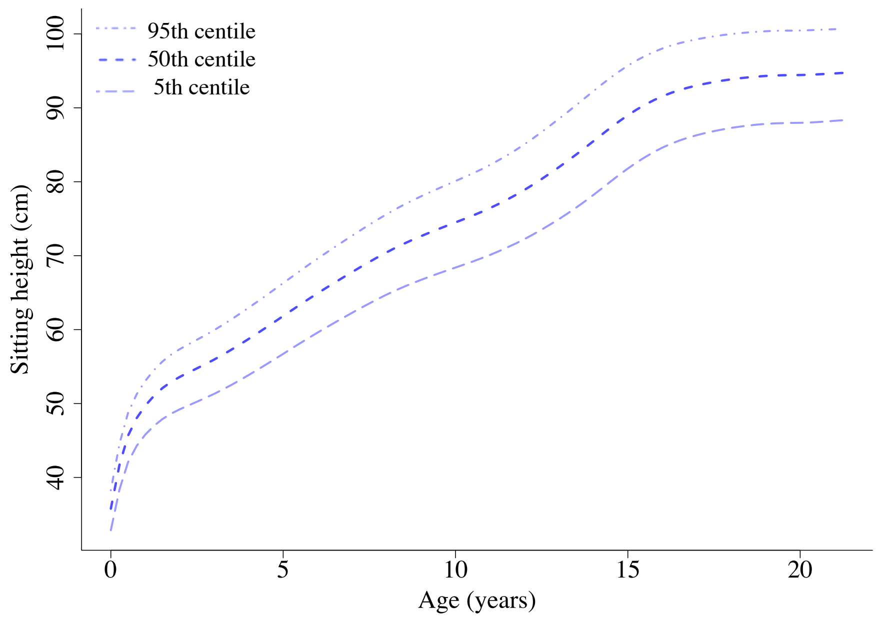
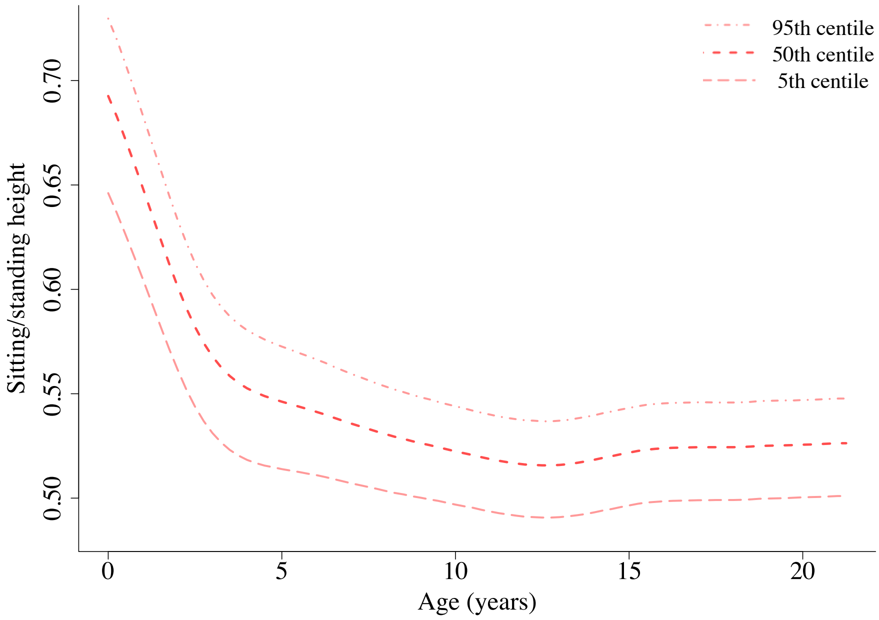


B

Figure 5: Sitting height (cm) against age (years) for the growth standard of females (A) and males (B). The lines represent the median along with the 5th and 95th centiles for the growth standard. The growth standard was redrawn from data provided by Fredriks et al [83, 87].



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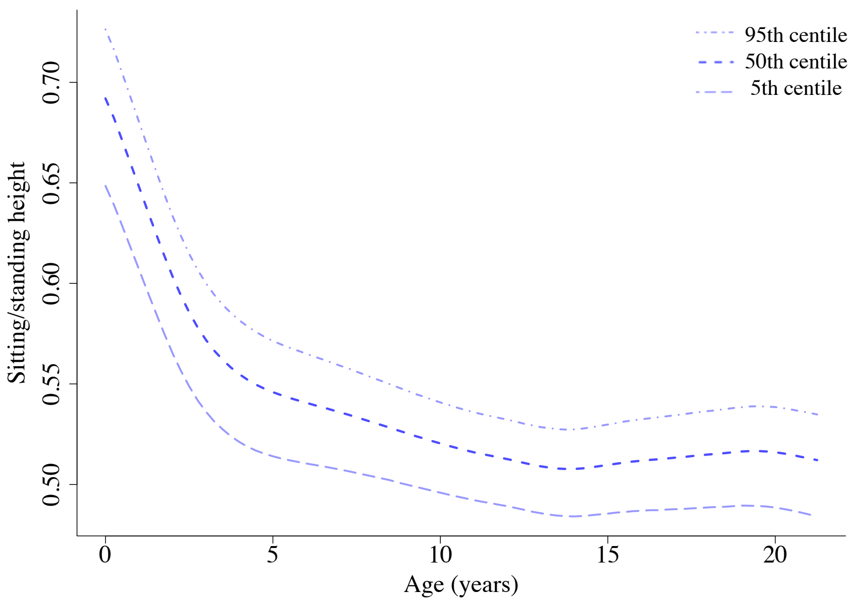


B

Figure 6: Sitting height to standing height ratio against age (years) for the growth standard of females (A) and males (B). The lines represent the median along with the 5th and 95th centiles for the growth standard. The growth standard was redrawn from data provided by Fredriks et al [83, 87].

2.2.2 Critique.

Unfortunately, all of the studies that have resulted in the published growth charts are from historical data, particularly when created for older children and adolescents. This can be seen as the source data for this age group for the WHO growth charts is from 1977 [80], and for the CDC data, 1963 to 1994 [81]. The UK-WHO uses data from 1995 and 1996 [82]. For sitting height and sitting height to standing height ratio, the Dutch standards are from 1996 to 1997 [83]. To use these charts in any form needs to take in to account the differences in size that may have occurred across the population since the source data was collected through improvements in diet and healthcare, particularly in affluent countries. However, given the amount of work that is required to collect the data to create these references for growth, it is hardly surprising that it is not an exercise that is repeated regularly.

In deciding what the correct growth standard to use against the data collected for this thesis, the age and the geographical location of collection of the source data must be considered. The data collected for this thesis is based solely in the UK and between 2010 and 2017, the standard used to reference height, weight and BMI against is the UK-WHO as the reference data that is the basis of the standard was collected at the closest time to this thesis. Again, noting Figure 4, there is little difference between them to visual inspection. For sitting height and sitting height to standing height ratio, the Dutch standards were chosen becasue they are geographically close to the UK and come from similar years to that of the UK-WHO standards.

2.3 The measurement of cosmesis.

2.3.1 Description.

Those who seek medical help for their scoliosis are mainly interested in how they look [91]. In some young people with scoliosis, perception of their own shape has been reported to lead to mental health problems that include eating disorders and depression [1, 2] and summarised by the literature review of Gallant et al [92]. Studies into what anatomical features of the shape of the back and torso cause the most concern to those with AIS have revealed that 75% of shape dislike is made up of waist asymmetry, shoulder height difference and scapula prominence [30]. Classifications of shape in AIS have concentrated on these features and are used by both patients and doctors to score the degree of external deformity [31, 93, 94]. All of these systems are predicated on 'normal' being a completely straight spine in the coronal plane with no asymmetry between the two sides of the torso; a completely symmetrical body. Sanders et al [94] have validated what was originally known as the Walter Reed Visual Assessment Scale [95] and now as the Spinal Assessment Questionnaire (SAQ). The SAQ is a series of seven sets of pictures, each demonstrating an aspect of the body (that are titled: curve, head over pelvis, rib prominence, shoulder level, flank prominence, scapula rotation and head, rib and pelvis). In each set of pictures there are five individual images that show a non-deformed torso through to a very deformed torso, demonstrating increasing deformity in that anatomical view. Individuals with scoliosis then indicate which of the pictures they feel best represents their own deformity. This is then summed to give a total score where the higher the score, the more deformied the subject feels their torso is. The SAQ has been streamlined by Bago et al [93] as the Trunk Appearance Perception Scale (TAPS). This uses only two of the sets of pictures from the SAQ (curve size as viewed from behind and the size of rib hump when bent forwards) and adds a frontal view. Bago reports that the TAPS score is easy to use and has characteristics that make it suitable for use by the patient in scoring their own deformity. The Trunk Aesthetic Clinical Evaluation (TRACE) score reported by Zaina et al [31] is an evolution from the Aesthetic Index [31]. The TRACE score is a combination of four different anatomical areas; the shoulders (looking at the differences in vertical height of the shoulders), the scapulae (looking at scapula prominence), the hemithorax (looking at rib prominence separate to the scapula assessment) and the waist (looking at waist asymmetry). The TRACE score has been used in the literature by the original authors in studies looking at the effects of bracing in scoliosis [96, 97].

2.3.2 Critique.

The literature on physical appearance is mixed. The literature that examines the relationship between mental health and physical appearance are not recent and are, in the main, retrospective in design of a small cohort of subjects [1, 2, 91]. Things are also complicated by the use of different methods of assessing mental health. The statistical analysis varies making anything other than a broad conclusion across all of the papers difficult. A valid conclusion would be that there is a relationship between AIS and mental health issues but more detailed conclusions would be difficult. However, the work of Donaldson [30] is useful as there is an identification of which body asymmetries cause concern.

The attempts to quantify and categorize the subject's own view of their overall body shape using different pictorial tools are again of different age and quality. Noting that there are several different methods reported for patient documentation of their own shape. This does suggest that none have been seen as entirely fulfilling the required need. This may well represent the inherent problems of taking the complex issue of the like or dislike of an aspect of body shape and forcing that view into a limited number of choices within a questionnaire.

Certainly, the author of this thesis does not entirely understand the different options that are to be assessed within the SAQ [94] and would struggle to complete it. The TAPS [93] score is a welcome simplification of the SAQ. The literature does not comment on further uses of the TRACE score [31] outside the paper that describes the measurement and this suggests that use outside the designer has been limited at best.

The methods used in the design of the WRVAS [95] are robust and demonstrate responsiveness of the parameter with increasing scoliosis curve size and an appropriate response when compared to planned or active treatment. This is particularly demonstrated with the 95% confidence intervals for the total score for the 'surgery recommended' to the 'post-surgical' groups. There is also no overlap of the mean or 95% confidence intervals between the scores of those with and without a noticeable deformity. The reliability, validity and responsiveness of the WRVAS, now known as the SAQ, is also assessed [94]. The methods described by Sanders et al [94] in the paper are thorough and the conclusion that the SAQ gives more information than the appearance domain of the SRS-22 is valid. The SRS-22 is the Scoliosis Research Society 22 question outcome questionnaire for measuring the effect of a scoliosis in four different domains – pain, function, mental health and self image [98] and has been the most widely used scoring questionnaire in scoliosis.

The methods used to validate the TAPS score [93] are well described in the paper in a cohort of 186 subjects. The scoring system is simpler than the SAQ and makes an assessment of the anterior aspect of the torso, discriminating between males and females. The TAPS score [93] is correlated in the paper with the appearance domain of the SRS-22, in a similar way as described for the SAQ with correlation values of between 0.43 and 0.54, only a moderate correlation at best.

Of note, there are no reports in the literature of a comparison of the SAQ [94], TAPS [93] or TRACE [31] scores with a quantitative measurement of torso shape.

In summary, there is no uniformly agreed method for a patient to document their own view of their deformity although there are several methods for this, both word and picture based. The SRS-22 [98] is a commonly used method (there are 432 papers with SRS-22 as a key word in an internet search in Pubmed via the The United States National Library of Medicine). The concern with the SRS-22 [98] is the scope of the four domains that need to be answered and discriminated within a small number of questions, particularly for the younger age groups [99]. As for the SAQ [94] and TAPS [93] scores, the TAPS score seems simpler (as noted previously). However, further work is required to investigate how the SAQ [94] and TAPS [93] scores relate to a quantitative measure of shape.

2.4 Methods of measuring surface shape with devices.

2.4.1 Scoliometer.

The simplest way of documenting surface shape and asymmetry of the back of the torso is by using a scoliometer [100]. The scoliometer is used when the subject is in the Adams forward bend position [33] and is straightforward to use. It is similar in nature to a spirit level and, when placed on the back, will allow measurement of the angle of trunk rotation (ATR). As such, the scoliometer has formed one of the main tools for historical school screening programmes [100] and research into torso shape [101, 102]. Bunnell concluded from his assessment of 1000 school-aged children that a small degree of difference between the two sides of the torso was common (ATR less than 3°) and was seen in 80% of the cohort [100]. He concluded that an ATR of 7° or more should be the trigger for further investigation looking for the presence of an underlying scoliosis. It is of note that the sensitivity and specificity of any screening examination for scoliosis in school children has been questioned and there is no current school screening performed in the UK [103].

The issue with the use of a scoliometer is the forward bend position, which is not an upright stance. The change in spinal shape that occurs with forward bend may change the shape of any underlying scoliosis. Making comparisons between shapes measured in the different body positions of forward bend and upright stance must be seen in the light of this caveat. The issue of how to measure the external shape of the torso in upright stance, the same posture as a radiograph, is addressed by surface topography techniques.

2.4.2 Surface topography techniques.

Within the study of human anatomy, topography is defined as 'the distribution of parts or features on the surface of or within an organ or organism' [104]. Techniques based on surface topography have been used for studying the external shape of the body in scoliosis for many years, since moiré fringes were proposed by Takasaki in 1970 [77] as a method of demonstrating the contours of the posterior torso. Asymmetries in shape lead to a visual difference in the number of contours seen over each side of the torso and the difference gives an objective measure of asymmetry. As a technique that does not use ionising radiation, this technique was, and is still, used as one of the methods of screening for scoliosis in schools [105]. Further systems have been developed since 1970 that capture the surface topography of the torso and have been used in scoliosis [78, 79, 106, 107, 108, 109, 110, 111, 112, 113, 114, 115, 116, 117, 118, 119, 120, 121, 122, 123, 124, 125, 126, 127, 128].

Turner-Smith et al [109] developed the Integrated Shape Imaging System (ISIS1) in Oxford in 1988. This system passes a horizontal blade of light down the back and this is 'seen' by a camera mounted at an angle relative to that of the light. A record of the 2D coordinates of the blade of light is captured and this is converted to a 3D surface for analysis. This system requires the identification of key anatomic landmarks that are marked with small stickers. These are the dimples of Venus, marking the posterior superior iliac spines inferiorly, the vertebra prominens, marking the upper thoracic spine superiorly and some of the spinous processes of the vertebrae in-between. The captured 3D data are analysed automatically to provide parameters describing the shape of the back. These are lateral asymmetry (LA), volumetric asymmetry (VA), coronal imbalance and a measure of the thoracic kyphosis and lumbar lordosis. LA is the ISIS equivalent of the Cobb angle, although it is noted by Turner-Smith et al [109] that there is not an exact relationship between the two measures. The initial step in determining LA is to identify the position of the centre of each vertebral body and through these centres, create a spine line marking the path of the vertebral column. The predicted centre of the vertebral body is found from the relationship of the angle of the skin over the paramedian areas from left to right including that over the spinous process (which is rotated away from the midline and the coronal plane by the scoliosis) and the coronal reference plane. The LA angle is then the angle subtended between the points of inflection in the spine line in a very similar fashion to the method of measurement for the Cobb angle. VA is a measure of the difference in the volume of one hemithorax in comparison to the other. The back is split into 10 transverse sections and the difference in the area of each section between the left and right sides of the back is summed and then divided by the length of the back. To allow for variation in the shape of a rib hump in a scoliotic subject, the parameter hump severity was created and this is the VA divided by the length of the back over which that VA was calculated. The image from ISIS1 took approximately 2 seconds to capture. The use of ISIS1 in both a normal and a scoliotic population has been described in the literature [91, 116, 123, 124, 125, 126].

The disadvantages of ISIS1 were related to the time required to acquire the image and the time required to process the image and provide the ISIS1 output. This was related to the techniques used with the blade of light, and the computing power available at the time. ISIS1 became obsolete as both of these issues changed and improved to become ISIS2 (discussed at length later in this review and in the Methods chapter).

Drerup and Hierholzer developed a similar system to ISIS1 in the 1990s using video rasterstereography that created a 3D reconstruction of spinal shape [122]. The system is described by Drerup as an improvement on ISIS1 as there is better resolution of the surface. This allows more advanced methods of analysis, in particular the identification of anatomical landmarks without the need for pre-marking with surface stickers and also, the faster acquisition of the surface image. This technology has developed into the Formetric system that is commercially available in the UK from Diers Medical [79]. It has been used to describe the results from scoliosis surgery [114, 115]. Other systems that have been used in the past include Quantec®, described, amongst others, by Oxborrow [127] and used by Sakka and Mehta [118, 119] and Goldberg [128], the Milwaukee Topographic scanner publicised by Thometz and Lyon [120, 121], the device used (but not named) by Pino-Almero et al [112], a laser based system reported by Poredos et al [117], the BIOMED-L® system reported on by De Korvin et al [111], the Vitus Smart 3D Body Scanner used by Gorton et al [113] and a non-invasive structured light method by Mínguez et al [106]. A group from Alberta, Canada have recently reported on the use of a full torso system (capturing the front and back of the torso at the same time) and they have used this to create asymmetry maps for the front and back of the torso [107, 108].

The Integrated Shape Imaging System 2 (ISIS2) was developed by Berryman and coworkers, and was first reported in 2008 [78]. This was an updated version of the ISIS1 system and uses the technique of Fourier Transform Profilometry to obtain 3D surface data from a 2D photograph. The similarities to ISIS remain in that stickers are applied to the back to mark the vertebra prominens, the dimples of Venus and some spinous processes. A full description of the workings of the ISIS2 system is found in Chapter 3 (the Methods chapter). The use of ISIS2 surface topography has been published in the literature since 2008 by both the designer [110, 129, 130, 131] and other units [132].

It is of interest to note that there are many different systems that attempt to address similar issues. Whilst this represents the gradual changing in technology over time, with improvements in technique and computing abilities, it must also be noted that the presence of many different systems does suggest that none of the systems are completely able to answer all of the questions on surface shape in AIS.

2.4.3 Methods of quantifying the amount of torso deformity.

There have been attempts to quantify the size and shape of the posterior torso using measurements derived from a surface topography technique. The most well known of these are the Posterior Trunk Asymmetry Index (POTSI) described by Suzuki et al [133] (which was further elaborated by Inami et al [134]), and the Suzuki hump sum [135]. The Suzuki hump sum is a sum of the rib humps in the posterior torso. Three measurements of rib hump height are made, one each at the thoracic, thoracolumbar and lumbar spines. At each level the difference in height between the left and right rib humps is divided by the width of the torso at that level and multiplied by 100. The three individual measurements are then summed (Figure 7).

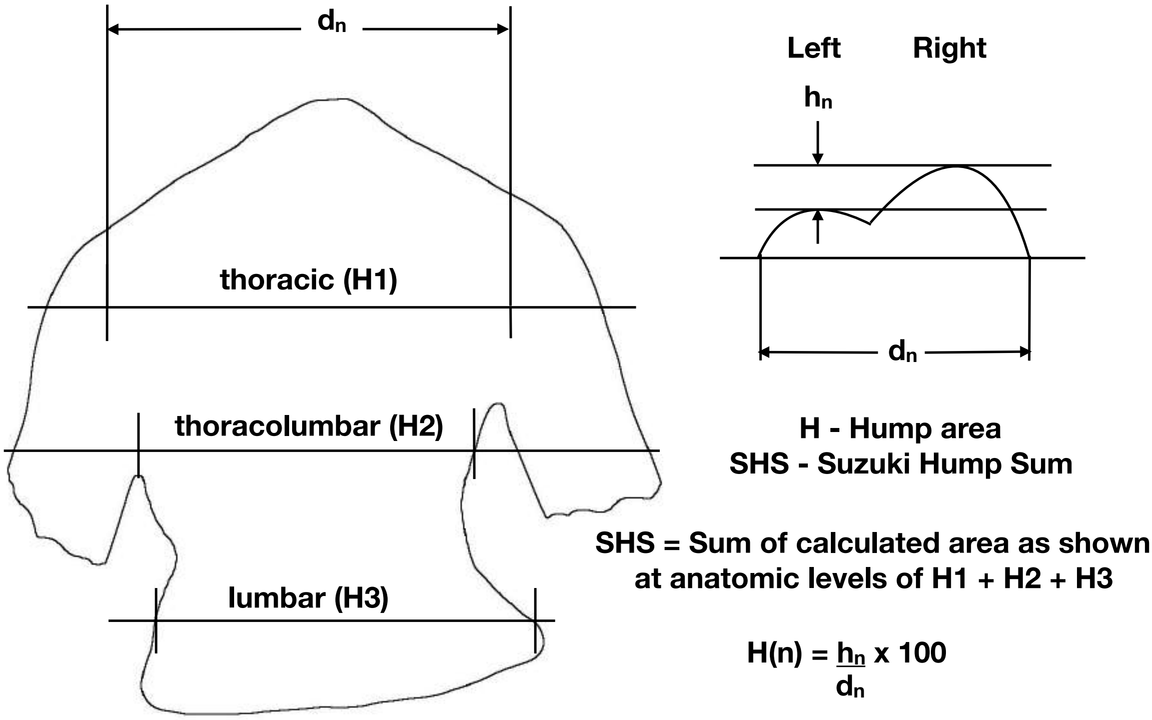


Figure 7: The calculation of the Suzuki Hump Sum redrawn from Patias et al [136].

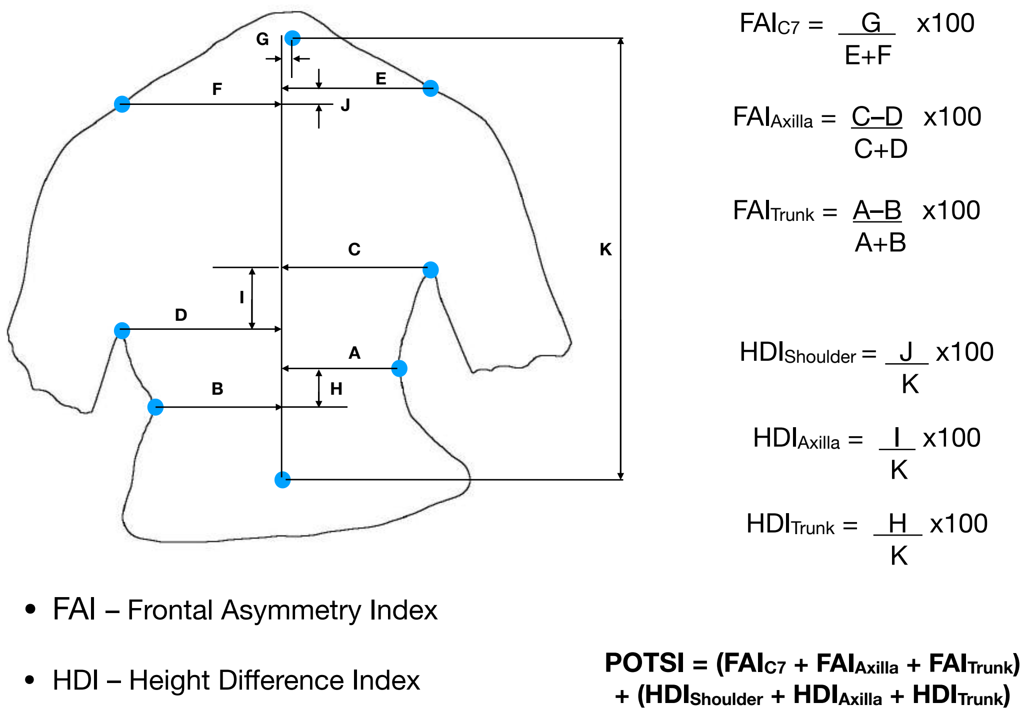


Figure 8: The POTSI score redrawn from Inami et al [134].

POTSI is a series of 2D measures taken from a moiré surface topography image in both the horizontal and vertical planes that are summed to give a total score (Figure 8). These measurements are the horizontal distance from the midline (defined as a vertical line that runs through the natal cleft) of the location of the C7 vertebral body (G), the difference between the distance from the midline to the edges of the body at the level of the most superior aspect of the posterior axillary fold (C–D) and the difference between the distance from the midline to the edge of the body at the most drawn in parts of the waist triangles (A–B). Note that each horizontal measure is divided by the total horizontal width of the body at the level of the anatomical measurement i.e. the level of the shoulders between the acromioclavicular joints (ACJ) for the C7 vertebral body (E+F), the distance between the axillary folds for the axillae (C+D) and the distance between the waist triangles for the waist (A+B). These figures are then made into percentages by multiplying by 100. The vertical measures are the difference in the height of the shoulders (as marked by the position of the ACJ) (J), the axillary folds (I) and the waist triangles (H). All of these measures are divided by the total vertical height between the level of C7 vertebral body and the most superior part of the natal cleft (K). Again the figures are then multiplied by 100. A larger score reflects greater asymmetry, but where that asymmetry is within the torso cannot be defined from the single number provided.

When reported with the Suzuki hump sum, a 3D measure of torso deformity is made. The POTSI score has been used and reported in the literature [106, 112, 137, 138]. Asher et al [137] used the POTSI score in 2002 to show that in a group of 45 patients operated on for scoliosis by one surgeon in one institution, the POTSI score had little correlation with the patient's own evaluation of their deformity as measured using the SRS-22 [98] outcome questionnaire, but that the thoracic component of the Suzuki hump sum was correlated to some domains of the SRS-22 (function and self image). Asher notes that this was a surprising result and suggests that objective measures of the back of the torso do not necessarily relate to what a patient sees of the front of their body in the mirror. Mínguez et al [106] use both the POTSI and DAPI (Deformity in the Axial Plane Index - reviewed below) as tools to explore the utility of their new surface topography method. The Denmark group [138] looked at 24 subjects with right thoracic scoliosis curves with significant thoracolumbar compensatory curves, operated on using a posterior pedicle screw technique, and assessed cosmesis, trunk shift and patient derived outcomes using the SRS-30 and EQ-5D outcome questionnaires. The results showed that, at follow up, POTSI measures correlated well with the mental health domain of the SRS-30 and partially with the other domains of self-image, pain and function. A similar method to the POTSI score is the Deformity in an Axial Plane Index (DAPI) described by Mínguez et al [106]. The DAPI score is calculated through the sum of the differences in the direct distances (i.e. not horizontal or vertical) between the most prominent points over the scapulae (B) and the least prominent points over the waist (C). Before addition the measured differences are multiplied by 100 and divided by the length of the line between the C7 vertebral body and the most superior point of the gluteal cleft (A) (Figure 9). The paper of Mínguez compares the POTSI and DAPI scores for a series of non scoliotic controls (n=56) and scoliotic subjects (n=30) and suggests that there is a strong correlation of both measures to changes in the Cobb angle with r^2 values of 44.6% for the relationship of POTSI and Cobb angle and 49.8% between DAPI and Cobb angle. Pino-Almero et al [112] compared POTSI and DHOPI (horizontal plane deformity index) to radiographs of 88 subjects with scoliosis to correlate surface topographical parameters against radiographic measures of kyphosis, lordosis and scoliosis and showed moderate positive correlation. For reasons that are not explained in the paper by Pino-Almero [112], the DHOPI measure is named the horizontal plane deformity index but it is the same measure as the DAPI and references the method to the original description of Mínguez et al [106].

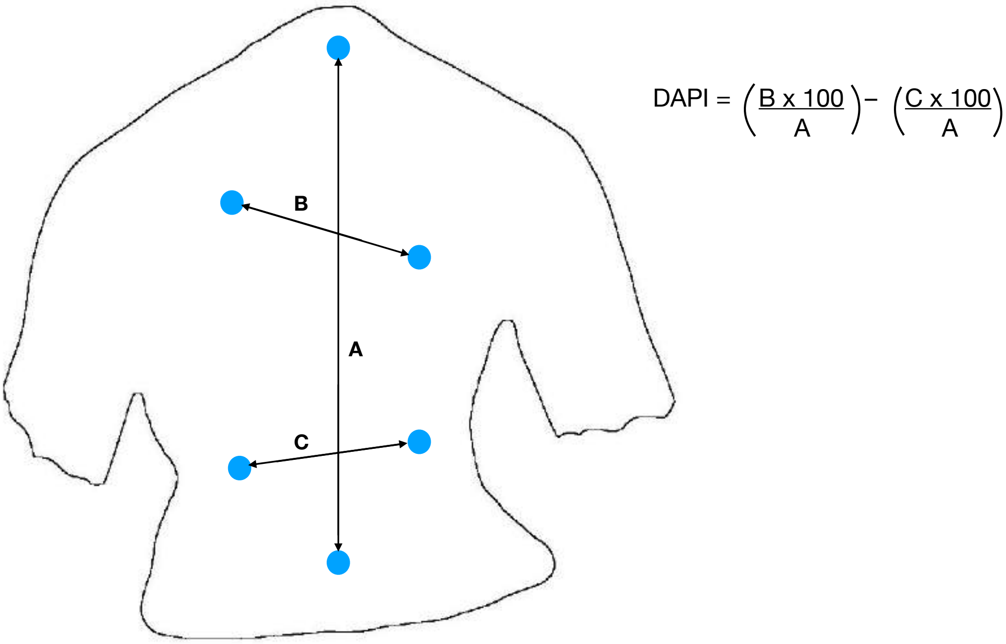


Figure 9: The measurements made for the DAPI parameter redrawn from Mínguez et al [106].

Both the POTSI [134] and DAPI [106] scores are attempts to describe the shape of the torso in 2D through mathematical formulae, which act to standardise the effects of different body shapes and sizes between subjects, making comparison across a population with AIS possible. The Suzuki hump sum [135] is a method of describing the 3D shape, and consequently the POTSI score and the Suzuki hump sum are often quoted together [137]. The downsides of the POTSI and DAPI scores are inherent to their design. There is no absolute measure to compare to, rather a number that describes the whole surface. This number does not come with any pictorial information and so the number provided is only useful in comparison to numbers from others. It cannot be picked apart for individual parameters. The Suzuki hump sum [135] again follows the same mathematical principles, creating a number that describes 3D shape. The individual components of the Suzuki mathematical construct are only three measures on the back and are not defined in anatomical location. Whilst it is important to allow any measure of 3D asymmetry to measure the most asymmetric area, rather than a fixed anatomical point which may not be the most deformed point, there is inherent variability here.

The POTSI [134] score is complex to use, as demonstrated in the convoluted description previously seen in this text (Figure 8), which also reflects the original description [134]. This might well explain the limited use of the technique in the literature to date by a small number of authors [106, 134, 135, 137, 139]. This issue is also seen with the DAPI [106] score, and raises issues around the clinical utility of the methods for the measurement of torso asymmetry from the surface topography techniques from which they arise.

The literature that does use the POTSI [134] or DAPI [106] scores and the Suzuki hump sum [135] are limited to small series of patients and the use of correlation for statistical analysis. There have been limited attempts to measure POTSI score from cohorts of children without spinal deformity [106, 135, 139].

However, the anatomical points used in the POTSI [134] score are those identified as of greatest concern to those with AIS [30] and so the score does have a basis with regard to patient concern.

2.4.4 Other methods of measurement of spinal shape.

Other methods have been reported for the monitoring and measurement of the size of the curve in scoliosis. Scolioscan is an ultrasound method that is performed in the upright position [140]. The ultrasound probe is passed down the spine and an image of the spine is created. The size of the scoliosis is identified through measurement of the angle between the most angulated vertebra at the top and bottom of the curve. The method used for this measurement is not clearly demonstrated in the paper, although the figures suggest that it is the Ferguson method [34] that is used rather than the Cobb method [7]. There is an accepted difference between these two methods in the literature [141] (although it is noted that the Ferguson method would be better suited in an automated measurement system [142]). The Bland Altman plots in this ultrasound paper suggest limits of agreement of $\pm$ 10° when comparing the measured Cobb angle from a radiograph and the angle derived from this ultrasound method. Further comment on the utility of this technique is not possible given the small number of publications using the technology. The advantage is the ability to measure spinal shape in upright stance without the use of ionising radiation. The difference in measurements of $\pm$ 10° would lead to a similar situation as that of surface topography, in that it would not be advised to use the methods interchangeably.

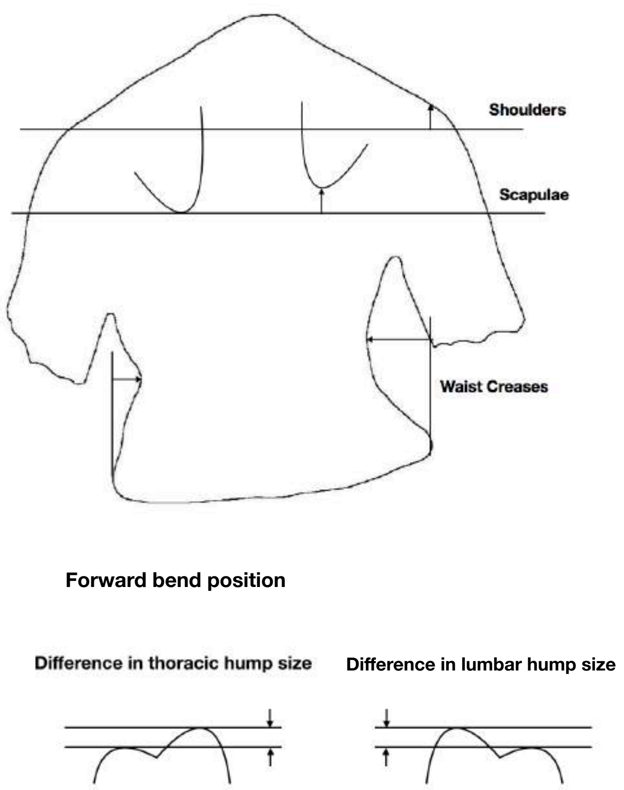
The use of MRI scanning is also described to monitor a scoliosis and, with the advent of the upright MRI scanner, the patient can be imaged in the weight bearing position similar and thus comparable to an upright radiograph [143]. Diefenbach et al [143] correlated the measured Cobb angles from both postero-anterior and lateral whole spine radiographs with the equivalent angles from an upright MRI taken within 7 days of each other. The acquisition time for the MRI sequences used was around 7 minutes. There was a high correlation for the coronal and lateral Cobb angles between the two imaging modalities. Intervertebral rotation was also measured from the MRI scans. This MRI paper concludes this radiation free method can be used to monitor scoliosis and goes on to address the issues of cost and time required for the MRI scan. The limitations to the technique are acknowledged and these include contraindications to an MRI scan (some metallic foreign bodies) and the inability to remain motionless for the time required. In the UK, access to the small numbers of upright MRI scanners via the NHS is strictly controlled for very specific indications. Monitoring of scoliosis is not one of those indications so this technique is not currently available in the UK within the NHS.

2.5 Normal torso asymmetry.

2.5.1 Description.

Previous literature has demonstrated that there is a range of body shapes that class as 'normal' [101, 102, 106, 134, 144, 145, 146, 147, 148, 149, 150, 151, 152, 153, 154, 155, 156, 157]. Willner [152] in his 1984 study examined 6464 children from Malmo, Sweden. Those who had a visible deformity of the spine or torso, in either erect stance or in the forward bend position, were then examined with moiré surface topography. A smaller subset of this latter group then had radiographs taken. Of the original 6464 children screened (3286 males and 3178 females), asymmetries were seen in 14.4% of males and 18.9% of females. Those who had a trunk asymmetry of greater than 1 cm in height comprised 1.2% of males and 2.4% of females. The method of measurement is described as a visual assessment without actual measurement. Of particular interest were those with visible asymmetry who did not show an asymmetric pattern of moiré contours, this was made up of 10% of boys and 14% of girls. In the opinion of Willner and co-workers, this excluded a diagnosis of scoliosis. Of the group that was left, 6.2% of boys and 8.3% of girls had an asymmetry less than one moiré contour, 6.2% of boys and 7.0% of girls had an asymmetry of between one and two moiré contours and 0.6% of boys and 0.9% of girls had asymmetry of greater than two moiré contours. In the children who subsequently had a radiograph taken, those with an asymmetry of less than one moiré contour and a small hump (less than 1 cm) had a coronal deviation in the spine, but no Cobb angle measured more than 6° and was thus classed as a physiological lean. In those with an asymmetry of more than one moiré contour and a hump of less than 1 cm in height, the Cobb angle on the radiograph was no larger than 11°. Larger hump heights and increasing moiré contour asymmetry led to greater Cobb angles and a diagnosis of scoliosis. The study concludes that an asymmetry in shape of less than 1 cm in hump height and less than one moiré contour difference is classed as physiological variation across the population and is not a precursor to developing scoliosis, requiring no further investigation or follow up.

Also in 1982, Vercauteren and colleagues [144] examined 270 Belgian school children at their annual school medical checks. The authors quote ranges that describe physiological, postural and structural asymmetry based on their findings. The children were measured in the erect and forward bend positions and assessments were made of the difference in vertical height of the shoulders (measured at the acromion and the inferior pole of the scapula), the maximum depth of the waist creases and the size of any thoracic rib hump or lumbar prominence (measured in the forward bend position) (Figure 10). Physiological asymmetry is defined as shoulder height difference of less than 1 cm at the acromion and inferior pole of the scapula, a waist crease difference of less than 1.5 cm, a rib hump of less than 0.8 cm and a lumbar hump of less than 0.5 cm (Figure 10). Structural asymmetry is defined as measures equal to or greater than the upper limit defined as physiologic asymmetry. Postural asymmetry is defined as a measure greater than the upper limit of physiological asymmetry (shoulder height, scapula height or waist crease differences), but not greater for rib or lumbar hump size which remain the same as is defined for physiologic asymmetry. The thrust of the Vercauteren paper is that structural asymmetry is found in the presence of increasing vertebral rotation, demonstrated by a greater rib hump or lumbar prominence. In the absence of this rotation there is then only a physiological or postural asymmetry and this is seen through the measures of shoulder height, scapula height and waist triangle depth.



	Physiologic asymmetry	Postural asymmetry	Structural asymmetry
Shoulder height difference	≤ 1 cm	> 1 cm	> 1 cm
Scapula height difference	≤ 1 cm	> 1 cm	> 1 cm
Waist triangle depth difference	≤ 1.5 cm	> 1.5 cm	> 1.5 cm
Rib hump	≤ 0.8 cm	≤ 0.8 cm	> 0.8 cm
Lumbar prominence	≤ 0.5 cm	≤ 0.5 cm	> 0.5 cm

Figure 10: The measurements made by Vercauteren et al [144] including a table defining physiologic, postural and structural asymmetry.

Burwell in his paper of 1983 [153] examined a group of 636 school children (315 males and 321 females) and used a combination of clinical examination in the upright position and the objective measurement of the size of any asymmetry using a ‘formulator body contour tracer’. This was a device allowing the actual shape of the back in a transverse plane to be transferred from the volunteer to be traced onto paper for analysis. The shape of the back was captured at the spinal levels T4, T8, T12, L3, and S2 with the child in the Adams forward bend position [33]. The raw data were standardised using the mean chest diameter of the entire cohort (21 cm), thus creating standardised trunk assessment scores (to allow comparison between children of different ages and body sizes). The angle of trunk inclination was also calculated. The main conclusions of the paper were that 25% of children had a rib or lumbar hump. Rib humps were more common in girls than boys (1.2 to 1 for thoracic rib humps and 1.4 to 1 for lumbar humps) and that right sided thoracic humps were 10 times more common than left. At the levels of T8, T12 and L3 in both males and females, the mean shape was an asymmetry higher on the right than the left, although the spread of the data through one standard deviation at all levels crossed the point of symmetry following a Gaussian distribution. Of the entire cohort, 51 (8%) children had spinal radiographs following their participation in the study. There were no curves found that were greater than 19° and most demonstrated that the rib hump and the spinal rotation was in the same direction. In 7 children the direction of the spine rotation and the rib hump was opposite to each other. Burwell et al concluded that 'measures of asymmetry' were seen across the group who clinically had no evidence of scoliosis in the forward bend position. These conclusions were very similar to that of Vercauteren et al [144].

Nissinen and co-workers have published three papers on a group of children from Helsinki [149, 150, 151]. In 1989, their first paper [149] reported on 1060 children (515 males and 545 females) with a mean age of 10.8 years, assessed for both trunk and spine deformity in the forward bend position. The rib hump was measured as the vertical height between the two sides of the torso using a water scale in progressively increasing amounts of flexion, and termed as the angle of trunk rotation (ATR). From the figure in the Nissinen paper [149], the water scale is a type of spirit level device for accurately identifying the horizontal. Assessment was also made of differences in limb length of both the arms and the legs, sitting and standing height, BMI, pubertal stage and trunk asymmetry using moiré contours. Radiographs of the spine were taken for those children with a rib hump of 6 mm or more. There was a rib hump of between 1 and 5 mm in 61%

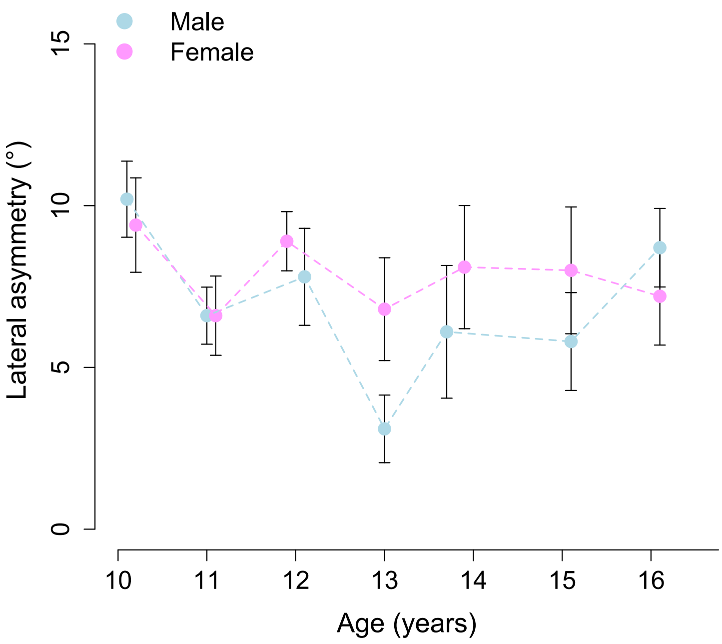
of all of the children with 20% of children having no rib hump and 19% having a hump of greater than 6 mm. Large humps (greater than 6 mm) were seen more frequently in girls than boys. The ATR range measured between 0-20° with a mean value of 5° in girls and 4° in boys. Differences in the pattern of moiré contours were common although the differences seen in the moiré contours did not seem to change between those children with a large rib hump with or without scoliosis and the rest of the cohort. In those that had a radiograph, the correlation between the moiré contour differences and the Cobb angle was poor. Spinal curves were seen in 5.6% of girls and 2.6% of boys with a mean Cobb angle of 7°. Of those who had a radiograph with differences in the moiré contours indicating trunk asymmetry, 9.6% had a straight spine.

The follow up paper of 1993 [150] was a re-examination of the same cohort but with a drop out rate of 19.3%. There were 855 children reattending for examination with a mean age of 13.8 years. Of the cohort, the majority had a degree of trunk asymmetry, with 47.3% having a right sided thoracic hump, 30.4% having a left sided thoracic hump and 22.2% being symmetrical. In the thoracolumbar spine, 50.1% had a left sided lumbar hump, 31.0% had a right sided lumbar hump and 18.9% were symmetrical. The side of the rib hump and lumbar hump were the same in over 80%, with a right sided rib hump being seen with a right sided lumbar hump and vice versa. The prevalence of scoliosis in this group was 9.2% for any spinal curves with a Cobb angle of greater than 10°. A small proportion (2.4%) of the group had no scoliosis on radiographs despite having a rib hump of greater than 8 mm. The 1993 paper notes that trunk asymmetry occurs during the pubertal growth spurt and that a rib hump of 6 mm or greater is an appropriate value for a radiograph querying the diagnosis of underlying scoliosis.

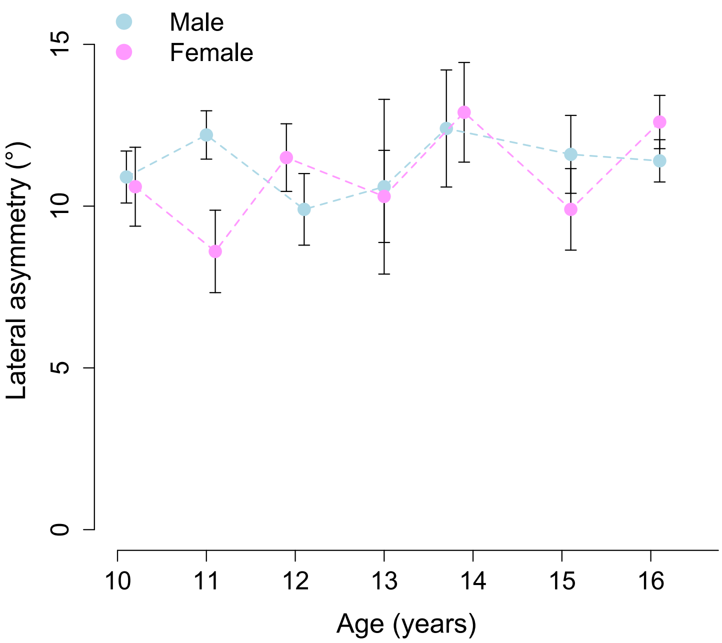
The final paper of 2000 [151] was a long term follow up of the original cohort. The participation rate was 54% of the original 1060 children, comprising 208 women and 222 men with a mean age of 22 years. Of the cohort, 30% showed no signs of asymmetry, defined as a rib hump of less than 4 mm in the forward bend position. Larger rib humps were seen, with 51% having a hump of between 4 and 9 mm and 19% with a rib hump of 10 mm or greater. There was a predominance for curves to be seen on the right side, as in the 1993 paper. There was no difference between the number with asymmetry between males and females. Interestingly, the side of the rib hump changed between puberty and adult life in a small proportion with 10% changing from a right to a left sided thoracic hump and 19% changing from a left to a right sided thoracic hump. This is also seen in lumbar humps with 10% left to right change and 16% right to left. Over the period between puberty and adulthood the proportion of large humps increased and this was seen during the pubertal growth spurt. Those with a large hump post growth spurt maintained this into adult life and this was found to be the most significant predictor of major torso asymmetry in adult life.

This series of three papers establishes a large cohort of prepubertal children, revisits them during and after the adolescent growth spurt and then again in early adult life. The papers demonstrate that trunk asymmetry in both the thoracic and lumbar regions is common and becomes more common during growth. The number who are truly symmetric is small. The side of the hump can change over time. Humps that are classed as large following puberty remain large into adult life. Most of those who have trunk asymmetry do not have scoliosis, although the larger the hump, the greater the possibility of underlying scoliosis.

Using the ISIS1 system, Carr et al measured the surface topography of 271 children (135 males and 136 females) without scoliosis between the ages of 10 and 16 years [145]. Whilst a cross sectional study by design, the data presented can be reanalysed and viewed in a longitudinal manner (Figures 11, 12 and 13). This reveals that in males LA is larger on the right than the left and this is also seen with VA. Over time, right VA increases in males whilst left does not. LA does not change over time. Maximal surface angle does not change over time. In females, right LA is again larger than left and gets larger with increasing age. Right VA is also larger than left but does not increase with time. Maximum surface angle remains constant throughout.

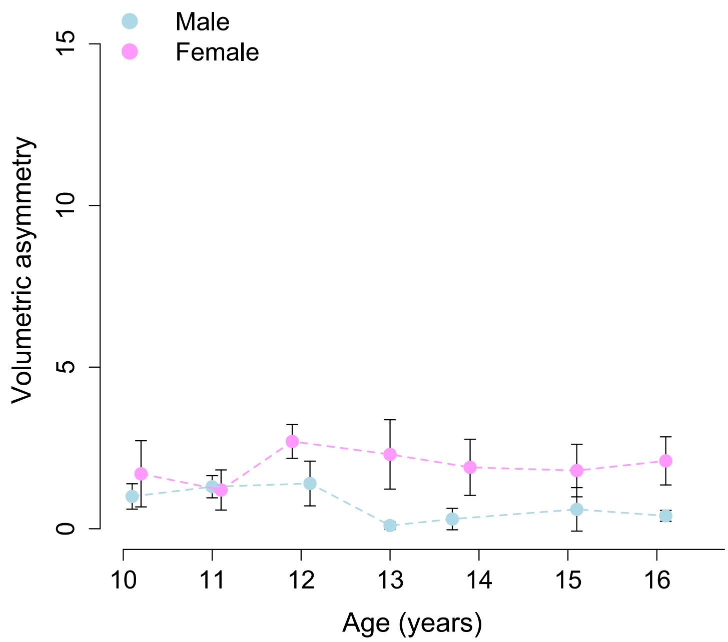


A.

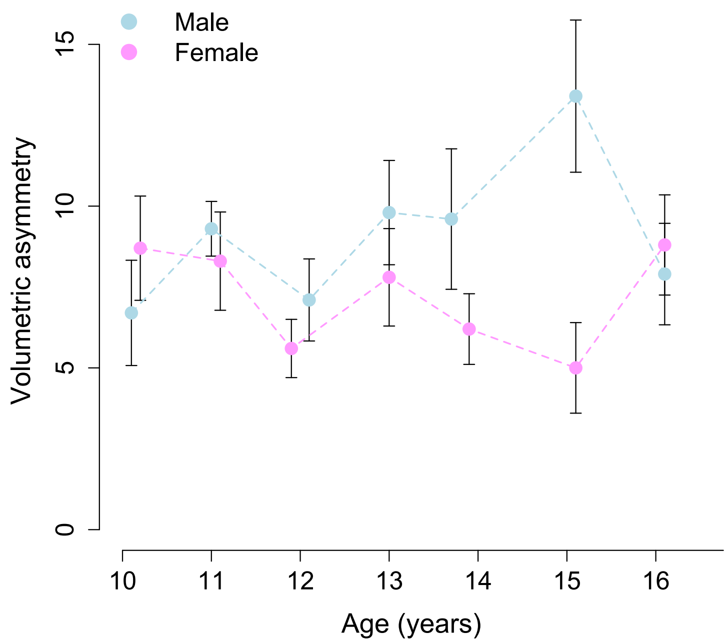


B.

Figure 11: The lateral asymmetry (°) for left (A) and right (B) sides of the back, viewed longitudinally. Redrawn from the data in Carr et al [145].



A.



B.

Figure 12: The volumetric asymmetry for left (A) and right (B) sides of the back, viewed longitudinally. Redrawn from the data in Carr et al [145].

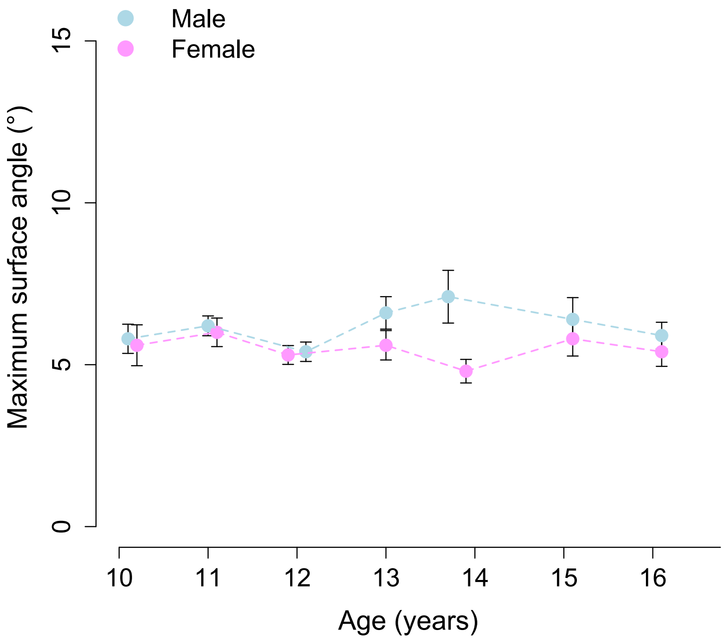


Figure 13: The maximum surface angle (°) viewed longitudinally. Redrawn from the data in Carr et al [145].

There have been some large cohort studies using a scoliometer to measure the size of rib humps reported from the school screening programmes in Greece [101, 102]. In the paper published in 2006, [102], measures were taken of 2071 children between the ages of 5 and 18 years of age in the standing and sitting forward bend positions measuring the angle of trunk rotation (ATR). Measures were taken over the central thoracic spine (T4 to T8), the thoracolumbar spine (T12 to L1) and the lumbar spine (L3 to L5). Their results divided the cohort into three groups; symmetry with an ATR of 0°, mild asymmetry with an ATR of less than 7° and more significant asymmetry with an ATR of 7° or more. A positive number indicated the right side was bigger than the left, a negative number the reverse. The results show that most of the cohort were symmetrical (approximately 80%). Asymmetry occurs to both the right and left sides of the body. As the degree of asymmetry increases the number with that degree of asymmetry decreases. In the 2008 paper [101], the data is presented in such a way as to allow reanalysis in a longitudinal fashion, in a similar way to the paper of Carr et al [145]. As the children increase in age, the degree of asymmetry also increases (Figures 14 to 16 where minus one is an ATR to the left of between 1° and 6°, minus two is an ATR of 7° or more to the left, plus one is an ATR to the right of between 1° and 6°, plus two is an ATR of 7° or more to the right). Right side asymmetry is more common than that on the left side. When comparing the scoliometer readings between the sitting and standing positions, standing increases the angle over sitting as shown by an increase of the number of subjects with asymmetry.

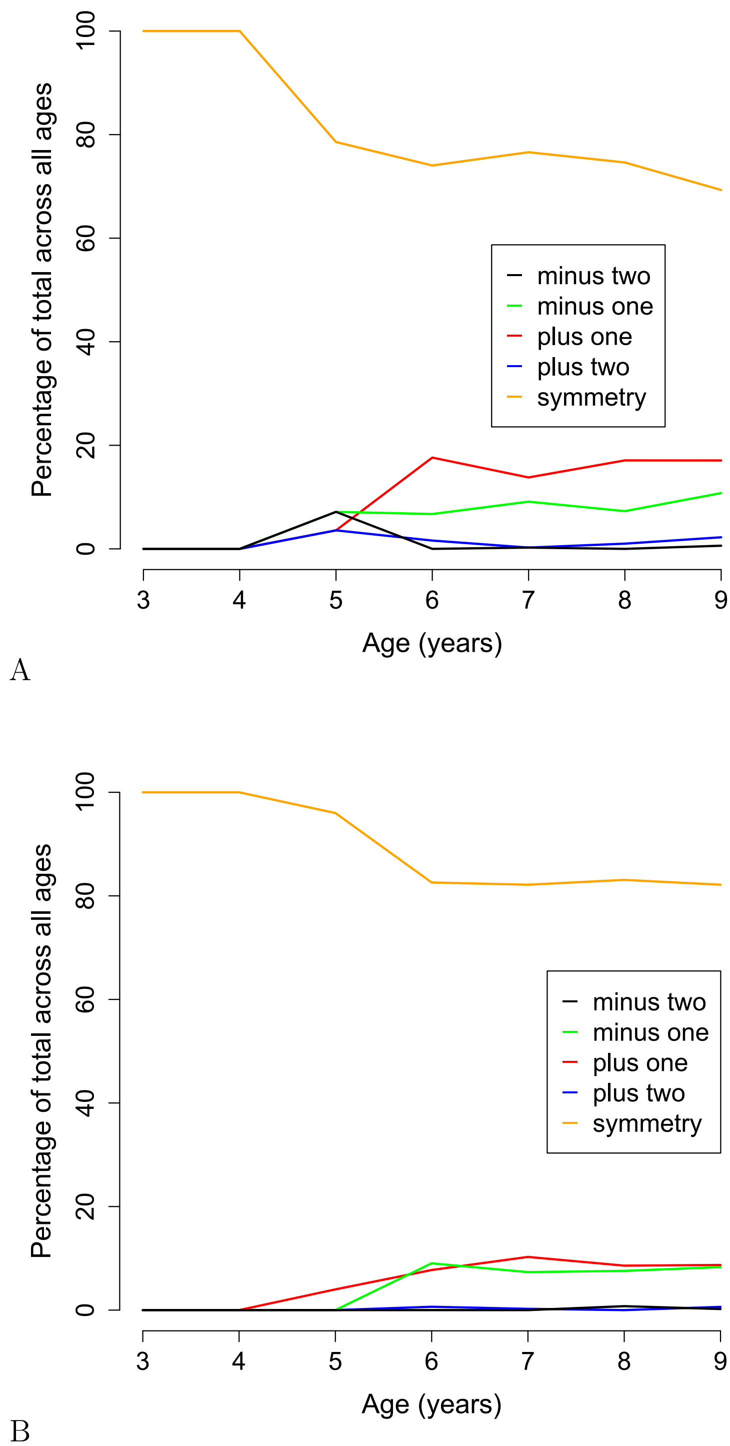
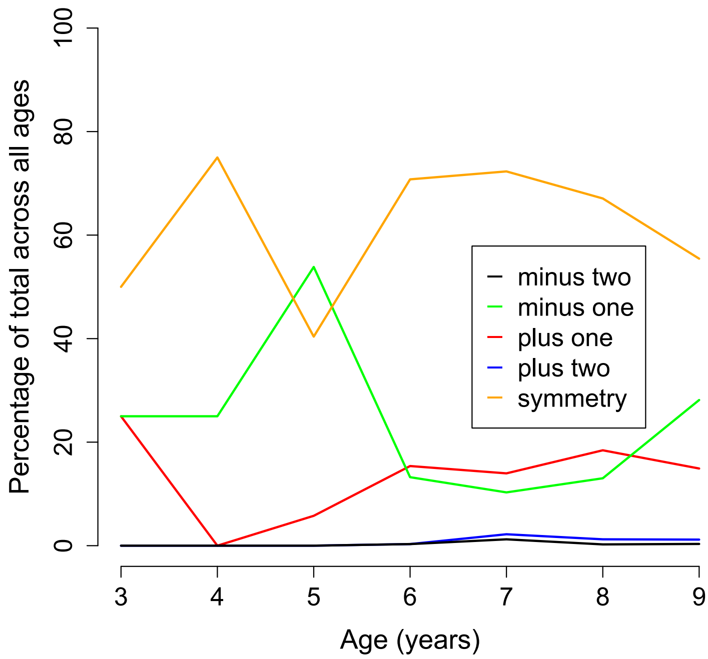
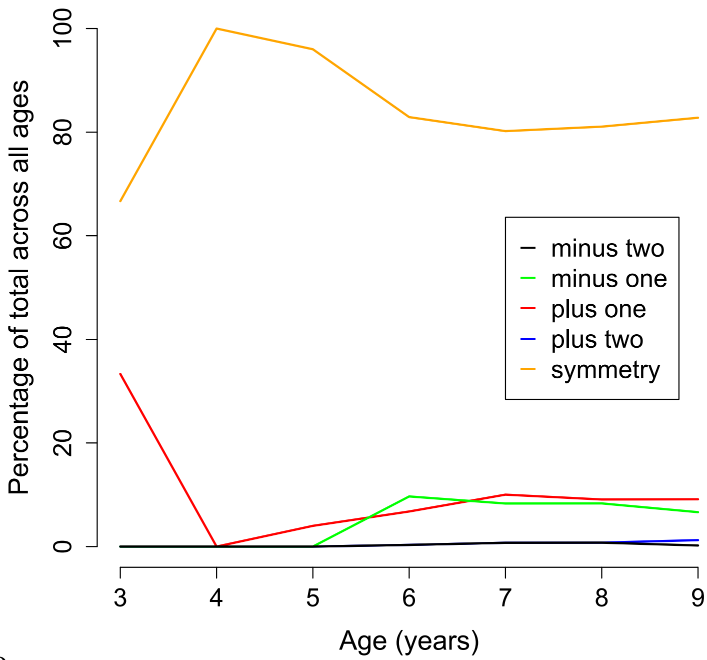


Figure 14: The percentage of the cohort with differing degrees of asymmetry versus age (years) for the thoracic spine in males (A) and females (B). Redrawn from Grivas et al [101].

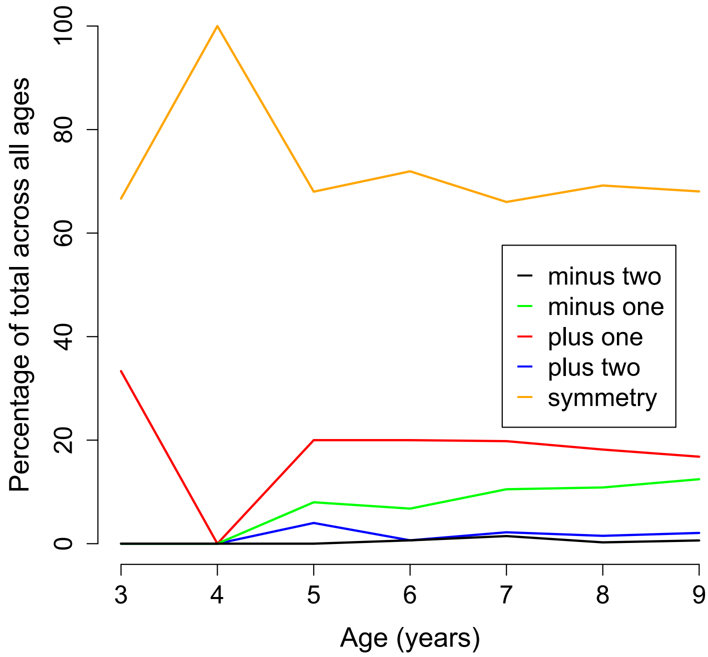


A

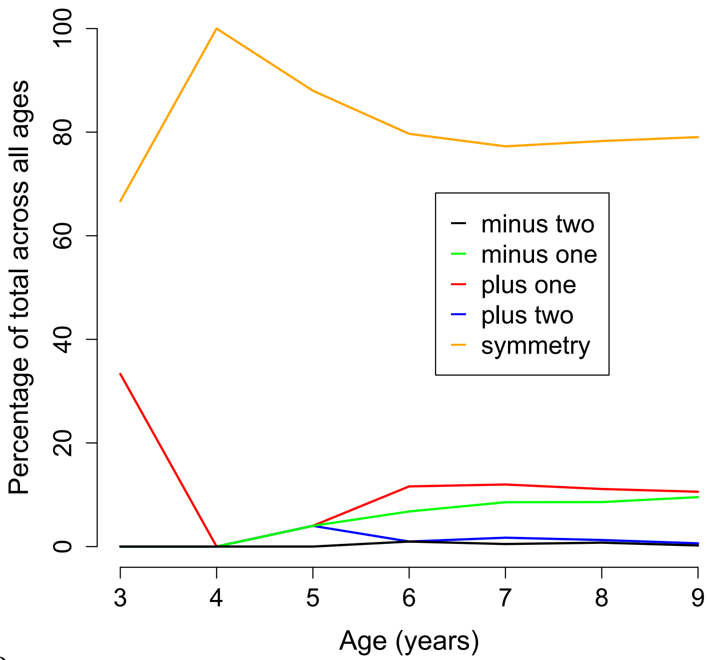


B

Figure 15: The percentage of the cohort with differing degrees of asymmetry versus age (years) for the thoracolumbar spine in males (A) and females (B). Redrawn from Grivas et al [101].



A



B

Figure 16: The percentage of the cohort with differing degrees of asymmetry versus age (years) for the lumbar spine in males (A) and females (B). Redrawn from Grivas et al [101].

Inami et al [134] used moiré contours to examine the surface shape of 55 children judged as non-scoliotic from a school screening programme. There were 52 females and 3 males of average age 14.5 years. The POTSI score was calculated along with the Suzuki hump sum. The mean POTSI score was 16.5 $\pm$ 8.2 with a Suzuki hump sum of 3.8 $\pm$ 2.2.

The POTSI and DAPI parameters have also been calculated for a series of non-scoliotic controls by Mínguez et al [106]. This group was seen to have a POTSI score of up to 30 and a DAPI score of up to 4. The Mínguez paper concludes that POTSI scores less than or equal to 27.9% or DAPI scores less than or equal to 3.9% would not be classed as pathological.

More recently, Ho et al [146] used a method that captured the whole torso, giving the surface topography of both the anterior and posterior aspects of the torso. The cohort consisted of 83 subjects (42 males and 41 females) between the ages of 10 and 18 years. The anterior and posterior images were matched together and analysed as a whole torso. There were two different patterns of asymmetry that were observed, 'thickness' and 'twist'. This was demonstrated through the use of colour maps where different depths of colour (red for distance away from and blue towards a coronal plane through the centre of the body) represented the shape of the torso. By comparing both aspects of the torso, thickness was defined as an increase away from the coronal plane in both the anterior and posterior aspects of a particular anatomical area, for example, the pectoralis region. Twist was defined as prominence on one side of the body (either anterior or posterior) but not on the other side, suggesting no increase in overall tissue but movement of that region relative to the coronal plane. The paper went on to note that each torso can have several areas of either thickness or twist in up to four anatomical areas (defined as the shoulder area, the pectoral or breast area, the abdominal or rectus area and the oblique area). There are no quantitative results presented in the paper, rather patterns of shape.

Other features of torso asymmetry have been reported in the literature. Akel et al [148] took clinical photographs in 91 children without spinal pathology who were attending for a chest radiograph for an unrelated problem. This study looked at the variation in shoulder height in this normal paediatric population, all of whom stated in advance that their shoulders were level. Measurements were taken directly from the photograph. It was shown that an equal shoulder height was seen in only 17 volunteers (18.7%). The mean difference in shoulder height was 7.5 ± 5.8 mm.

Documenting what is normal around the position of the waist is made more difficult by a variety of definitions of what classes as the waist [154, 155], which depends partly on why it is being measured. As noted previously, Vercauteren [144] notes physiological waist crease measurements as the difference in the horizontal depth of the left and right waist creases made from a vertical line lateral to the body (i.e. from the outside in) as less than or equal to 1.5 cm, called 'depth discrepancy of waist triangles'. A concept defined by Mason and Katzmarzyk [157] is that of the minimal waist, namely the smallest circumference around the waist. This is based on information drawn from published anthropometric standards [156]. Standards exist for waist circumference for children with centile charts [147] but not for the distance to the waist crease from the midline of the body.

2.5.2 Critique.

The literature on torso shape in those without spinal deformity is varied. There are different techniques used for measuring different parameters which vary from paper to paper. This, in part, reflects the time over which the literature has been written and the investigations performed which have changed as technology has improved. Certainly, the estimations of rib hump height made in the forward bend position by Willner [152] are very different to the colour maps documenting the shape of the entire torso in upright stance published by Ho et al [146]. Making a comparison between these papers becomes difficult due to the problems of identifying parameters with the same definitions between the studies. In particular is the measurement of rib hump height where the techniques of measurement, the anatomical location that measure is taken in along with the position the subject is placed for the measure all vary [101, 102, 149, 150, 151, 152, 153].

The paper by Vercauteren [144] is of more interest because there are absolute values supplied for a number of torso parameters in both upright stance and a forward bend position. There is no standardisation of these parameters across the cohort to allow for different sizes and shapes of children, and so the parameters must be viewed in this light. The conclusion drawn by Vercauteren [144] that asymmetry can be dividied in to physiological, postural and structural types causes some issues to arise. There is no definition of what the difference is between physiological or postural, other than stating that they 'occur so commonly as to be considered physiological' [144]. Table 1 of the paper demonstrates that the difference between postural and structural asymmetries is the amount of rib hump or lumbar prominence. The assumption is made that rib humps and lumbar prominences are related to vertebral body rotation caused by an underlying scoliosis. The method of measurement of the rib humps and lumbar prominences is a measure of height, not a measure of rotation.The papers that could have been referenced to help define the boundary of physiological / postural and structural are those of Nissinen et al [149, 150, 151]. The Nissinen papers [149, 150, 151] are more recent than the Vercauteren paper [144] and so understandably are not referenced by Vercauteren. Observation of the papers together shows that the degree of rib hump named structural by Vercauteren [144] lies close to the range noted as seen in the normal cohort reported on by Nissinen [149, 150, 151] and raises the concern that the boundary to structural change defined by Vercauteren may be incorrect. The technique described by Burwell

[153] has not been repeated in the literature and is complicated, slow and operator dependent.

Furthermore, there is the issue of how any of these parameters changes over time, especially over the rapid change of the adolescent growth spurt. To present all ages together would lose any change caused through the effect of the different ages. Only the series of papers by Nissinen [149, 150, 151] try to observe change over time, and thus with growth. Unfortunately this is by returning to the cohort on only two occasions after the initial enrollment, where a large number of the original participants are no longer involved. No attempt is made to deal with this loss of data, other than acknowledging it. The concern this raises is that the conclusions drawn from the series of papers may be flawed due to a selection bias. Of particular concern is the observation of a change in sides of the rib hump seen in some of the cohort in the final paper at 11 years post enrollment. This observation is not repeated elsewhere and could be a result of this bias.

Of note, the same issue of change within the study cohort through sex is poorly addressed within the literature, with the results of both sexes being reported together. Given the anatomical changes of adolescence that exist between males and females, to not subdivide the results for sex raises concern over whether there is a difference that is not being identified within the data.

Carr et al [145] used a cross sectional study design which, if analysed longitudinally, does potentially lead to inappropriate conclusions being drawn. However, the study by Carr [145] used the ISIS1 technology which makes the results comparable to those gained using ISIS2 and thus influenced the study design for this thesis. This is because the results show change in the ISIS1 parameters with age. This will be investigated further within this thesis using more modern longitudinal analysis techniques.

The papers by Grivas [101, 102] are of interest as there is a large number of subjects. These subjects are measured using a scoliometer in the forward bend position, both standing and sitting. Whilst this is interesting and demonstrates a measure of asymmetry, the same issues concerning the use of the scoliometer exist, as previously highlighted in this literature review, are seen. However the common features between the papers by Grivas [101, 102] and those of Nissinen [149, 150, 151] and Carr [145] is the greater number of subjects with a right sided prominence. Within the 2008 paper of Grivas [101] there is a high number of subjects recorded as being symmetric and not having any asymmetry. This seems at odds with the results of Nissinen [149, 150, 151]. Why there is a difference is not clear, and whether this is because of a difference in the populations being measured or down to differences in the techniques of measurement remains unclear. Again, these observations influenced the development of the work reported in this thesis.

The recent paper by Ho [146] uses a different philosophy of measurement and does not quote any numerical values for any parameters. The imaging of the entire torso, both the anterior and posterior aspects, is novel and the paper by Ho [146] shows that there is a link between the two. Unfortunately, without further details of the mechanics of the system and software used for analysis, it is not possible to repeat the observations or take them further.

The paper by Akel [148] is interesting and relevant, as it is the only paper that relates a measure of shape back to the subjects view of that shape. In this case, the difference of shoulder height is measured and a mean and standard deviation is quoted. This is a particularly useful figure as, given that all of the children involved believed that their shoulders were level, the value for the difference in shoulder height gives a figure at which the subject is unaware of the asymmetry that is present. This information allows the assessment of the 'minimally important clinical difference' (MCID). This is a concept that draws a distinction between statistical significance which may not be apparent or relevant to the subject, and the smallest change that means something to the subject [158]. The range of shoulder height difference quoted by Akel et al [148] is 7.5 ± 5.8 mm. A conclusion that could reasonably be drawn from this is that children without spinal deformity are unaware of a shoulder height difference of approximately 10 mm. Similar data for other measures of variability of body shape including waist height and scapular position does exist within the literature, and 10 mm is the figure for the same parameter in Vercauteren et al [144]. Similar figures that describe this variability for these other parameters are only really quoted by Vercauteren et al [144] and are between 10 and 15 mm. Consequently, to remove any ambiguity, a value of 10 mm as the MCID for points around the torso, such as in Akel [148] and Vercauteren [144] seems reasonable. Unfortunately, as mentioned earlier, the measure of the waist triangles described by Vercauteren [144] is designed, along with all of the other measures taken in that paper, to be independent of the location of the midline. Apart from the waist, all of the measures in the Vercauteren paper [144] give a difference in vertical height and do not deal with distance from the midline. Alternative measures of the shape and position of the waist within the torso are varied [154, 155] with the most attractive concept for this work described by Mason and Katzmarzyk [157].

2.6 Measures of kyphosis and lordosis.

2.6.1 Description.

As well as assessment of symmetry in the coronal plane, the sagittal plane can also be measured. Normal values for kyphosis and lordosis have been documented in the past using a variety of techniques including radiography and surface topography [27, 29, 145, 159, 160, 161, 162, 163, 164, 165, 166]. Fon et al in 1980 [159] measured the kyphosis in 316 lateral radiographs of subjects between the ages of 2 and 79 years of age. Kyphosis was measured between the upper and lower endplates of the vertebral bodies marking the points of inflection in the curve in the sagittal plane. Willner and Johnson [27] measured the sagittal profile of 1101 children aged 8 to 16 years (565 boys and 536 girls) using the spinal pantograph, which is a manual device with a wheel that was run down the spinous processes in the midline from the upper thoracic spine to the lower lumbar spine. The traced out shape was then transferred to paper where it could be measured using the Cobb technique [7] at the points of inflection. This was a cross sectional study that was then viewed longitudinally. More recent studies have either used radiation based methods (conventional radiographs or CT scanning) [29, 160, 161, 162, 163, 164, 165] or surface techniques [145, 166] to obtain normal values. Bernhardt and Bridwell [164] measured the kyphosis between T3 and T12 and the lumbar lordosis between L1 and L5 from radiographs in 102 subjects regardless of the location of the points of inflection. Similarly, Ghandhari et al [161], analysed radiographs of 98 subjects between the ages of 8 and 19 years. Mac-Thiong initially published on 180 subjects between 4 and 18 years of age [162] and this was subsequently expanded to 341 subjects with spinal radiographs using the arcs of a circle to mark the limits of kyphosis and lordosis [163]. Propst-Proctor and Bleck [165] analysed 104 lateral radiographs measuring kyphosis between the end plates of T5 and T12 and lordosis between L1 and L5. Schlösser et al [29] examined the vertebral body inclination angles for all vertebrae between C7 and L5 in 156 subjects between 7 and 18 years.

None of these studies is truly longitudinal in nature (and it would be unethical to perform repeated radiographic examinations on normal children). Those studies that report a mean value for kyphosis and lordosis for an entire age range are reported in Table 1. Whilst cross sectional studies are not longitudinal studies, it is possible to plot the results as longitudinal studies in the absence of true longitudinal data. Those papers that report a range of values dependent on age are plotted based on the data provided in the source paper. Results are presented as mean values for the entire group or subgroups by Carr et al [145], Ghandhari et al [161], Mac-Thiong et al [163], Propst-Proctor and Bleck [165], Schlösser et al [29] and Shefi et al [160]. Bernhardt and Bridwell [164], Giglio and Volpon [166] and Mac-Thiong et al [162], using the cross sectional cohort data that they present, analyse their data with linear regression techniques to attempt to predict the values versus age for the cohort.

Table 1: The results of papers quoting mean values of thoracic kyphosis (°) and lumbar lordosis (°) in adolescents and young adults.

Paper	Number of subjects	Age (years)	Method of measurement	Thoracic kyphosis (°). Mean (SD and range)	Lumbar Lordosis (°). Mean (SD and range)
Bernhardt [164]	102	4.6 to 29.8	Radiographic T3 to T12 and L1 to L5	36 (10, 9-53)	44 (12, 14-69)
Fon [159]	49	2 to 9	Radiographic between points of inflection	Males 21 (8, 5-40)	Not reported
				Females 24 (7, 8-36)	Not reported
	50	10 to 19	Radiographic between points of inflection	Males 25 (8, 8-39)	Not reported
				Females 26 (7, 11-41)	Not reported
Ghardhari [161]	98	8 to 19	Radiographic T1 to T12 and L1 to L5	47 (13, 6-73)	39 (12, 2-67)
Mac Thiong [162]	35	4 to $<$ 10	Radiographic using arcs to identify points of inflection	38 (10)	46 (12)
	145	10 to 18	Radiographic using arcs to identify points of inflection	44 (10)	49 (12)
Mac-Thiong [163]	341(including original 180)	4 to 18	Radiographic using arcs to identify points of inflection	All group 44 (12, 5-76)	48 (12, 15-101)
				Males 44 (12, 5-76)	47 (11, 15-74)
				Females 44 (10, 15-71)	49 (12, 21-101)
Propst-Proctor [165]	104	2 to $<$ 20	Radiographic T1 to T12 and L1 to L5	27 (IQR 21-33)	40 (IQR 31-50)
Schlösser [29]	156	7 to 18	Radiographic T1 to T12 and L1 to L5	Males 34 (9)	54 (10)

Willner and Johnson [27] (Figure 17), showed that in both males and females, the thoracic kyphosis decreases during early adolescence although in females it remains at a minimum value of 27° for several years, whereas in males it does not. The values then climb to a maximum value of 34° in females and 37° in males at the age of 18 years. The measurement of lumbar lordosis climbs in females from 33° to 38°. In males the lumbar lordosis alters with age but on average returns to the same value at the age of 16 years as seen at 8 years of age.

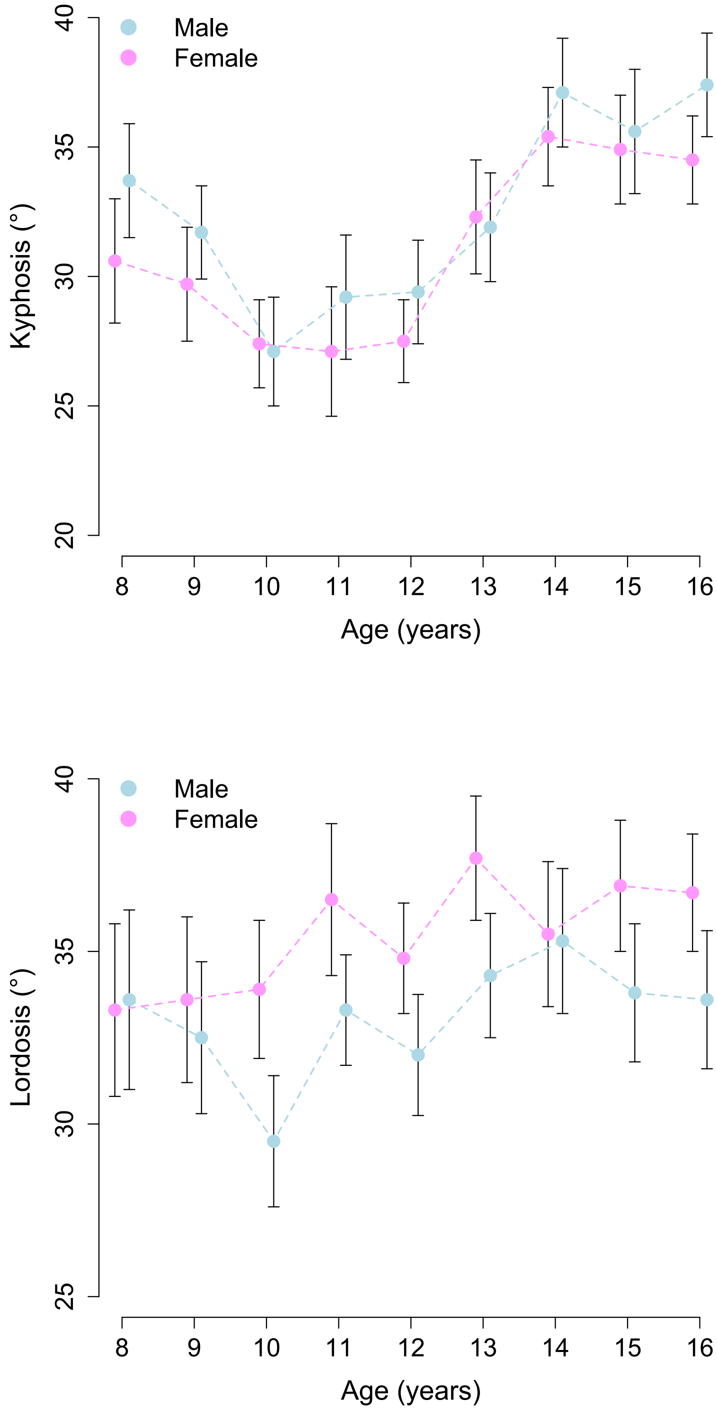
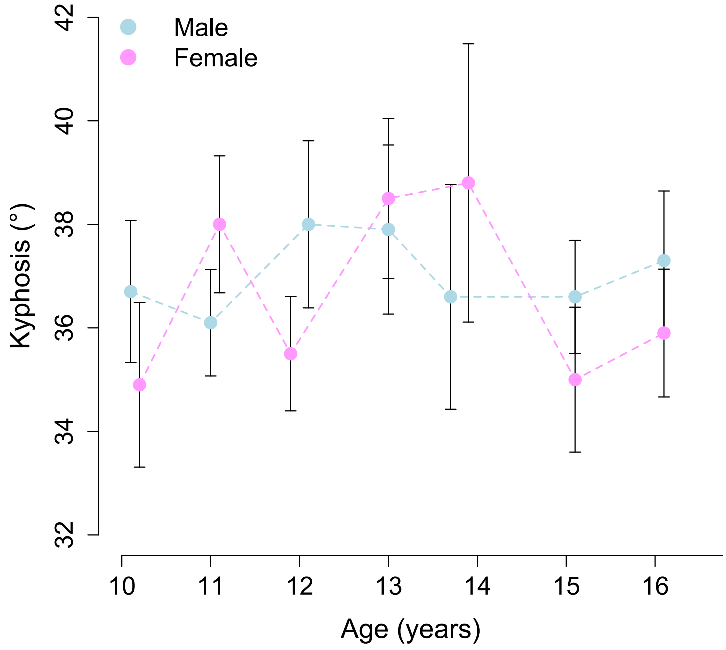


Figure 17: The mean kyphosis and lordosis angle (°) with 95% confidence intervals versus age (years) for both males and females (jitter applied for clarity). Redrawn from data in Willner and Johnson [27]. Jitter is a graphical technique that moves a pixel by an amount to make the graphic more understandable. In this case, jitter is applied as so not to obscure the 95% confidence intervals that would otherwise be on top of each other.

As noted previously, Carr et al [145] used ISIS1 to measure the kyphosis and lordosis angles from 271 children between the ages of 10 and 16 years. When the data is plotted longitudinally (Figure 18) there is no difference in the kyphosis angle with age or sex but in lordosis, females have a larger angle than males although this reduces with age.



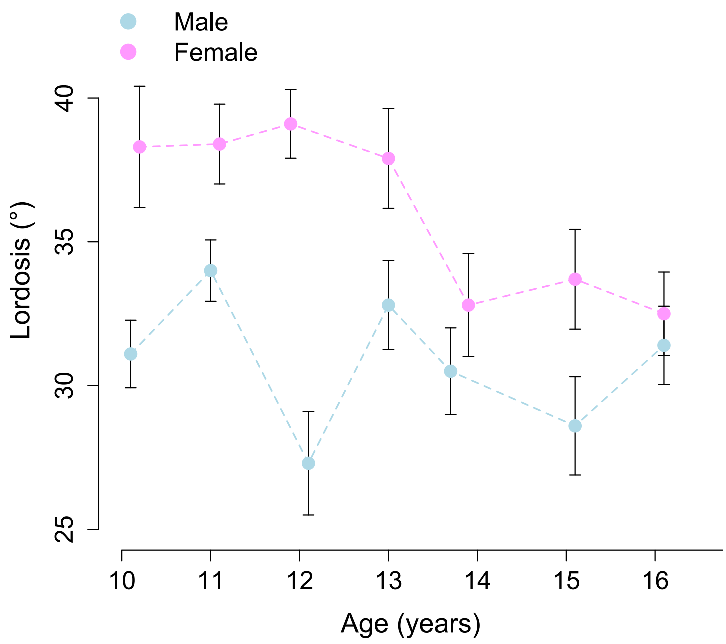


Figure 18: The mean kyphosis and lordosis angle (°) with 95% confidence intervals versus age (years) for both males and females. Redrawn from data in Carr et al [145].

In the paper of Giglio and Volpon [166], a spinal pantograph was used to measure kyphosis and lordosis in a method similar to Willner [27] in 718 subjects aged 5 to 20 years. As shown in Figure 19, the data is not smooth when viewed longitudinally with both kyphosis and lordosis climbing with age (although it does appear that kyphosis is greater in males and lordosis greater in females).

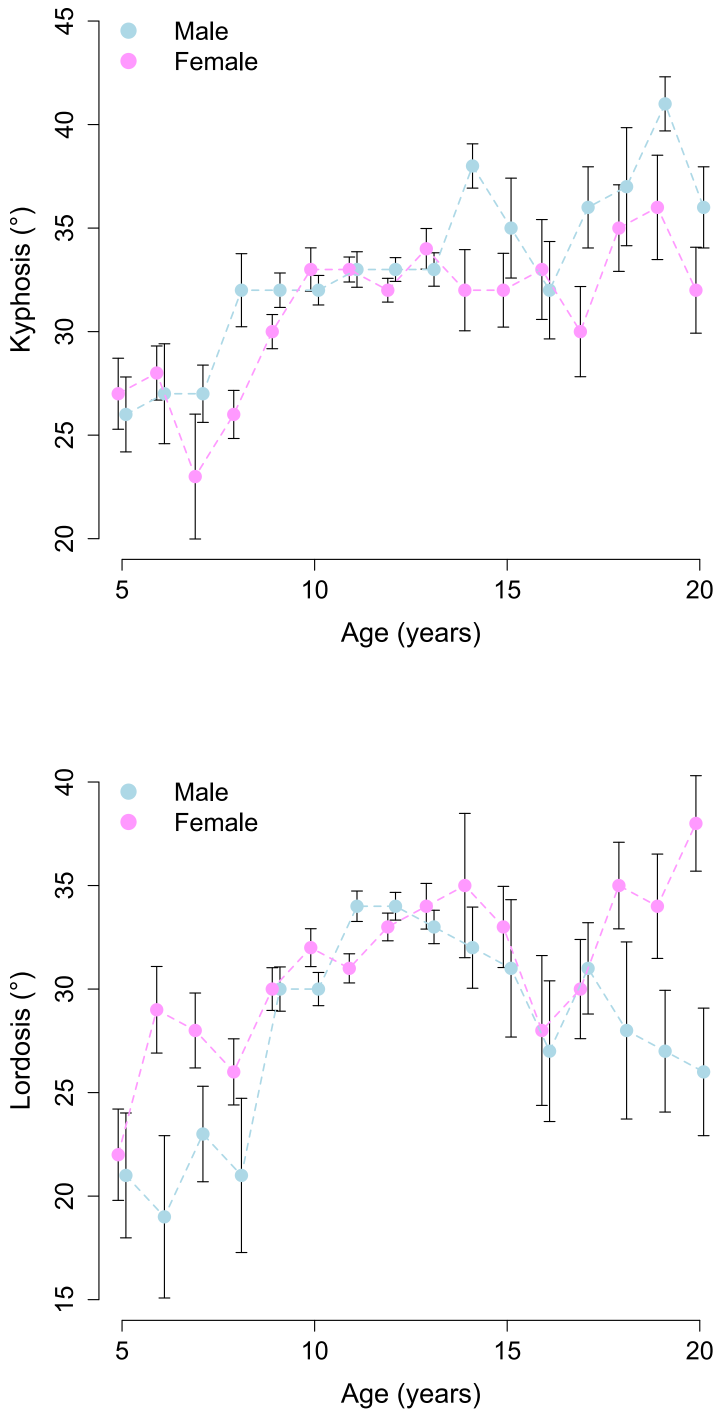


Figure 19: The mean kyphosis and lordosis angles (°) with 95% confidence intervals versus age (years) for both males and females (jitter applied for clarity). Redrawn from data in Giglio and Volpon [166].

Shefi et al [160] examined the lordosis from 210 patients (males and females combined) who had undergone CT scans in the supine position for abdominal pathology between the ages of 2 and 20 years by measuring between L1 and S1. Figure 20 shows this data plotted longitudinally. It shows lordosis increasing with age until the age of 15 years where there is a decrease.

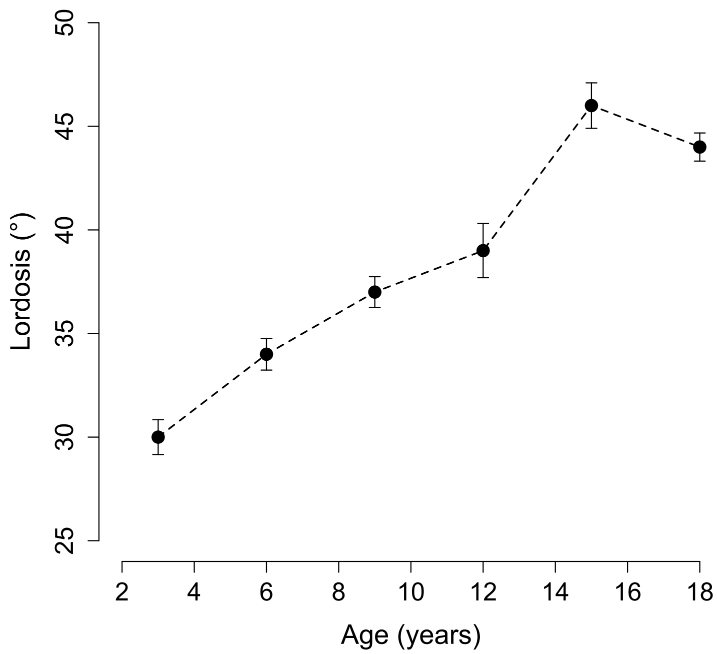


Figure 20: The mean lordosis angles (°) with 95% confidence intervals versus age (years) for all volunteers. Redrawn from data in Shefi et al [160].

2.6.2 Critique.

This review of the published literature on the measurement of thoracic kyphosis and lumbar lordosis in children and adolescents demonstrates the inherent difficulties in the making of these measurements. These studies are a mix of radiographic and skin surface measurement techniques. The vertebral levels from which the measurements are made vary. Within the radiographic studies, this variation in levels is to deal with the difficulties of visualisation of the upper thoracic spine through the shoulder girdle. The point where kyphosis becomes lordosis is pre-defined at the T12-L1 disc space in some papers regardless of where the actual point of inflection is, or is measured to that point of inflection regardless of the distance from T12-L1. In some papers there is a subdivision in the results for males and females and some present the results all together ignoring the possible effects of sex. The results are reported as overall mean values in some papers ignoring any effect of age. Other papers present results in age ranges or by year. What is common to all of the papers is that the cross sectional design used as the study method has then been viewed in a longitudinal manner, either through suggestion or through the use of linear regression techniques. This leads to the possibility of misinterpretation of the data and erroneous conclusions. This is particularly seen when comparing the results of the papers of Willner and Johnson [27] and Giglio and Volpon [166]. In both of these papers the methodology used was very similar. However the results are markedly different particularly in the development of thoracic kyphosis raising questions on how best to interpret these papers.

The use of a supine CT scan for the measurement of lordosis adds further to the confusion to the extent that the result should be discounted entirely [160]. The effect of gravity on the curve is removed by the process of lying down. Lying supine also has effects on the rotational position of the pelvis and this will secondarily affect the lordosis. There may also be a patient selection bias in selecting those who attended hospital for abdominal pathology. Intra-abdominal pathology can lead to guarding of the abdomen by tensing the abdominal wall to try and relieve pain [167]. This will have an effect on the shape of the spine and thus also the published results.

These papers highlight that there is a need for a new form of measurement and analysis for kyphosis and lordosis that measures all of the spine in upright stance and a neutral sagittal balance (not leaning forward or backward). This is of particular interest given the current thoughts around the pathogenesis of AIS being in the sagittal rather than coronal plane [28].

2.7 The use of surface topography in the management of scoliosis.

2.7.1 Description.

Surface topography has been used in the past to document the effects of scoliosis and how surgery affects the shape of the spine [91, 106, 107, 108, 109, 110, 111, 112, 114, 115, 116, 122, 125, 126, 130, 134, 168, 169, 170, 171, 172, 173, 174, 175]. A number of these publications are analyses showing that the surface topography measure that is being described correlates with the radiographic Cobb angle in the same population [107, 108, 110, 112, 114, 115, 122, 130, 170, 172, 173, 174, 175, 176]. Whilst there are a number of different surface topography techniques used in these papers and thus some slight differences between the surface topography parameters described, all of the papers describe results that allow the conclusions to suggest that there is a role for the use of surface topography methods to replace, or at least reduce the radiography used in the management of scoliosis. Surface topography is also described for monitoring of scoliosis [111, 119, 169, 171]. Sakka et al [119], using the Quantec® scanner, have shown that over repeated measurements there is an acceptable correlation between the surface topography measure (the Q angle) and radiographic Cobb angle. A similar conclusion was made by MacArdle et al [171] for thoracic sagittal curvature using the Quantec® scanner. Another similar study, using the BIOMOD- L® system was performed by De Korvin et al [111] who enrolled 100 children who had serial radiographs and surface topography images annually for three years. There was a mix of those who were observed and those who wore a brace during this time. The conclusion of this paper was that a 2° increase in surface topographical parameters would indicate a 5° increase in the radiographic Cobb angle. Komeili et al [108] used a whole torso scanning system that created colour maps indicating asymmetry in the surface shape of the torso about a vertical line of best symmetry between the two halves of the torso (which the paper notes may not be the midline). There were 46 subjects with AIS who were imaged twice, with a year between the images. There was good inter-observer reliability in analysis of the images and comparison with the radiographs showing that the deviations in the colour maps were in similar anatomical areas to the apices of the curves from the scoliosis. Hong et al [169] used the same surface topography technique as Komeili [107, 108] and used the results with previously published decision trees [107] to determine whether it was possible to reliably predict moderate or severe curves based on the surface topography images. The sensitivity of the analysis was reported as 95% with a negative predictive value of 90%. Specificity was 35% and the positive predictive value was 53%. Hong concludes that the use of decision trees would allow a reduction in the number of radiographs used in the clinical setting.

Surface topography has also been used to describe the effects of scoliosis and scoliosis surgery on the torso [91, 106, 109, 116, 125, 126, 134, 130, 168]. The ISIS1 system was used by Jefferson et al [125] both pre-operatively and 1 year post-operatively in 34 patients, who underwent scoliosis surgery for AIS. The surgery was performed with either Harrington instrumentation, Harrington Luque instrumentation (combining the Harrington rod with Luque sublaminar wires) or as a costoplasty. Those who had a costoplasty had previously undergone some form of scoliosis fusion surgery. Surgery with Harrington instrumentation led to statistically significant changes in the Cobb angle and the ISIS parameters of LA, maximum surface angle and hump severity. There was no significant difference in VA. The parameters of those with Harrington Luque implants and those who underwent costoplasty all decreased; however, the numbers in each group were too small for meaningful statistical analysis.

The POTSI index was used by Inami [134] describing the technique on 155 subjects with a scoliosis greater than 10° who were unoperated and 40 subjects who had undergone scoliosis surgery with Harrington instrumentation. There was a weak positive correlation between an increase in Cobb angle and an increase of the POTSI score. There was a decrease of 22.6 points in the POTSI score from the pre-operative score of 46.9 to the post-operative score of 24.3. Of interest, there was no change in the Suzuki hump sum between the pre-operative and post-operative group.

Theologis et al published two papers using ISIS1 [91, 126]. In the 1993 paper [91], 100 patients with AIS underwent ISIS scans, clinical photographs and radiographs. There were 10 non-medical judges who scored the clinical photographs for the severity of the deformity of both the spine and the torso creating the cosmetic spinal score (CSS that was scored 1-10 with 1 the least and 10 the most cosmetically acceptable back in the view of the judges). The Cobb angles and ISIS1 parameters were analysed against the average CSS. There was a negative correlation between the CSS and the Cobb angle (r=-0.49) and between the CSS and hump severity (r=-0.63). The correlation was stronger for CSS and surface topography rather than CSS and Cobb angle. Those who had undergone intervention through bracing or surgery showed improvement in the CSS, with surgery giving a greater change than bracing. In the 1997 paper [126], Theologis followed up 78 patients with a right thoracic AIS pattern who underwent ISIS1 scans and radiography. The group was then reviewed undergoing further ISIS1 scans every 3 to 6 months and radiographs every 6 months for a minimum of 18 months until either skeletal maturity or until a management decision had been made initiating either bracing or surgery (indicating curve progression). Progression of a Cobb angle of 5° was deemed a clinical change of note. The ISIS1 parameters of LA, VA and hump severity detected progression earlier than would have been done using radiographs for those whose Cobb angle ended at greater than 50° and were thus considered for surgery. In those who progressed 5° or more but were less than 50° and thus were braced, the ISIS parameters were not statistically significant. However, LA did predict progression in step with changes in the Cobb angle.

Griffiths et al [168] used the Quantec® system with a custom made grid over the back to allow comparison of the left and right sides following surgery in 9 patients. Whilst the authors all agreed that surgery had improved their appearance, a quantitative reduction in the asymmetry was only seen in 7 patients.

Mínguez et al [106] in their comparison of the POTSI and DAPI scores, demonstrated that both scores were increased by the presence of an underlying scoliosis with correlation r values of 0.668 for POTSI to Cobb angle and 0.706 for DAPI to Cobb angle.

Using ISIS2, Berryman et al [130] describe and establish the accuracy of repeat measurements of the size of the rib hump, measured in millimetres of height, in 60 subjects. It was found that the limits of agreement of a repeated measure was ± 10 mm, thought to reflect changes in the shape of the thorax caused through breathing and posture. In 2009 using a similar method, Berryman et al established that VA could also be monitored using ISIS2 [177].

McMaster and McMaster [116] reported on 37 patients with more than 2 year follow up with ISIS1, who were treated for AIS with staged anterior release of the spine and internal thoracoplasty followed by posterior spinal instrumentation and they showed that the rib deformity did not reoccur. This was attributed to the internal thoracoplasty.

2.7.2 Critique.

The literature around the use of surface topography techniques in the management of scoliosis reflects the observations already made in this literature review around normal torso asymmetry. There are a number of different systems reported that are measuring slightly different parameters in groups of patients who have undergone a number of different operative and non-operative treatments over several decades. In part this again is due to the changes in technology and computing power that has led to advances in what is possible. These papers do particularly comment on the difficulties of relating surface topography measures to Cobb angle, either as a Cobb proxy [108] or as a number denoting the amount of asymmetry such as POTSI or DAPI [106]. Also of note, is the view of Theologis [126], that a change in the Cobb angle of more than 5° is clinically significant and again, this provides a numerical value that could be used as an MCID for this thesis.

The literature does demonstrate that surface topography measures do change with a change in the size of the underlying curve [126] and with scoliosis surgery [91, 106, 109, 116, 125, 126, 130, 134, 168]. The work by Berryman et al [110, 130, 177] has also shown that there is repeatability of the ISIS2 system. Unfortunately, lesser papers such as that by Griffiths et al [168] do little to advance knowledge in this area, other than noting a interesting finding that would potnetially benefit from further investigation in the future.

A fair conclusion would be that surgery changes the shape of the back. However, extrapolating work that dates back many years performed with spinal instrumentation and techniques now obsolete to cases using modern corrective surgical techniques is not possible. Consequently, what happens to the torso shape in modern surgical practice is not clear and there is a role for surface measurement systems to investigate this further.

2.8 Analysis of differences in 3D shape.

The measurement and analysis of 3D shape is a widely required technique, across a number of different disciplines [178, 179, 180, 181]. The benefits of 3D shape analysis is in the ability to quantify the differences seen between shapes and assign that difference the appropriate significance relevant to that situation [182]. The assessment of 3D shape can be subdivided into historic methods and more latterly, methods that do or do not rely on landmarks [182].

Traditional methods are based on the use of linear distances to describe the shape [183]. These distances are then analysed using multivariate statistical techniques. Unfortunately, due to iatrogenic inaccuracies caused through the loss of the geometric structure in the use of these techniques, particularly in biological structures, the fitting of the model created was not accurate enough and newer methods have been developed [182].

The more recent methods can be deivided into those using landmarks to anchor the analysis to fixed points common to all shapes and landmark free methods. The use of landmarks can be labour intensive as they all need to be identified in each shape prior to the analysis. Landmark methods include those that are based on deformation such as Finite Element Scaling Analysis, those based on superimposition such as Procrustean Analysis and those based on linear distances including Euclidean Distance Matrix Analysis [182]. These techniques all act to compare one shape against a reference shape. A noted benefit of superimposition methods is the production of an interpretable visual image of the output of the analysis [182]. Procrustes techniques, so called because of the link to the name Procrustes from Greek mythology [184] (see Section 8.1 for more details), allows the assessment of the mean shape and variability of that mean shape [185]. Procrustes analysis has been used with spinal and scoliosis research in the past [186, 187, 188].

Landmark free methods are methods of analysing the global 3D shape without the use of landmarks [182], and these include shape statistics-based methods, function analysis, view-based methods, topology-based methods and hybrid methods [182, 185].

A recent literature review on the subject [182] concludes that for most applications, a method based on Procrustes techniques is widely accepted. This has been taken further with the development of techniques based on Procrustes methods by Dryden and Mardia [185], making the mathematics required more accessible to the non-specialist. Procrustes analysis uses landmarks common to all of the shapes to be analysed and removes the effects of location, rotation and scale to allow the mean shape, and the variability of that mean shape to be assessed independent of the effects of size.

2.9 Conclusion.

This review of the literature demonstrates the previous research in this area and crystallises how the research presented in this thesis will be original in the field. To the onlooker, scoliosis is a mix of a spine and a torso deformity, measured through cosmetic scores and radiographic angles with outcomes that differ depending on whether you are the patient, parent or surgeon. This perception is partly due to the difference in outcomes between these groups. A patient wants to look symmetrical and not deformed, whereas a surgeon wants the best possible, safe correction balancing long term function against ongoing deformation of the spine. This may or may not be the same. However, the patients' perceived end result of a back with no asymmetries and straight spine is through their understanding that this is normal across the population. This literature review has shown that a normal population is neither straight nor symmetrical and that these parameters change with age. It is unfortunate, although understandable, why many cross-sectional studies of growth have been analysed in a longitudinal fashion, ignoring the error that this creates. Any study that uses ionising radiation must have the safety of the subjects uppermost in the minds of the researchers. Thus the repeated radiographs necessary to conduct a truly longitudinal study are not appropriate and the radiographic studies that have been performed are often measures from radiographs taken for an unrelated reason.

There are significant methodological flaws in the literature, in particular that around the measures of kyphosis and lordosis. A cross-sectional study can be looked at as a longitudinal study but must be interpreted with caution as there will be loss of detail through this method. Studies that do not allow for the differences in age or sex in any parameter associated with growth are 'too broad a brush' to allow the mean result for the entire cohort to be applied with confidence (as reflected by the large confidence intervals that surround that mean value). It is also concerning that there are studies using imaging techniques taken for other reasons and making measurements that are then presented as the definitive result without identifying the errors inherent in this approach. The other difficulty with radiographic studies are the differences in measurement limits especially in the measurement of kyphosis. The upper limit of measurement is variable to cope with the difficulties of defining the upper thoracic vertebrae through the bulk of the shoulders overlying the spine. This is especially true in the older studies where the quality of radiographs was poorer. Consequently, measures are taken from an upper measurable limit that can be from the vertebral bodies of T1 to T5. It may be that the upper thoracic spine contributes little to the overall kyphosis of the thoracic spine but this has not been shown and so leaves an unanswered question about the methods used and the compatibility of the studies. All previous studies in this area must be treated with caution.

The studies that look at the overall torso shape are hampered by the techniques available to perform the study. The studies of Grivas et al [101, 102] have large numbers of children enrolled, but the scoliometer is a blunt tool measuring the angle of trunk rotation in three areas only. This does not represent a total description of the torso shape. The use of moiré contours or other forms of surface topography give a more complete picture. Unfortunately the results are difficult to compare due to the differences in the way parameters have been measured. The end conclusion is that there are differences in the shape of the torso that change with age but putting quantitative data to this is difficult. The use of scoring systems such as POTSI and DAPI are a way of regulating this problem and their use has been associated with some success.

The above issues are then repeated when reviewing the literature on the effects of surgery for scoliosis with regards to the surface topography of the torso. The papers quote different parameters in different ways to show the effects of surgery on the torso. Comparing the studies is difficult because of these issues. In the end, the conclusion is that surgery changes the shape of the torso towards a more symmetrical shape but further comment is not possible. There seems to have been no attempt made to assess the effects of scoliosis or surgery on the shape of the spine and torso compared to the shape of a non-scoliotic population. The work presented here will fill this gap in the literature, identifying normal posterior torso shape stratified for sex and age, how that shape is affected by scoliosis and how the surgery for scoliosis changes that shape in comparison to normal shape.

In regards to the best way of measuring the shape of the spine and torso, undoubtably it is using one of the surface topography methods. This is because surface topography offers the ability to measure the entire back in upright stance. Furthermore, multiple parameters can be measured from one image, and the data can be saved and reanalysed at a later date. There is no guidance in the literature as to which of the surface topography systems is recommended. It is with the user to appreciate the abilities and drawbacks of each system used when assessing their own data.

3 Methods.

3.1 Introduction.

This thesis analyses back shape asymmetry in two cohorts of scoliotic and non-scolioitc adolescents. This Methods chapter describes the design of the study and the two cohorts. Further, there is a description of the ISIS2 equipment used for the study and how it operated. Finally there is a description of how the ISIS2 images were processed to provide data on the 3D position of the key anatomical points of interest around the torso.

3.2 The design of the study.

The non-scoliotic cohort were children from a local school. This was a private, fee paying school. The reason that this school was selected was that the children of the author of this thesis go to this school and are in the age group of interest. It was felt that there would be better recruitment to the study as, with the author known to both the parents and children, there would be a greater interest in participation.

The non-scoliotic cohort of the study was designed to be a longitudinal analysis of growth through the adolescent growth spurt in both boys and girls. This was performed through repeat measures of the same children on a yearly basis. The first enrollment to the study was in 2011. The cohort of particular interest were the year groups which included children who would become 8 years of age (youngest year group) and 9 years of age (next youngest year group) during that school year. This was because these year groups would be in the study for the longest time and represented the greatest ability for longitudinal data. It was acknowledged within the study design that some children would exit the study at the end of secondary school, before moving on to further education. For the younger year groups, there would be 7 yearly measurement opportunities until the children reached the end of secondary education and so the study was designed to take place over 7 years. This time also covered the age of the adolescent growth spurt and it was also felt that measurement in the older child in further education would not add to the questions posed in this thesis.

Once ethical approval had been granted (NRES committee West Midlands - South Birmingham 11/H1207/10) (the protocol and participant information sheets are found in the appendices), the study was publicised by the author at the school. This was in the form of talking to parents and students at parent evenings, briefing the teaching body and presenting at whole school events in front of all students. Following this, all eligible students were approached using documents sent home with the child. These included age appropriate participant information sheets, parental information sheets along with consent and assent forms. If the child wished to participate, then the completed forms were collated and a timetable of appointments was created and administered by the school secretary. Inclusion criteria for the non-scoliotic cohort parts is seen in Table 2.

Participants were recruited every year between the first measurement in 2011 and the measurements taken in 2015 (the first five years of the study). This was because of the need to develop a cohort of younger children who could then be measured serially over their growth spurt. By 2015, further recruitment of younger children did not occur as these children would not have reached their growth spurt by the end of the study. Withdrawal was seen every year, due to children becoming too old for the study or leaving the study for a variety of other reasons. These reasons included leaving the school, an inability to attend the appointment to be measured for academic reasons and as desire to no longer be involved. In an attempt to maintain the cohort, measurement appointments were offered at the end of the school day. The study team also interacted with the school to maintain the idea of participation as a positive activity within the minds of the student body through presentation in school gatherings.

Table 2: Inclusion criteria for the non-scoliotic cohort

Aged between 8 and 16 years
Able to read English
Able to stand still for the time required to capture the ISIS2 image
No previous history of spinal or thoracic surgery
Parents willing to consent to child participation in the study
Child willing to assent to participation in the study

Table 3: Inclusion criteria for the scoliotic cohort

Aged between 10 and 18 years at time of first image
No thoracic surgery other than that undertaken for scoliosis
A diagnosis of AIS
Bracing not used as part of the management
Both pre-operative and post-operative ISIS2 scans available for review

The equipment for the study was transported to the school and assembled in advance of any measurement taking place. The height and weight of the participant was measured using a free standing stadiometer which was placed against the wall. For sitting height a stool of known height was used. The stadiometer was confirmed to be accurate though the measurement of a pole of known length and an object of known mass.

On arrival, each participant was asked to sign next to their name to confirm their willingness to continue in the study. They were prepared for the study (removal of upper body clothing, positioning of the collar and modesty sheet, positioning of the skirt around the waist and application of stickers (for further information see Section 3.2). Height was measured in the same way on every occasion, with the participant standing on the stadometer having removed their shoes first and their head in aneutral position. The participant back was to the wall and the child was confirmed to be still.

Sitting height was measured in the same fashion, but the participant sat on a stool with their back against the wall. Weight was measured with the child free standing (away from the wall) and stood still.

The ISIS2 image was then captured following the methods detailed elsewhere (for further information see Section 3.2). The ISIS2 image and analysed data is automatically saved. Measures of height, weight and sitting height were recorded on to a custom made database which ran alongside the ISIS2 database. All participants were measured by the same individual at all visits.

This proedure was repeated every year for each child. Pseudonymisation of the participants was performed with a unique code that allowed linkage of the same participants across the years of measurement.

The scoliotic cohort comprised of ISIS2 images taken as part of standard care at the regional scoliosis centre. These images and associated analysis results were all saved in the ISIS2 database at the hospital. Most, but not all, of the scoliotic cohort also had height, weight and sitting height recorded. This was performed using the same method as described for the non-scoliotic cohort. The ISIS2 images were recorded by a number of individuals as part of the scoliosis service. All individuals had been trained and had been assessed against the necessary competencies, in a similar way as a series of radiographers will take radiographs that form part of clinical practice and of research studies.

A further ethical application was made and granted (NRES Committee East Midlands - Northampton 15/EM/0283) to allow review of this scoliotic data,. The protocol is included in the appendices. As the data was collected as part of routine care and no further interaction with the patient was needed, consent for review of the data was not to be required. Both the date of birth and date of scan of the participant was recorded and so the database was subdivided in to those between the ages of 10 and 18 inclusive.

This became the scoliotic cohort. The inclusion criteria for the scoliotic cohort can be seen in Table 3.

Following a review of the literature of the features of concern for young people with scoliosis, the standard data output of ISIS2 for both the normal non-scoliotic cohort and the scoliotic cohort was augmented with information around the position of the shoulders, axillae and waist. Consequently, a customised ISIS2 application was created to allow these points to be identified and recorded for further analysis. The author then analysed every ISIS2 photograph within the custom application (as described below). Once this was done, the application produced the standard ISIS2 output, including information on spinal position and shape and the amount of asymmetry along with the x, y and z coordinates of the torso points of interest. These are the same points used for the POTSI score [134] and the DAPI score [106].

There was no missing surface topography data. For every ISIS2 image taken, the recorded data set was complete. For the normal non-scoliotic cohort, every participant had height, weight and sitting height recorded at every visit and again there was no missing data. Measures of height, weight and sitting height were not recorded for all of the scoliotic cohort, with these measures becoming part of standard care at a point after the ISIS2 became used within the scoliosis service.

The mix of ethnicity between the normal non-scoliotic and scoliotic cohorts were recorded through observation of the ISIS2 image in each of the cohorts.

3.3 A description of the ISIS2 system.

The tool used to make the measurements of the surface of the back and torso was the Integrated Surface Imaging System 2 (ISIS2) as described by Berryman et al [78]. The ISIS2 system is an evolution of the ISIS1 device [109]. Whilst there are some similarities between the systems, ISIS2 uses different techniques of data acquisition and analysis compared to ISIS1. The ISIS2 system was used as it is a radiation free, surface topography system, that allows risk free repeated measures in the non-scoliotic cohort. It is already in use as standard care for all scoliosis within the hospital, so all of the scolioticcohrt have these images and it is important to use the same system for both cohorts to allow comaprisons to be made.

The ISIS2 system consists of a patient stand and a separate digital camera and projector, mounted on a telescopic actuator [78]. The patient stand is a frame with a black background (Figure 21). There is a horizontal bar against which the abdomen is rested. There are also arm support bars that hold the arms away from the sides of the torso without the need for the use of the trunk muscles of the subject. The abdominal bar and arm supports also help to prevent sway of the subject. Prior to the start of measurement, a retractable vertical screen that is part of the patient stand is deployed. Horizontal lines are projected on to this from the slide projector (Figure 22) and measurement of these lines allows calibration of the system.

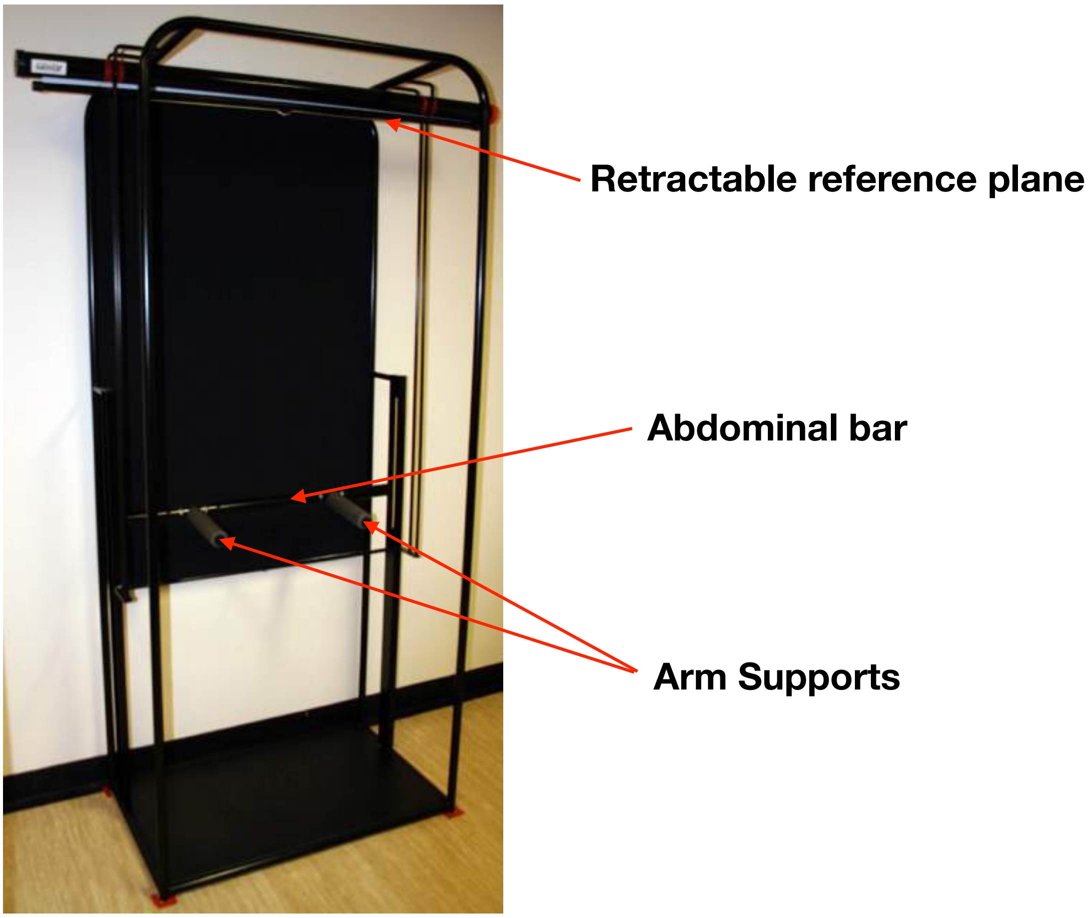


Figure 21: The patient frame showing the retractable reference plane, the abdominal bar and the arm supports.

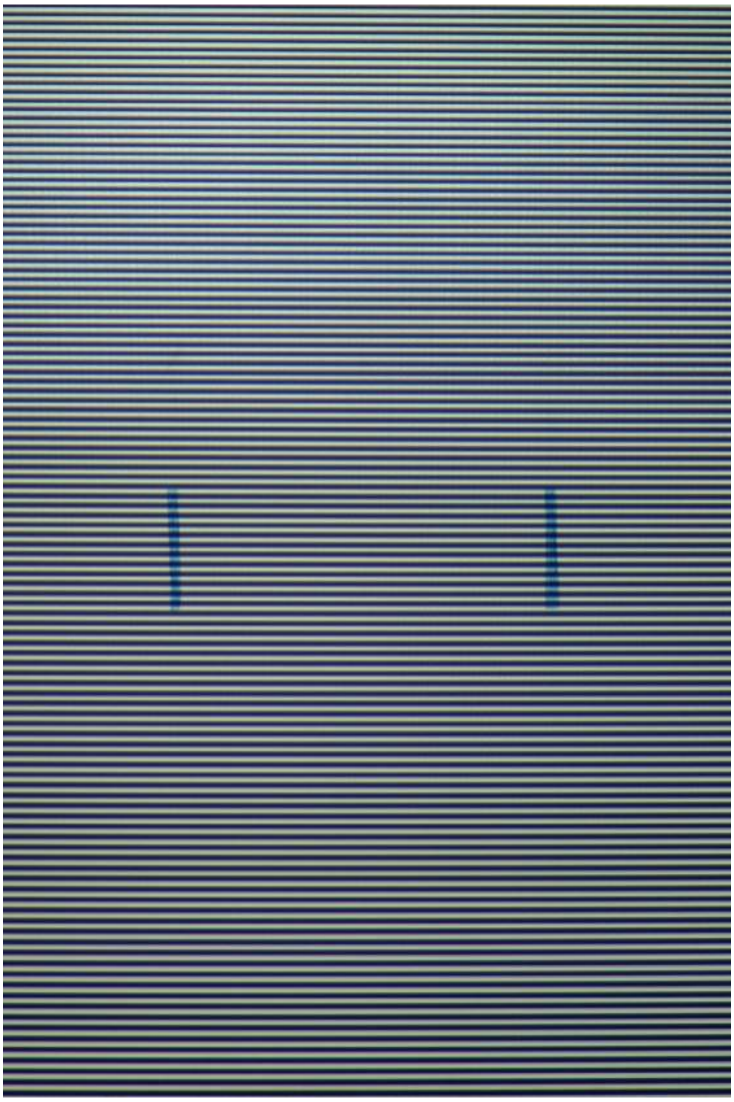


Figure 22: The reference plane with projected horizontal lines, used in the calibration of the ISIS2 system.

When using ISIS2, the subject undresses from the waist upwards so that the full back can be seen. There may be a need to loosen the waist of the trousers or skirt to expose the dimples of Venus (also known as the lumbar dimples) on either side of the sacrum. If necessart, the hair is tied up and held off the back to prevent interference with the image. A black collar is placed around the neck of the subject at the level of the mid cervical spine. A black apron is placed around the trunk at the hips, below the dimples of Venus, at the level of the top of the trousers or skirt. These are required to define the superior and inferior limits of the image for the analysis. To preserve the modesty of female subjects (and males should they wish), there are collars with an attached piece of material that hangs down from the collar to cover the anterior chest, and the apron covers from the waist down. Using palpation, landmarks are identified and turquoise coloured stickers are placed over these landmarks (Figure 23). The most superior landmark is the vertebra prominens (VP), marking the junction of the cervical and the thoracic spine. The VP is the spinous process of the C7 vertebral body and is shaped anatomically like a thoracic spinous process rather than the bifid spinous processes of the rest of the subaxial cervical spine. Inferiorly, stickers are placed on the dimples of Venus on either side of the midline. The dimples of Venus are a result of subcutaneous ligaments between the skin and the posterior superior iliac spine (PSIS). By design, ISIS2 defines the position of the sacrum as the midpoint of a line connecting the two PSIS locations. Stickers are then placed over some spinous processes between the VP and the sacrum. Whilst there is not a requirement to mark all of the spinous processes, it is recommended to mark a minimum of four taking care to include a sticker at the apex of all curves. In the post-operative subject where the spinous processes have been removed as part of the operative procedure and there is a linear scar on the back, the VP and sacrum are identified as for a non-operated subject. The scar is marked in several places between the VP and sacrum acting as a proxy for the position of the spine.

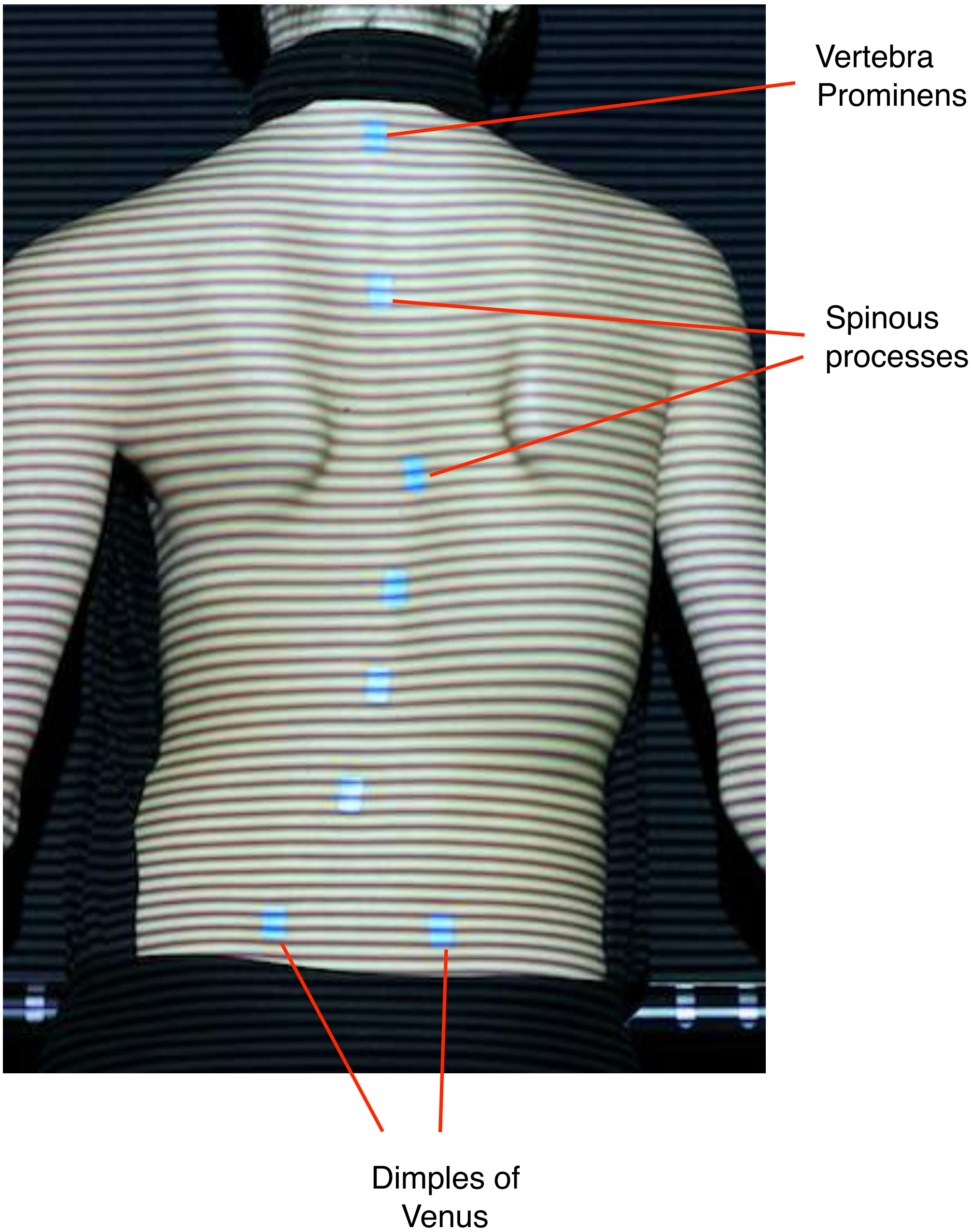


Figure 23: Example photograph of the back showing the turquoise stickers and the projected pattern of lines needed for ISIS2 calculation of the 3D back surface.

With the reference plane removed the subject then stands in the frame and the image of the back is captured using the ISIS2 system. The captured image is from a digital camera connected to an Apple® computer. The image is then processed using the ISIS2 software to create both graphical and numerical information that describe the shape of the spine and torso in 3D. Horizontal lines are projected onto the back of the subject from a slight downwards angle to create the image that the system can analyse. Whilst if projected onto a flat vertical surface (such as the reference plane) these lines would be horizontal, with a body present, the pattern of the projected lines is altered by the shape of the body. The 3D information is created using a Fourier Transform Prolifometry technique, through the phase shift in the lines over the back compared to that of the lines on the reference plane, calibrated by the distance between the centre of the imaging plane to the centre of the reference plane, and the centre of the projection plane combined with the separation of the lines over the back [78]. The 3D surface is tilted and rotated relative to the reference plane through the VP and PSIS points to eliminate any effects of stance prior to analysis.

Typical examples of graphical output of ISIS2 are shown in Figures 24 (mild deformity) and 25 (severe deformity) with both pictorial and numerical information. The information is arranged in three horizontal rows. The top row shows a wire plot of the back similar to the Adams forward bend position (the Height map) and shows the contours and asymmetries that would be seen clinically. The Contour plot is to the right of the Height map and is a 3D representation of the back of the subject. In some ways this plot can be viewed in a similar way to an Ordnance Survey® geographical map with contour lines representing points of equal height on the back. Using a heat colour map, the height of the back away from the reference plane is shown with red being the greatest distance and blue the least distance. Small solid blue circles represent the stickers that were placed on to the back. The solid blue triangles are the most prominent points on the back. In the middle panel are three plots that represent the three planes of deformity; axial (called transverse in the plot), coronal and sagittal. All of the plots have red, blue and green markers or lines on them. The blue markers represent the spine; the green and red markers represent the paramedian region seen at 10% of the back length away from the spine. In the transverse plot, there are 19 horizontal lines (including through the VP and sacrum) that are associated with a value of skin angle in degrees. This is the ISIS2 equivalent of a scoliometer measurement at those horizontal levels and this demonstrates the changes in shape over any rib or lumbar hump. Printed under this plot are the parameters measuring rotation (which is the amount the image was rotated by the system prior to processing to make the PSIS stickers equidistant to the reference plane) and the back length (a measure of the vertical height of the back between the VP and sacrum ignoring the effect of any coronal offset). The coronal plot is most similar to the radiograph used for the measurement of the Cobb angle. The blue line is the line through the stickers. The dashed black line shows the projected position of the centre of the vertebral bodies and the lengths of the horizontal black lines represent the amount of volumetric asymmetry (VA. See later in this paragraph for a description of this parameter). The points of inflection in the spine line are indicated as dotted lines and the angle between the dotted line and the horizontal is measured using the same method as the Cobb angle [7]. Addition of the angles seen between two lines of inflection will give the parameter lateral asymmetry (LA), which is the Cobb angle proxy. In the work presented here the LA has been further defined as the main or compensatory curve, dependent on the size of the curve with the main curve defined as the larger curve. The direction of the curve is included. The curve has also been given an anatomical location based on the location of the apex following the rules of the Lenke classification [55]. Associated with this plot are the numerical values of coronal imbalance, LA for both the main and compensatory curves and volumetric asymmetry

(VA). Coronal imbalance is the horizontal distance between the VP and the sacrum. If the VP was directly above the sacrum then coronal imbalance would be zero. VA is a measure of the rib hump (see later description of this parameters in Figure 26). The sagittal plot is very similar to the coronal plot and is equivalent to a sagittal radiograph of the spine. Again the blue line is the line through the stickers with the point of inflection between kyphosis and lordosis marked with the middle of the three solid black lines. The solid black lines at the top and bottom indicate a location 0.05 multiplied by the back length, proximal of the sacrum and distal of the VP. At the location of the apex of the curve, the line and figure represents a linear measure in millimetres of the kyphosis or lordosis taken from a straight line (also seen) between the VP and the sacrum. The paraspinal green and red lines are of different shapes to the blue line in this plot. This is because these lines are to the right and left of the spine and show the sagittal shape of the paraspinal region (that will include the rib hump which will affect the shape of the sagittal profile at that level). The numerical values with this plot are flexion (or extension if a negative value), kyphosis and lordosis and sagittal imbalance. The parameter of flexion is an angular measure of the position of the VP relative to the sacrum as the subject stands for the image. If the VP was directly above the sacrum, then the flexion angle would be zero degrees. Kyphosis is the angle between the line just inferior to the VP and the point of inflection and lordosis is the angle between the point of inflection and the the line just superior to the sacrum in a similar way to the Cobb angle on a sagittal radiograph. The point of inflection is variable and not defined as the anatomical T12/ L1 disc space. Sagittal imbalance is the sagittal equivalent of coronal imbalance.

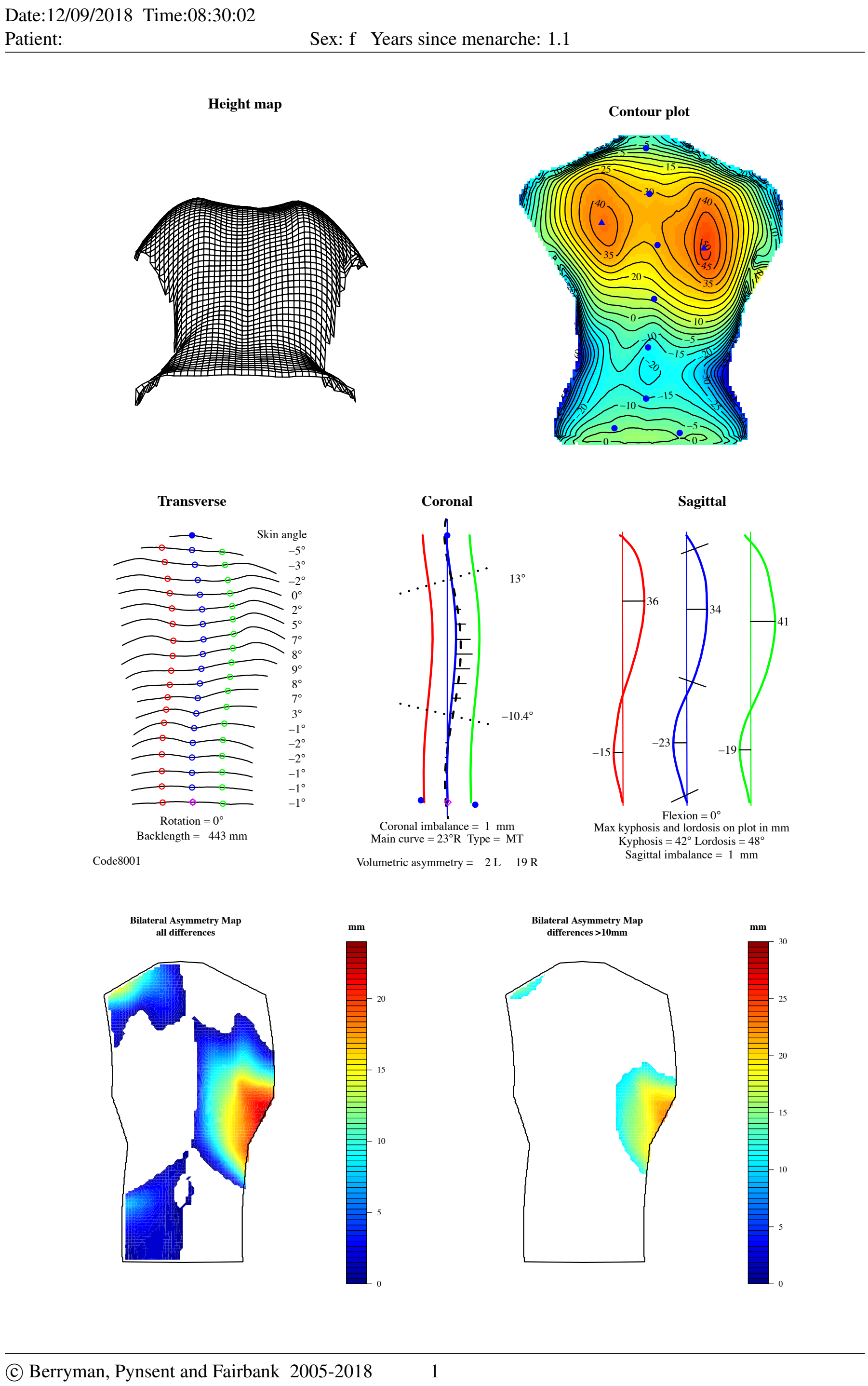


Figure 24: The graphical output of ISIS2 for a moderate curve.

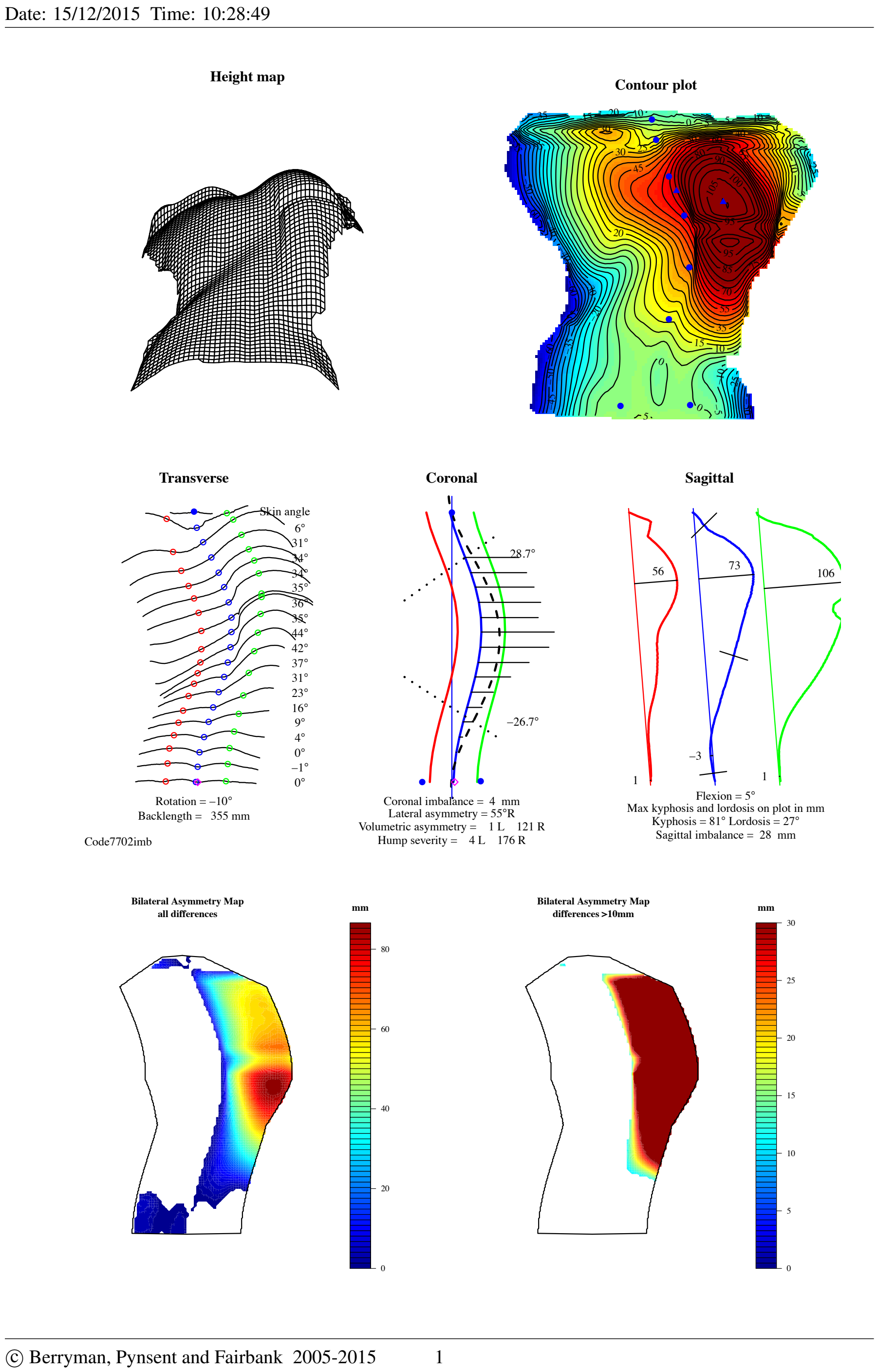


Figure 25: The graphical output of ISIS2 for a severe curve.

The bottom panel shows the asymmetry of the torso around the spine line. On the left is an image where any asymmetry is shown, and on the right, an image showing only asymmetry of a height of 10 mm or more. The size of the asymmetry is shown in a heat map format as previously described with blue the least asymmetry and dark red the most asymmetry as seen on the scales next to the plots. The numerical parameter associated with this plot is the VA. VA is a unit-less parameter that is calculated for each rib or lumbar hump and comes with an L or R to denote the left or right side. It is calculated as the sum of the difference in area in 19 transverse sections through the back normalised and scaled for the length of the back (Figure 26). Volumetric asymmetry does not make an assessment of the shape of the rib hump. It simply gives an overall value, a higher values meaning a larger volume of the hump.

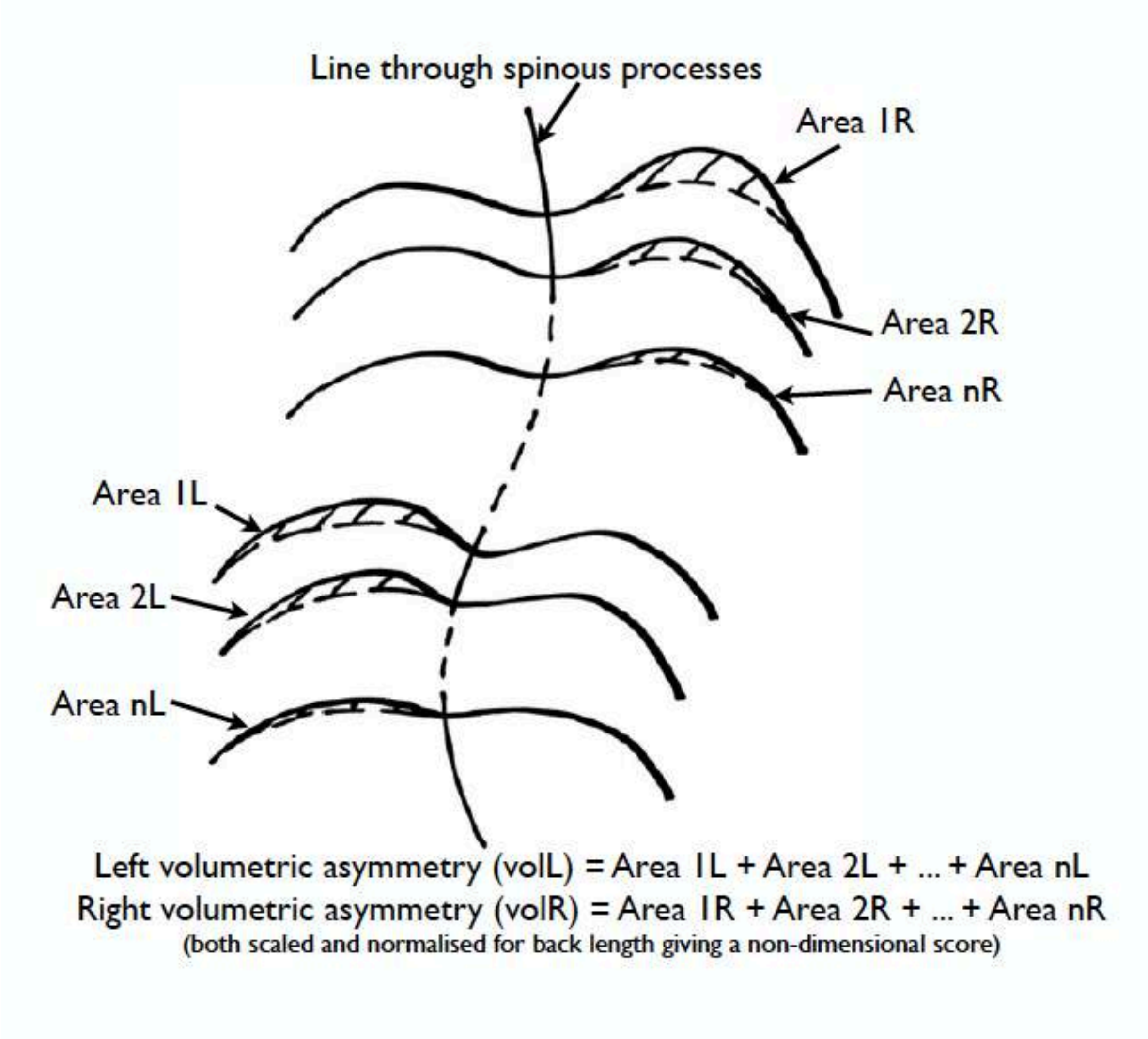
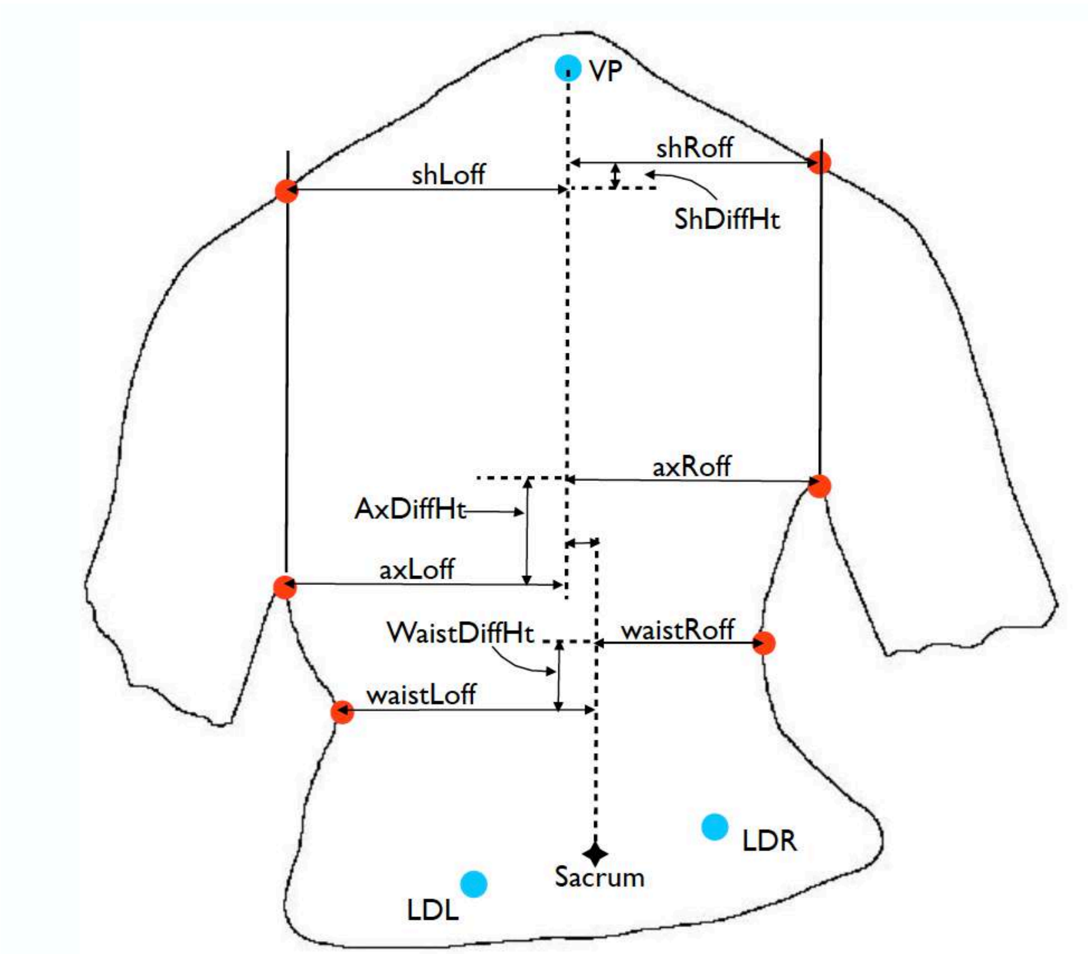


Figure 26: The calculation of volumetric asymmetry (VA) (as seen in [129]).

The method for taking the images was the same in both the non-scoliotic and scoliotic cohorts. The non-scoliotic cohort was a prospective serial measurement of the children on an annual basis between 2011 and 2017. The scoliotic cohort was an analysis of images taken as part of usual care in the scoliosis clinic.

For the work presented here, all of the images were reanalysed by the author using a custom interface allowing the identification of the extra points on the torso, which are demonstrated as solid red circles on Figure 27. This was performed by clicking with the mouse on the relevant points using the ISIS2 photograph as shown in Figure 28 (where the red circles are shown in the same anatomical points as in Figure 27). These points were the axillae (defined as the most superior part of the posterior axillary fold on either side of the body), the shoulders (defined as the point where a vertical line from the axillary point crosses the edge of the body on the shoulder girdle) and the waist points (defined as the most indrawn part of the waist on either side, also known as the minimal waist [157]). The 2D parameters obtained from the locations of these torso points are defined in Figure 27 and Table 4 and are differences in vertical height (DiffHt) or distance from the midline (DiffOff). For the shoulders and axillae, the midline was defined as a vertical line inferiorly from the VP and for the waist a vertical line superiorly from the sacrum. In the absence of any deformity, these lines would be the same. If the right point was further from the midline or vertically higher than the left, this point was defined as a positive value. If the left was higher or further from the midline than the right point then this was given a negative value. The locations of the torso points and the associated 2D parameters were stored in the ISIS2 database automatically and there was no transcription of data at any point. The data was later exported as a .csv file for processing and statistical analysis in R [189].



VP = vertebra prominens

● **LDL, LDR = lumbar dimples (dimples of Venus) left and right**

✦ **Sacrum location as calculated as mid point between lumbar dimples**

For WaistDiffHt, AxDiffHt, ShDiffHt +ve means right side higher
Offset parameters are all +ve, whether to left or right

AxDiffOff = axRoff - axLoff
WaistDiffOff = waistRoff - waistLoff

Figure 27: The torso points of interest as discussed in Table 4[129].

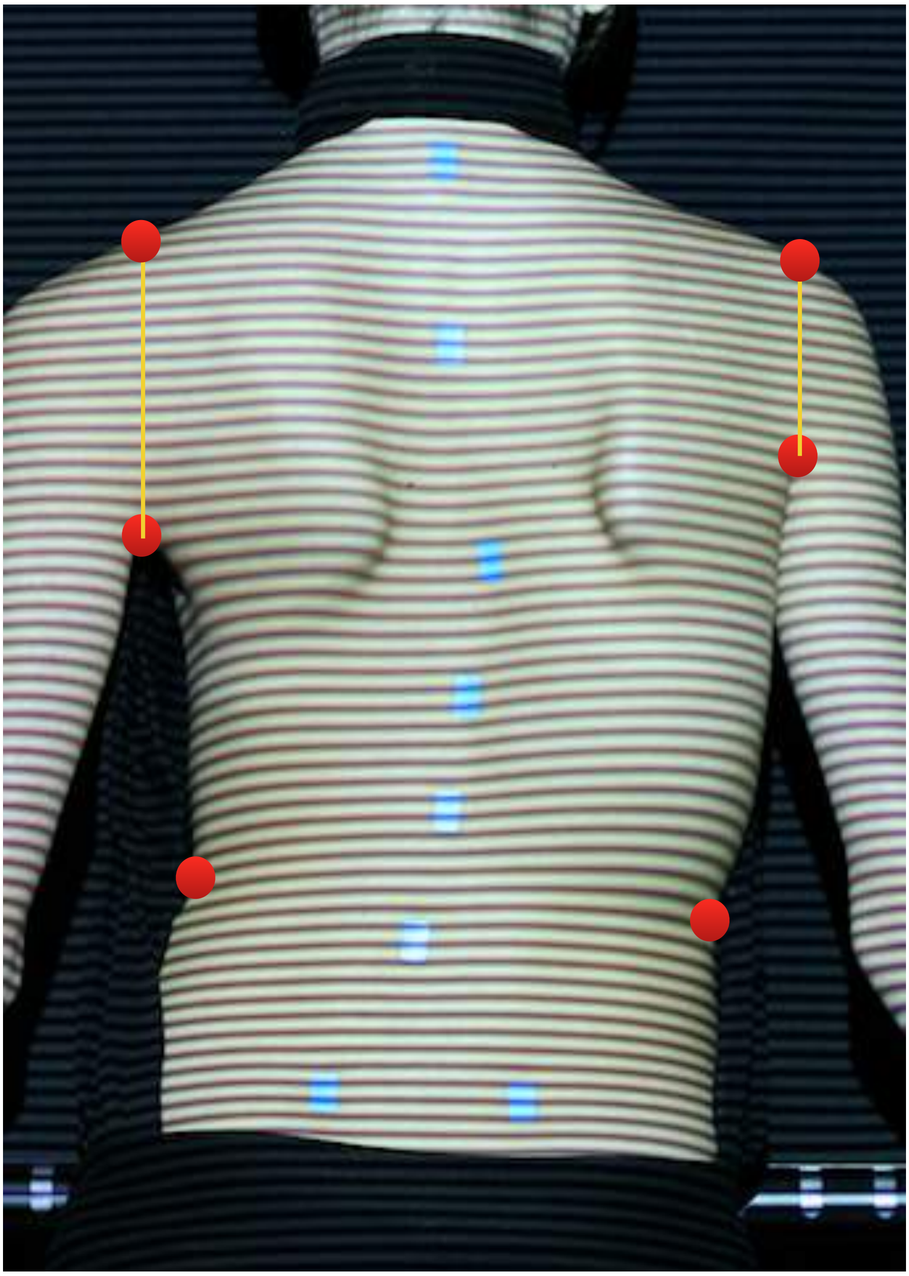


Figure 28: The torso points of interest shown on the ISIS2 photograph.

Table 4: The definitions of the parameters in Figure 27[129].

Orientation	Torso parameter (mm)	Definition
Vertical measurement	ShDiffHt	The difference in vertical height between the shoulder points
	AxDiffHt	The difference in vertical height between the axillary points
	WaistDiffHt	The difference in vertical height between the waist points
Horizontal measurement	axRoff	The horizontal distance from the midline to the right axillary point
	axLoff	The horizontal distance from the midline to the left axillary point
	waistRoff	The horizontal distance from the midline to the right waist point
	waistLoff	The horizontal distance from the midline to the left waist point
	AxDiffOff	The difference between axRoff and axLoff
	WaistDiffOff	The difference between waistRoff and waistLoff
	Total Axillary Width	The sum of axRoff and axLoff
	Total Waist Width	The sum of waistRoff and waistLoff

The most prominent points over the back (Figure 29) are points with 3D locations represented by the blue triangles seen on the ISIS2 contour plot. The parameters generated from these points are defined in Table 5. Again these parameters represent the difference between the right and left sides and are given positive and negative values as previously described.

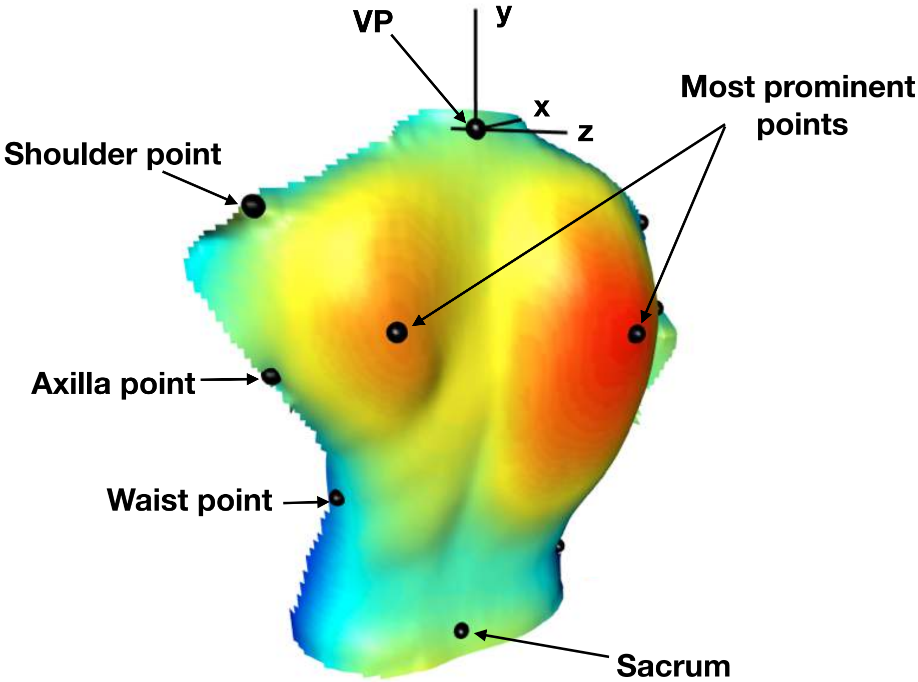


Figure 29: The location of the most prominent points as defined in Table 5.

Table 5: The definition of the most prominent point parameters shown in Figure 29.

Orientation	Torso parameter (mm)	Definition
Horizontal (the x parameter seen in the coronal plane)	MaxDiffOff	The difference in horizontal distance between the right most prominent point and the midline and the left most prominent point and the midline
Vertical (the y parameter seen in the coronal plane)	ScapDiffHt	The difference in vertical height between the right and left most prominent points
Depth (the z parameter seen in the sagittal plane)	ScapDiffDepth	The difference in 3D height between the right and left most prominent points

In the following chapters, box and whisker plot are used in conjunction with a superimposed mean with its 95% confidence interval. For clarity, Figure 30 shows the construction of this type of plot and defines the different parts of it. Of note, some of the box plots have a 'devils ear' appearance where the box and whisker plot has angulated corners rather than a box shape. This occurs when the number of participants is small such that the calculated error for the 95% confidence intervals for the median are greater than the value of the 25th and 75th quartile.

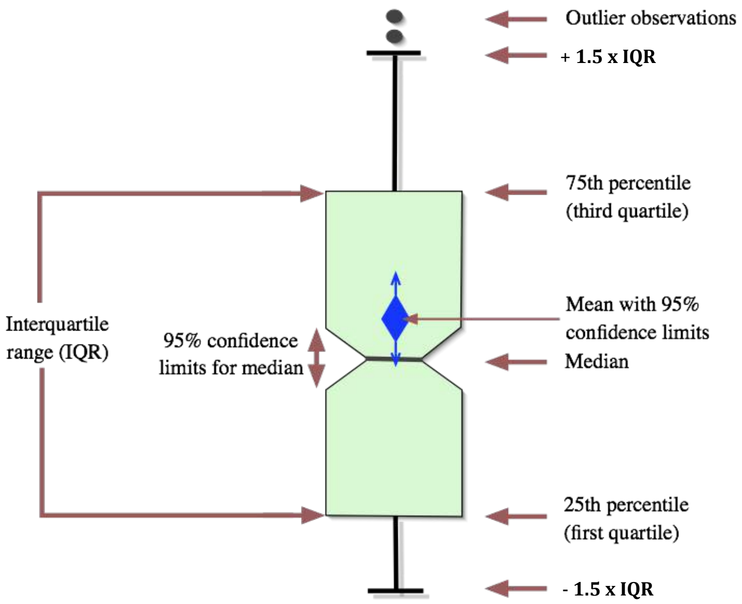


Figure 30: The definition of the format of a box and whisker plot.

In conclusion, this chapter describes the design of the two arms of the study, the non-scoliotic and scoliotic cohorts. It then describes the techniques of measurement using the ISIS2 system, and the processing that was performed after the images were obtained to allow identification of the torso points for analysis. Finally, an explanation of the measured parameters that will be discussed within the results chapters are defined, along with the components of a box and whisker plot.

Using this information:

- A description of growth and the proof of normality using growth charts is found in Chapter 5 - Proof of normality of the non-scoliotic cohort using published growth standards.
- A description of the growth of non-scoliotic children is found in Chapter 6 - The normal child.
- The effects of scoliosis and subsequent surgery for scoliosis on the shape of the torso compared to the shape of the non-scoliotic child is found in Chapter 7 - Pre-operative to post-operative change.
- The use of Procrustes analysis to examine the 2D and 3D shape of the torso in non-scoliotic, pre-operative and post-operative subjects is found in Chapter 8 - Procrustes analysis of torso shape.

4 Recruitment demographics and measurement reliability.

4.1 Introduction.

This chapter describes the demographic details of the non-scoliotic and scoliotic cohorts, including consort diagrams to describe the enrollment of the study as it progressed over time. There follows an analysis of the potential sources and sizes of measurement error throughout the stages of data collection prior to analysis.

4.2 The demographic details of the non-scoliotic and scoliotic cohorts.

In the non-scoliotic cohort there were 831 individual measures made in 196 subjects measured between 2011 and 2017. In the scoliotic cohort there were 3446 measures made in 1369 subjects measured between 2008 and 2017. Of that cohort there were 289 scoliotic subjects who had undergone both pre-operative and post-operative ISIS2 scans. These 289 paired images formed the pre-operative and post-operative scoliotic cohort that were subsequently used in the analyses. Table 6 summarises the demographic details of the non-scoliotic and scoliotic cohorts. The range of age for the post-operative cohort is greater than 18 years of age; however the diagnosis is AIS as it was made prior to the 18th birthday and it is thus included. Both of the groups were of predominantly Caucasian descent. The time from the pre-operative scan to surgery was a mean of 413 days (SD 304 days, range of 1 to 2057 days) and from surgery to the post-operative scan was a mean of 263 days (SD 270 days, range of 34 to 1984 days).

The scoliotic data set has two separate components as not all of the scoliotic cohort have recorded heights and weights. Thus for the analysis of standing height, weight, BMI, sitting height and sitting height to height ratio, a smaller subset of the scoliotic cohort was used. This is different to the data set used for the rest of Chapter 7. Statistical analysis comparing the distribution of the torso point data between the complete scoliotic data set and the subset of the scoliotic data set with height and weight data demonstrated no significant difference. This comprised 155 subjects who had a main thoracic curve pattern (135 females and 20 males) and 18 females with a main thoracolumbar curve pattern. There were no males with a main thoracolumbar curve pattern.

Table 6: The demographic details of the non-scoliotic and scoliotic cohorts.

Cohort	Mean age (years)	SD (years)	Range (years)	Males	Females	Number of individual images
Non-scoliotic	12.9	1.9	9.2 to 17.9	117	79	831
Scoliotic	15.3	1.9	9.9 to 19.0	293	1076	3446
Pre and post-operative cohort	13.9 (at pre-operative image)	1.5	9.9 to 17.9	39	250	289 (pairs of images)
Pre and post-operative cohort	15.7 (at post-operative image)	1.6	12.0 to 19.0	39	250	289 (pairs of images)

All of the subjects in both the non-scoliotic and scoliotic cohorts were measured at least once and in some cases many times. Table 7 and 8 show the number of measures per participant for both the non-scoliotic and scoliotic cohorts.

Table 7: The number of measures per participant for the non-scoliotic cohort.

Number of times measured	1	2	3	4	5	6	7
Number of subjects	16	14	21	57	45	30	13

Table 8: The number of measures per participant for the scoliotic cohort.

Number of times measured	1	2	3	4	5	6	7	8	9	10	11	12
Number of subjects	482	356	204	162	76	44	25	12	6	0	1	1

4.3 Consort diagrams.

A consort diagram of the recruitment and withdrawal of participants in the non-scoliotic cohort is shown in Figure 31 and for the scoliotic cohort in Figure 32.

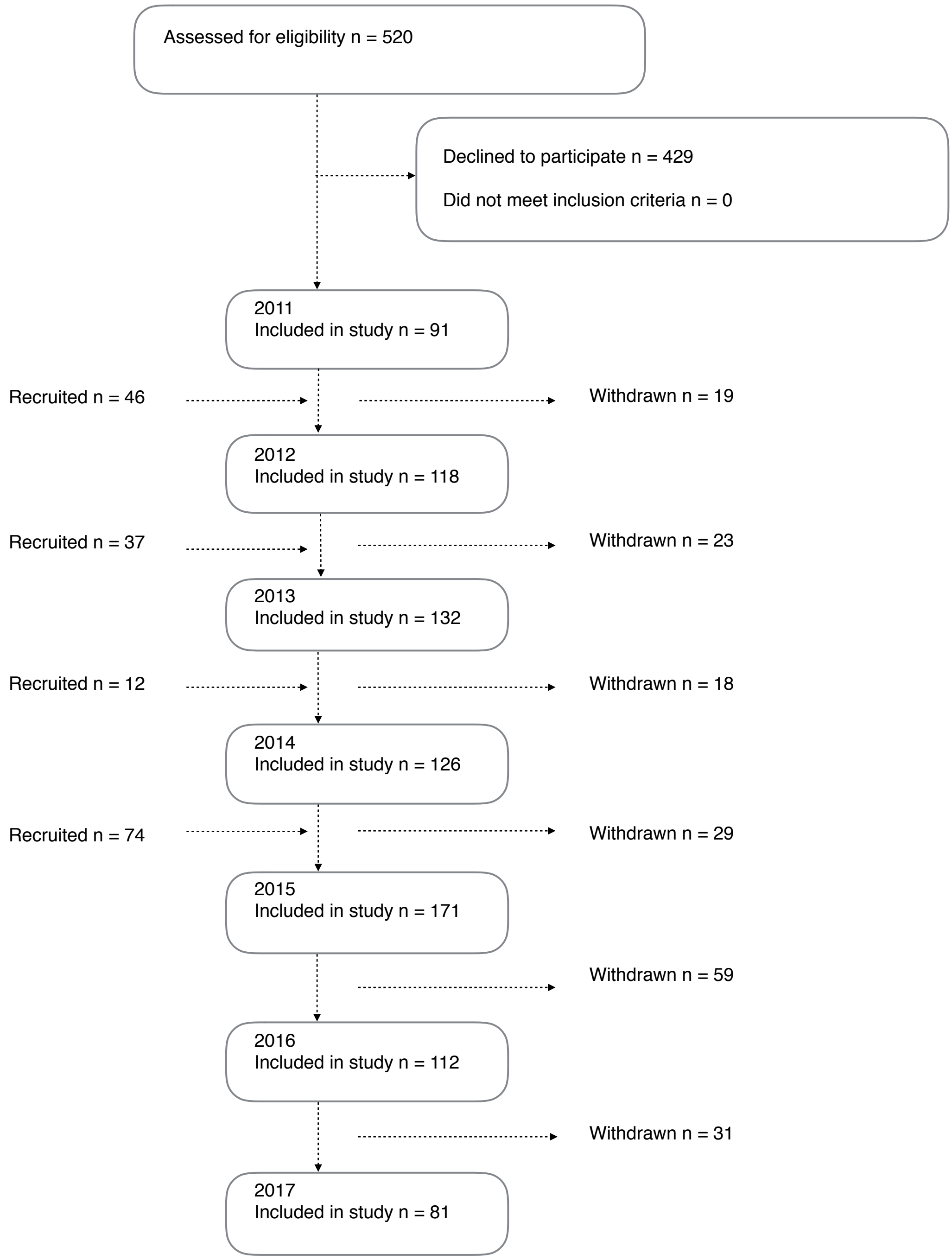


Figure 31: A consort diagram of the numbers recruited and withdrawn from the non-scoliotic cohort between 2011 and 2017.

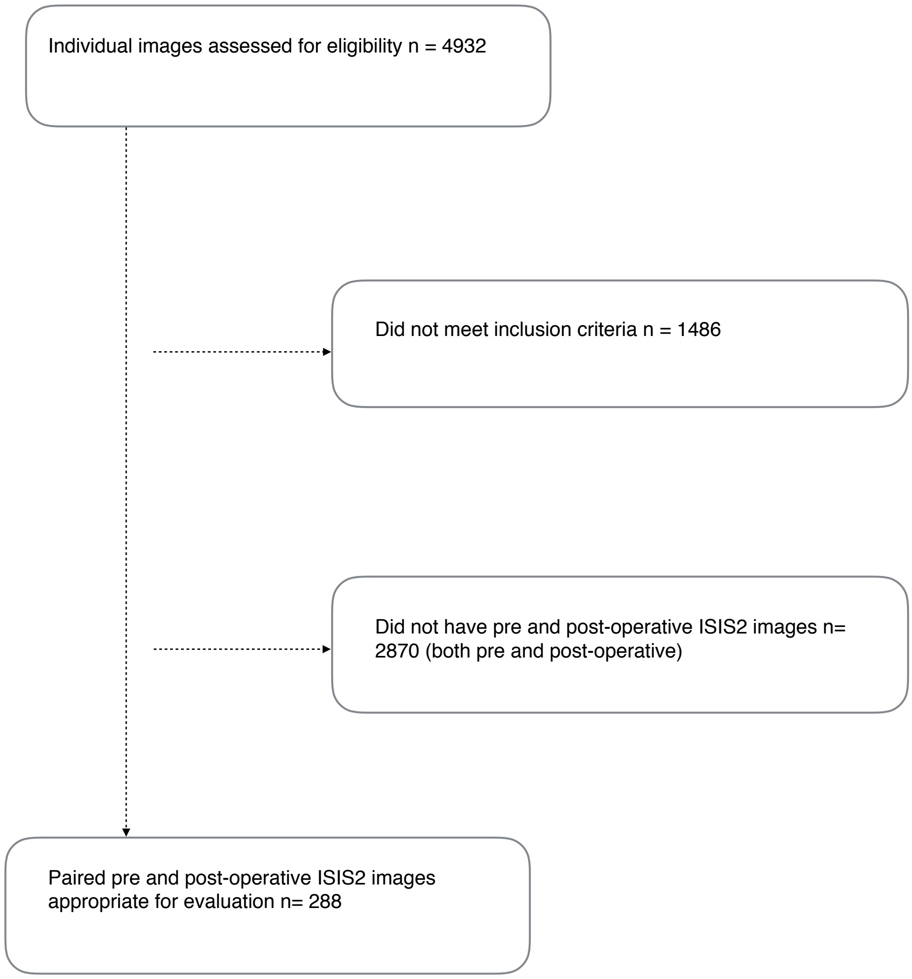


Figure 32: A consort diagram of the numbers recruited and withdrawn from the scoliotic cohort.

4.4 Sources of measurement error.

There are three potential sources of error that could occur in the measurement of the subjects in this thesis. The first is related to the ISIS2 system itself, in particular in the calibration of the system and the reliability of the measures produced. The second source of error is in the preparation of the subject for the taking of the ISIS2 image, including the palpation of the bony landmarks and the placement of the stickers used in the analysis of the image. The third is associated with the later identification of the torso points from the captured ISIS2 image.

The assessment of these potential sources of error has been performed in three stages as reported here. Of note, only the assessment of intra and inter-observer variability of the identification of the torso points was performed as part of this thesis. The variability of the preparation of the subject for the ISIS2 image and any errors within the system itself were performed prior to this thesis [78].

4.4.1 Error inherent in the ISIS2 system.

The error inherent in the ISIS2 system is best described in the methods paper describing the ISIS2 system by Berryman et al [78] where the issue of error within the acquisition of the images is addressed. The paper notes that there is an error in the fringe frequency (the horizontal projected lines on the back) of approximately 0.2% and that this converts to a level of accuracy of measurement to $\pm$ 1 mm. The paper further notes that an accuracy of $\pm$ 1mm is less than the amount of movement seen during normal respiration, which would be more with a deeper breath [78].

4.4.2 Error through the application of the stickers and preparation for the ISIS2 image.

In 2012, as part of the use of the ISIS2 system for standard clinical care, the variability of repeated measures was undertaken to address the variability caused through the preparation of the subject for an ISIS2 image. This was not undertaken as part of this thesis and was not published. Bland Altman analysis [190] was used to identify the mean difference along with the 95% limits of agreement for repeat measures. This included the removal of the stickers and application of new stickers by a different individual along with asking the subject to leave the measurement frame, walk around the room and come back to the measurement frame for a fresh image. Bland Altman analysis [190] does not provide a value of statistical significance, rather numerical values in the units of the original measures. Bland Altman analysis [190] establishes the numerical difference between the mean values of the two data sets along with the 95% confidence interval of that difference, known as the 95% limits of agreement. The interpretation of those measures is assessed relative to the original data. There were 35 sequentially measured, different individuals that formed the basis of this audit.

The result of the audit are shown in Table 9. All of the standard ISIS2 output parameters are shown. The parameter 'Lateral asymmetry' is the equivalent of the 'main' parameter in this thesis and represents the largest measured coronal deviation of the spine.

The most useful indication of variability from Table 9 is the measurement of back length as this is the only linear measure common between the audit of reliability and the work in this thesis. The back length measure has a mean difference of approximately 6 mm with 95% limits of agreement of approximately 15 mm. All of the other measures, particularly those which are discussed in the results chapters of this thesis, have a mean difference of less than 2°.

Table 9: Audit of the variability of repeated measures using ISIS2

Parameter	Mean	Standard deviation	Limits of agreement
Back length (mm)	5.71	7.57	-9.66 to 21.09
Pelvic rotation (°)	0.43	2.35	-4.36 to 5.21
Flexion / extension (°)	0.23	2.31	-4.48 to 4.93
Coronal imbalance (mm)	0.63	9.16	-19.25 to 18.00
Lateral asymmetry (°)	0.43	3.44	-6.56 to 7.42
Max skin angle (°)	- 0.18	1.41	-2.94 to 2.58
Min skin angle (°)	- 0.01	1.44	-2.84 to 2.82
Kyphosis angle (°)	1.46	2.91	-4.46 to 7.38
Lordosis angle (°)	1.09	4.17	-7.39 to 9.56
Volumetric asymmetry left	0.11	2.19	-4.34 to 4.57
Volumetric asymmetry right	0.34	4.12	-8.02 to 8.71

4.4.3 Error from the identification of the torso points.

Method. Intra and inter-observer error in the identification of the torso points following the capture of the ISIS2 image was assessed. Using a random number generator, 33 images were identified. These were then analysed by the author for the identification of the torso points and this information was recorded separately to the main database. The same 33 images were analysed by a different individual (Dr Berryman) for inter-observer error. The images were then analysed again after one month by the author for intra-observer error.

Both intra and inter-observer error were assessed using two different methods. As each torso point had an x, y and z coordinate, the location of these coordinates were compared using Bland Altman analysis [190]. The disadvantage of Bland Altman analysis in this scenario is that each of the x, y and z values for each torso point is examined in isolation and not referenced as part of the larger 3D shape. Thus Procrustes analysis was used. The methods behind Procrustes analysis are described in Chapter 8. In brief, Procrustes analysis is a method that removes the effects of location, rotation and scale from a series of shapes with common landmarks, leading to a mean shape [185]. Statistical tests are described that assess the significance of the difference between the mean shapes by Hotelling [191], Goodall [192] and James [193].

Results. The results of the intra-observer and inter-observer error using Bland Altman analysis is as documented in Tables 10 and 11. In summary, when assessing the intra-observer error, the mean difference is less than 1 mm apart from the points representing the waist in the y axis (WaistDiffHt) on both the right and left sides. For the waist points in the y axis the mean difference is less than 2 mm. For inter-observer error, when using a Bland Altman analysis, the mean difference is less than 5 mm for all torso points in all planes. For both intra and inter-observer error, when using all of the statistical tests used within a Procrustes analysis, there was no significant difference found (Table 12).

Table 10: Intra-observer reliability of torso point identification

Torso point	Mean difference (mm)	Upper limit of agreement (mm)	Lower limit of agreement (mm)
Shoulder point x on right	0.42	3.79	-2.94
Shoulder point y on right	0.97	6.79	-4.85
Shoulder point z on right	-0.90	3.19	-4.99
Axillary point x on right	0.14	2.75	-2.46
Axillary point y on right	-0.39	4.45	-5.23
Axillary point z on right	-0.03	6.44	-6.51
Waist point x on right	0.32	4.97	-4.33
Waist point y on right	1.56	10.71	-7.60
Waist point z on right	-0.41	6.79	-7.60
Shoulder point x on left	0.80	5.83	-4.23
Shoulder point y on left	-0.15	5.67	-5.97
Shoulder point z on left	0.46	6.44	-5.52
Axillary point x on left	0.50	5.12	-4.13
Axillary point y on left	-0.18	7.22	-7.57
Axillary point z on left	0.40	3.43	-2.64
Waist point x on left	0.15	5.43	-5.12
Waist point y on left	-1.62	19.03	-22.27
Waist point z on left	0.18	6.60	-6.24

Table 11: Inter-observer reliability of torso point identification

Torso point	Mean difference (mm)	Upper limit of agreement (mm)	Lower limit of agreement (mm)
Shoulder point x on right	-0.62	2.23	-3.47
Shoulder point y on right	1.39	4.76	-1.98
Shoulder point z on right	-0.12	3.75	-3.98
Axillary point x on right	-0.26	2.40	-2.92
Axillary point y on right	4.06	9.43	-1.31
Axillary point z on right	2.12	8.03	-3.78
Waist point x on right	-0.76	0.99	-2.51
Waist point y on right	-0.25	14.08	-14.59
Waist point z on right	0.84	5.47	-3.79
Shoulder point x on left	-1.36	2.57	-5.29
Shoulder point y on left	-0.18	3.04	-3.40
Shoulder point z on left	-0.67	2.61	-3.94
Axillary point x on left	-0.78	2.81	-4.37
Axillary point y on left	3.27	10.55	-4.01
Axillary point z on left	1.70	7.08	-3.68
Waist point x on left	-0.58	2.02	-3.17
Waist point y on left	-1.62	16.37	-19.61
Waist point z on left	-1.48	6.72	-9.68

Table 12: Procrustes analysis of inter and intra-observer variability of torso point indentification

	Intra-oberver	Inter-observer
Root mean square difference	0.898	1.861
Hotelling's test	1.00	0.97
Goodall's test	1.00	1.00
James' test	1.00	0.97

4.4.4 Sum of errors.

To combine these errors gives a mean variability within any measure made between individuals of 11 mm (the sum of a mean of approximately 6 mm variability in back length from the 2012 work, 1 mm from the paper of Berryman et al [78] and 4 mm from the inter-observer difference, the largest amount from all of the measures, from this thesis).

4.4.5 Conclusion of sources and amount of measurement reliability.

An assessment of potential error within an experiment must take into account all the sources of error at every stage of that experiment. Across the identified potential sources of error identified in this thesis, the mean value is 11 mm. To put this in to some context, the measures from the papers of Akel et al [148] and Vercauteren et al [144] both quote a range of values for different parameters measured in children without spinal or thoracic deformity to identify a range of normality for body shape. In the paper by Akel et al [148], the difference in shoulder height in children who believed their shoulders to be level was 7.5 ± 5.8 mm. The paper by Vercauteren [144], which is a review of children from Belgian schools, notes values of between 5 mm and 15 mm. The value of 11 mm is similar to the values from the Akel [148] and Vercauteren papers [144]. Within the values quoted from the literature, the variability of the measures and techniques used within this thesis are very similar. Based on that observation, it is felt by the author that the amount of variation demonstrated as occurring within the data for this thesis is acceptable and any change documented that is more than 11mm is outside the measurement error of the data acquisition.

5 Proof of normality of the non-scoliotic cohort using published growth standards.

5.1 Introduction.

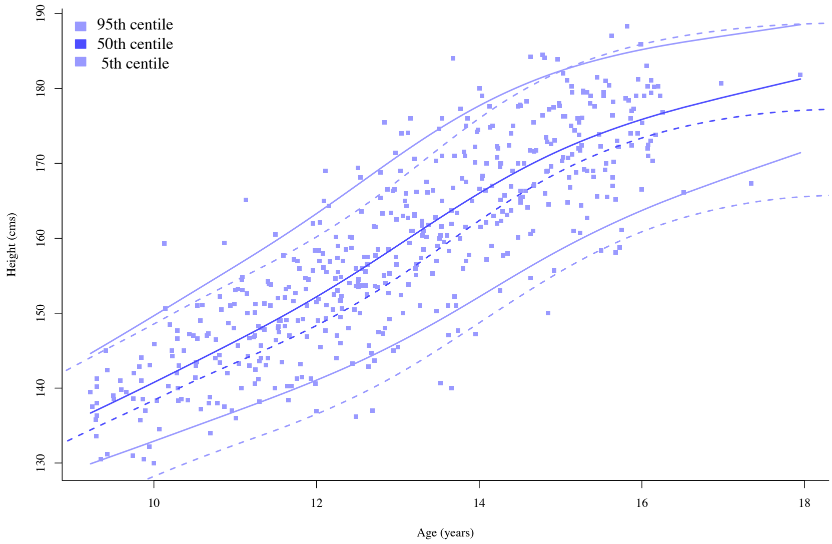
To draw conclusions from a sample population that are applicable to the wider population, it is important to establish that the sample is representative of that wider population. In this case, it needs to be established that the 'normal' non-scoliotic population is a representative sample of the wider population of children and adolescents. This is possible through the comparison of parameters from the sample population against agreed standards for that parameter in the wider population. In this case the standards available are the UK-WHO growth standards consisting of standing height, weight and body mass index (BMI) from the Royal College of Paediatrics and Child Health (RCPCH) [82, 86] and for sitting height and sitting to standing height ratio from Fredriks et al [83]. The test of normality is based on an assessment of the distribution of the sample data, in this case the parameters from the non-scoliotic cohort in comparison to the appropriate growth standard.

5.2 Methods.

The standards for standing height, weight and BMI were created showing the median (50th centile) along with the 5th and 95th centiles. In a similar fashion, the standards for the sitting height and sitting to standing height ratios were also created. The data from the non-scoliotic cohort were plotted to the appropriate standard. The median, 5th and 95th centiles of the non-scoliotic data were calculated and fitted curves produced to represent this were also plotted. The percentage of data from the non-scoliotic cohort below the 5th centile, the median and above the 95th centile of the growth standard is documented. For a perfect fit of the non-scoliotic data on to the growth standards, there would be exactly 50% of the non-scoliotic data above and below the median value of the growth standard, with 5% of the non-scoliotic data below the 5th centile and 5% above the 95% centile of the growth standard. The standards were extracted for plotting and the non-scoliotic data added in R [189] using the childsds package [87].

5.3 Results.

Figures 33 to 37 show the standards for standing height, weight, BMI, sitting height and sitting height to standing height ratio with the measured data for that parameter plotted on to the standard. Table 13 shows the percentage of the non-scoliotic cohort below the 5th centile, the median and over the 95th centile for males and females across the different growth standards.



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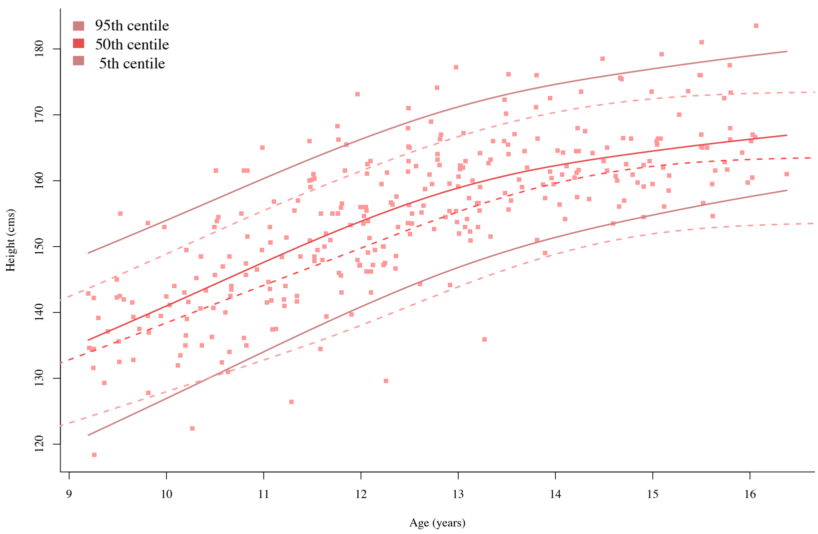
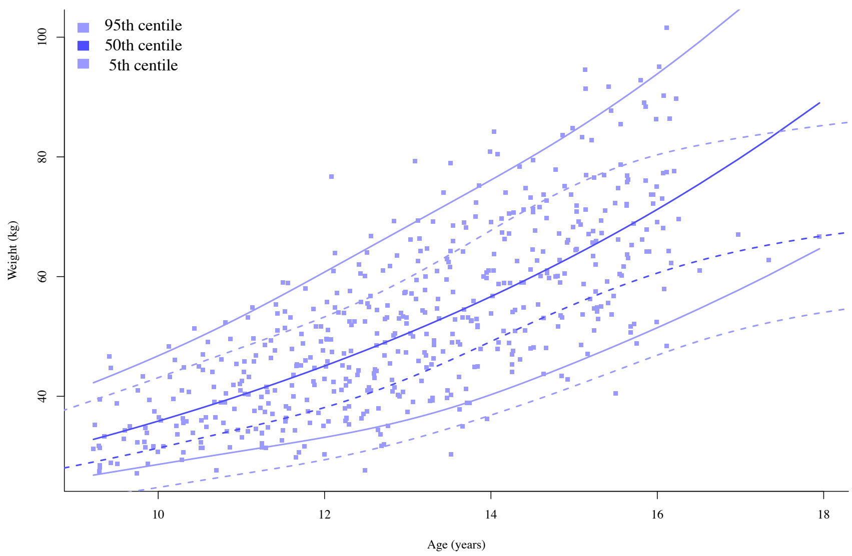


B

Figure 33: Standing height (cm) against age (years) for the growth standard with the males (A) and the females (B) in the non-scoliotic cohort as individual points. The solid lines represent the median and the 5th and 95th centiles for the non-scoliotic data, with the dashed lines representing the median and 5th and 95th centiles for the growth standard. The growth standard was redrawn from data provided by RCPCH [82, 87].



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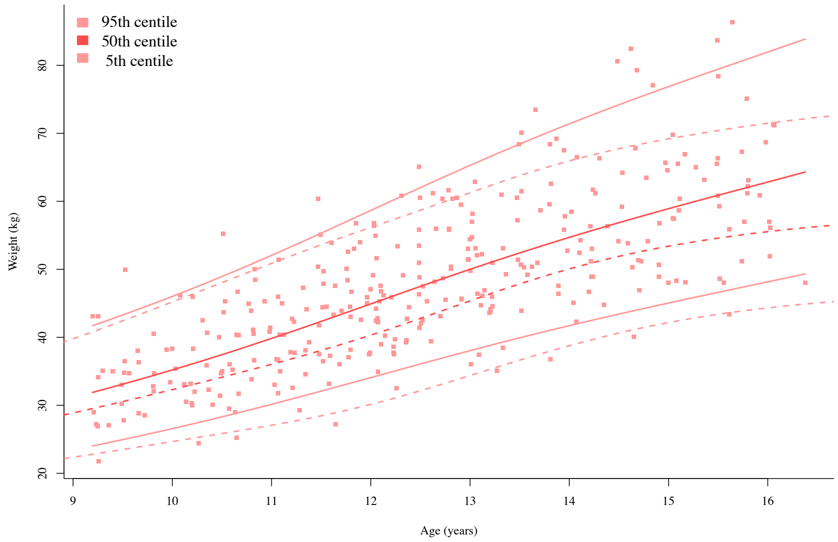
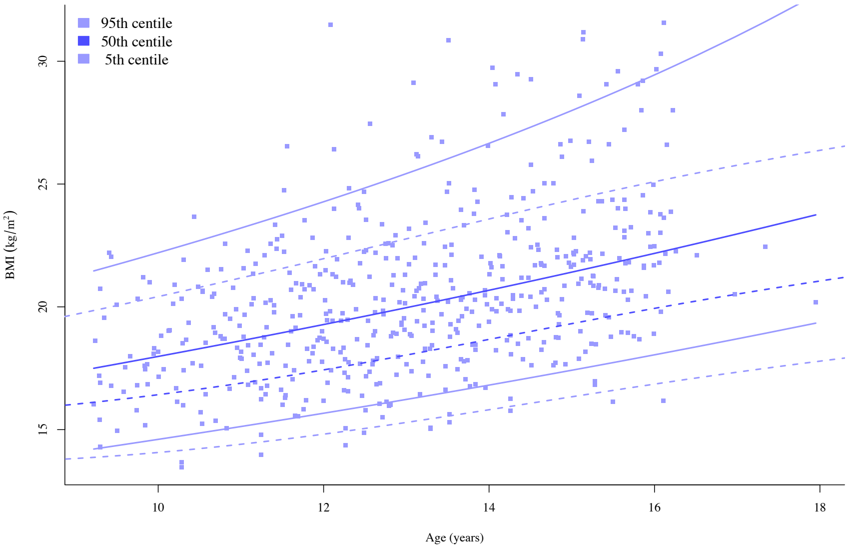


B

Figure 34: Weight (kg) against age (years) for the growth standard with the males (A) and the females (B) in the non-scoliotic cohort as individual points. The solid lines represent the median and the 5th and 95th centiles for the non-scoliotic data, with the dashed lines representing the median and 5th and 95th centiles for the growth standard. The growth standard was redrawn from data provided by RCPCH [82, 87].



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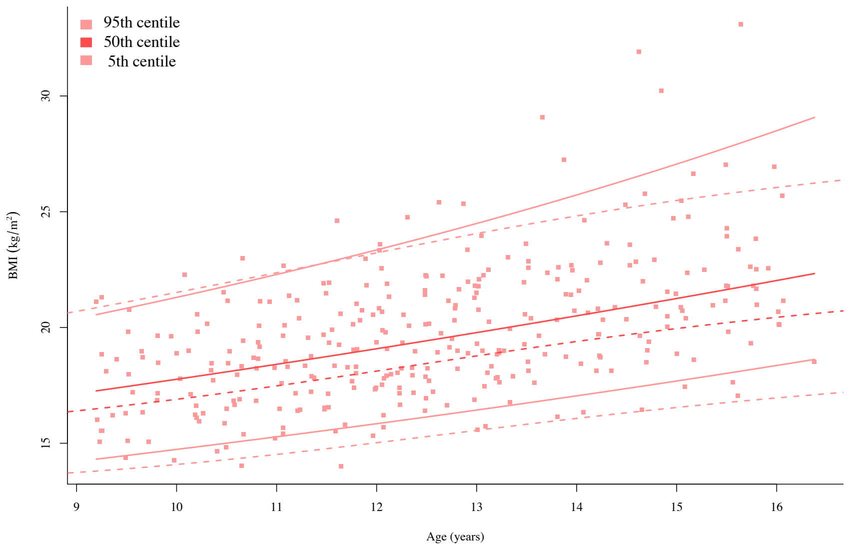
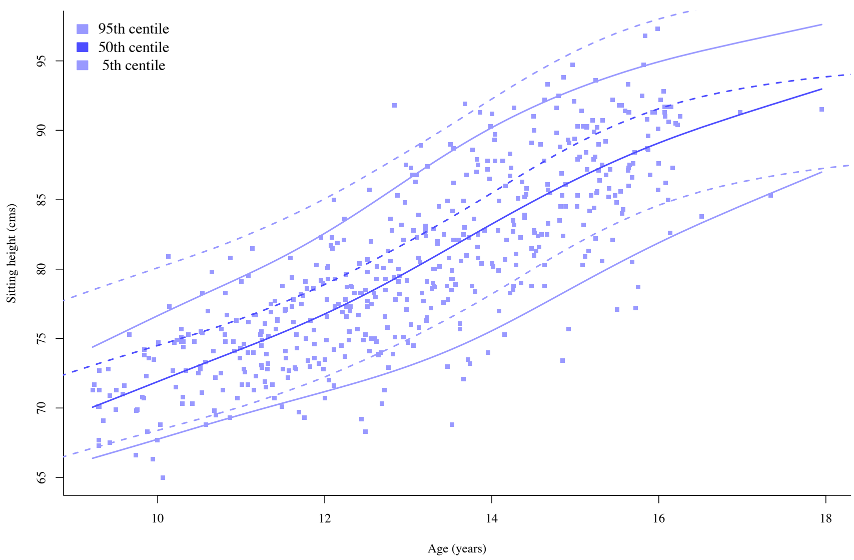


B

Figure 35: BMI (kg/m^2) against age (years) for the growth standard with the males (A) and the females (B) in the non-scoliotic cohort as individual points. The solid lines represent the median and the 5th and 95th centiles for the non-scoliotic data, with the dashed lines representing the median and 5th and 95th centiles for the growth standard. The growth standard was redrawn from data provided by RCPCH [82, 87].



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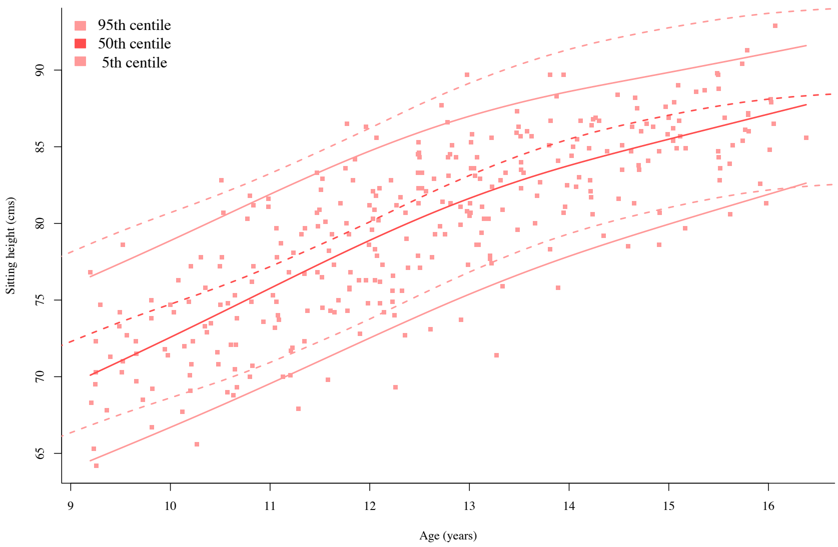
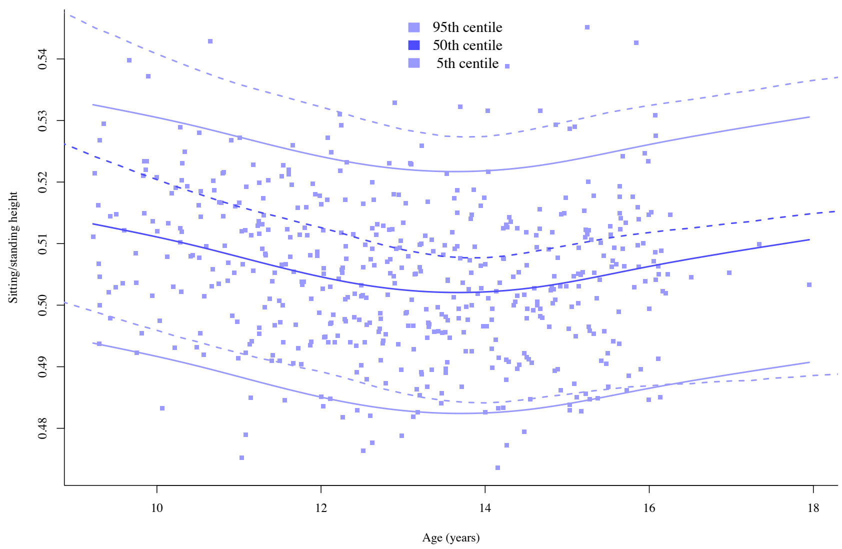


B

Figure 36: Sitting height (cm) against age (years) for the growth standard with the males (A) and the females (B) in the non scoliotic cohort as individual points. The solid lines represent the median and the 5th and 95th centiles for the non-scoliotic data, with the dashed lines representing the median and 5th and 95th centiles for the growth standard. The growth standard was redrawn from data provided by Fredriks et al [83, 87].



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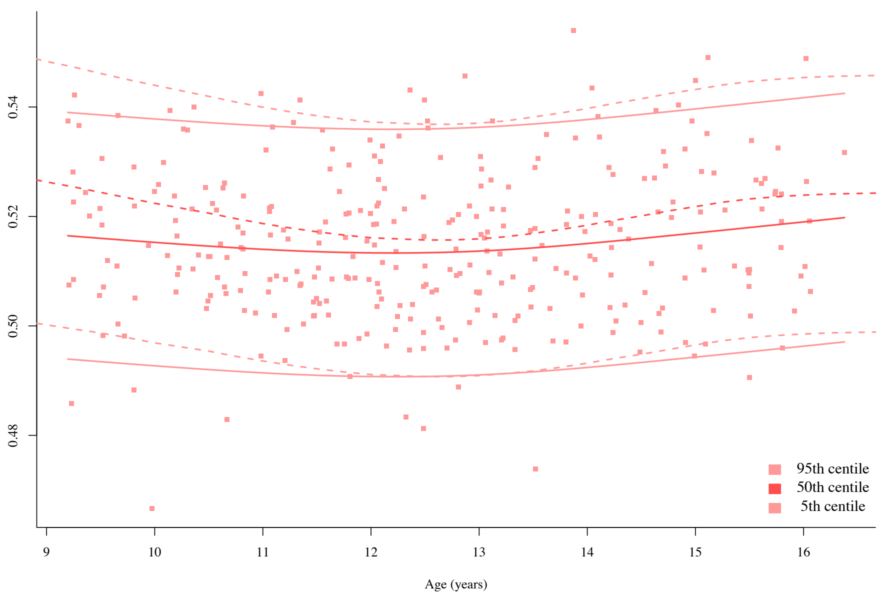


B

Figure 37: Sitting height to standing height ratio against age (years) for the growth standard with the males (A) and the females (B) in the non-scoliotic cohort as individual points. The solid lines represent the median and the 5th and 95th centiles for the non-scoliotic data, with the dashed lines representing the median and 5th and 95th centiles for the growth standard. The growth standard was redrawn from data provided by Fredriks et al [83, 87].

Table 13: The percentage of the non-scoliotic cohort below the 5th centile, the median and over the 95th centile for males and females across the different growth standards.

Parameter	Sex	% under the 5th centile	% under the median	% over the 95th centile
Standing height	Male	6	50	5
	Female	4	53	6
Weight	Male	5	50	5
	Female	4	50	5
BMI	Male	4	50	5
	Female	6	50	5
Sitting height	Male	5	50	6
	Female	5	50	5
Sitting: standing height ratio	Male	4	50	5
	Female	3	52	7

5.4 Conclusion.

An inspection of the distribution of the non-scoliotic data against the growth standards shows that most of the data lies around the median and within the 5th and 95th centile for both males and females. Whilst the fitted lines for the median, 5th and 95th centiles are not exact, the amount of data above and below these lines is very similar to that if the models were exact. Visual inspection of the data points themselves with respect of the growth standards along with the fitted models show the data to be normally distributed.

There are differences between the amount of data above and below the median for males and females. Males were generally taller and females generally heavier than the corresponding UK-WHO standard. This may be because the non-scoliotic cohort were from a private, fee paying school in the UK, and some of pupils are residential.

Consequently they are fed three meals a day, seven days a week and could be expected to come from a relatively affluent background. This, when compared to some of the populations used for the UK-WHO standards may represent a mismatch. Additionally the UK-90 data that contributes to the UK-WHO is from the 1990s. It is to be expected that health and thus growth and development will have improved for most people since the 1990s to date [194].

The data for the non-scoliotic cohort is mainly distributed around the median on the sitting height standard. As noted previously within the literature review (Section 2.1), northern mainland Europeans are some of the tallest populations in the world [88] and any differencse between the standard and the non-scoliotic data may be due to this. Given the historically similar racial and historical background within Northern Europe [89], the comparison between the populations of the UK and the Netherlands is considered appropriate and the variability in the relative contributions to height from trunk and leg length will be similar. Thus the use of the sitting height to standing height ratio removes the majority of the differences between the UK and the Netherlands caused by measures of absolute height.

Given this analysis of the non-scoliotic growth relative to the growth standards, it is concluded that the non-scoliotic population, whilst not a perfect fit to the growth standards, is a representative sample of the wider population.

6 The normal child.

6.1 Introduction.

In the management of any deformity of the body, it is imperative that there is an understanding of what is the range of 'normal'. This is because the treatment of a deformity invariably requires intervention that will attempt to recreate normality as far as possible. This intervention prevents further deformity from occurring and may improve function. The literature covering the variation in body shape in children without spinal deformity has already been discussed in Chapter 2. The major weakness of these studies are their cross sectional designs and that, in the main, they focus on one part of the torso in isolation, rather than looking at the torso as a whole.

This chapter documents the change in parameters of the spine and shape of the torso in the non-scoliotic cohort. These parameters are sub-divided for sex. The parameters are examined in different ways, relating the parameter to increasing age and to other parameters such as comparing torso point position and spinal shape for example. As these measures were collected serially, from the same children on multiple occasions over a seven year period, longitudinal analysis techniques are used, particularly using linear mixed effect modelling. The chapter is subdivided into sections and these are defined in the methods that follow. This chapter defines normative values for a non-scoliotic cohort for future work.

6.2 Methods.

There are many anatomical parameters that are measured and analysed in this chapter. Thus to simplify the chapter structure, the text has been divided into sections of measures of a particular aspect of the body. For each of these sections, there will be a review of the methods pertinent to the analysis followed by the results and conclusions drawn from the parameters presented.

The sections presented here are:

1. Measures of torso size. This group is comprised of measures of standing height, weight, sitting height and BMI. These are complemented by the measure of back-length, defined as the vertical distance in 2D between the VP and the sacrum. Also shown are parameters of width, defined as both the horizontal distance in 2D between the axillae (total axillary width) and the waist points (total waist width). These parameters are grouped together as they represent the growing torso as a whole.

2. Measures of torso asymmetry. This group is comprised of parameters relating to the individual 2D torso points of the shoulders, axillae and waist as defined in Figure 27 and Table 4 in the Methods chapter. This analysis identifies any difference in the location of these points between the left and right sides of the torso.

3. Measures of spinal shape. This group is comprised of parameters assessing the shape of the spine in both the coronal plane (measured as the coronal curve) and the sagittal plane (measured as kyphosis and lordosis). The relationship of coronal curve with asymmetry of the torso points from the measures of torso asymmetry is also demonstrated.

4. Measures of 3D shape. This group comprises measures of the 3D nature of the torso, namely the position of the most prominent points on the back and volumetric asymmetry as defined in Figure 29 and Table 5 in the Methods chapter.

In all groups the effect of increasing age and the effects of sex on the parameters are assessed.

6.3 Measures of torso size.

6.3.1 Methods.

All analysis of the measured child was performed using R [189]. The results for each parameter are illustrated first as a composite box and whisker plot allowing a comparison of males and females and second as a spaghetti plot, showing an assessment of any trend to be seen using the ggplot2 package [195]. The composite box plots were created by subdividing the data into sex and then into segments of 1 year (i.e. those indicated as being 9 years at the time of analysis had ages of ≥ 9 years and < 10 years). The format of the box and whisker plot is defined in Figure 30. The spaghetti plots demonstrate the data in a different way. Each individual line (light blue for males and pink for females) represents a single subject. The dark blue line is a LOESS line (locally estimated scatterplot smoothing) [196]. This shows a local mean for the data and demonstrates any change with increasing age. The grey funnel around the LOESS line (more easily seen at the youngest and oldest age groups) is the 95% confidence interval of the LOESS line.

LOESS is a local regression method that uses both a least squares regression technique along with non-linear regression, providing a smoothing function to scatterplot data [197]. The benefit to this technique is its ability to inspect the regression curve, identifying both the relationship described by the regression along with any effects of outliers within the data [197]. Being non-linear, LOESS does not force a linear (and thus a fixed, straight line relationship between the variables) regression onto the data inappropriately. Other scatterplot smoothing functions can be used such as LOWESS (locally estimated weighted scatterplot smoothing), regression and smoothing splines, kernal regression and others [197]. The advantage of LOESS is its ability to provide the user with a regression without having to specify a fitting function in advance. The disadvantage of LOESS is the inability to return a mathematical function that explains the regression. LOESS will be used here to demonstrate the mean change in the parameters against the increasing age of the child, based on not being able to assume in advance that the relationship will be linear [80].

To assess the statistical significance of the effect of age and sex on the measured parameter, linear mixed effect modeling (LMEM) was performed using the lme4 package [198]. This method of analysis allows for the error that may occur with repeated measures from the same individual over time [199]. Previous methods of longitudinal analysis have included multivariate analysis of variance (MANOVA) and generalised estimating equations (GEE analysis) [200]. To be compliant with the required assumptions for the analysis of linear data, the data must have interdependence [199]. This can be a problem when sampling measures from the same individual on multiple occasions, as is required for a longitudinal analysis, as those measures are not interdependent. LMEM solves the interdependence problem by accepting (and thus assigning) an amount of random error to each measure [199] making, for the purposes of the analysis, each measure independent of all other measures. The other benefit of LMEM is that the whole data set is used, rather than just an average of that data set [199]. To perform LMEM, it is necessary to identify in advance the fixed effects (those that are measurable) and the random effects (representing the inherent error in the experiment that cannot be controlled for and must be accepted).

In the linear mixed effect model the fixed effects were age and sex with the random effect of repeated measures. The models were constructed using a random intercept and random slope method. To examine the significance of one of the fixed effects, models with and without that effect were created and then compared using analysis of variance (ANOVA) [199]. The level of statistical significance was pre-defined as $p < 0.05$. In each case the model was examined for 'goodness of fit' through an assessment of the residuals, both as a plot of the residuals and as a qq plot. The effect of outliers was assessed and the models adjusted if there was an undue effect from an outlier. If the model was unable to converge, the model was simplified, first through removing random slopes, and then moving to a linear model without mixed effects. Linear analysis was chosen following inspection of the data that demonstrated that there was no added benefit from a non-linear analysis through the comparison of the Akaike information criterion (AIC) [201] values for different models constructed of both linear and non linear types. The AIC value for the linear model of standing height was 4770 and for the non-linear model was 5693. The AIC is a technique that allows the user to estimate the relative quality of statistical models, first described by Akaike in 1974 [201]. In essence, there is always a loss of some information from the data set when constructing a statistical model. The better the model, the less the data loss and the lower the value of AIC. The AIC is thus a measure of that data loss and gives the user a measure of which model is the best fit for their data.

When documenting the outputs of LMEM, each fixed effect is reported with a Chi squared value, the p value, the coefficient and the standard error of that coefficient. The Chi squared value is the value derived from that statistical test that underlies the comparison of models performed using the ANOVA test. The p value is the level of significance attached to that Chi squared value and is pre-defined as statistically significant at a fixed value. In this case, the coefficients are the amount that is added to move in the model from each parameter within the fixed effect. For example, within the fixed effect of sex, the coefficient is the amount within the model between males and females. For age, it is the amount describing the difference within the model between the youngest and oldest, i.e. the slope of the model line. The coefficient is always measured in the units appropriate to the model and represents the true size of the association. The standard error is a measure of variability, or uncertainty, of the coefficient. The construction of the models is such that a positive coefficient describes the change from female to male or from the youngest age to the oldest age, and vice versa for a negative coefficient. This is demonstrated graphically in Figure 38 where the coefficient for standing height (the left hand plot) is the slope of the line measured in cm/yr and the coefficient for sex (the right hand plot) is the difference between males and females measured in cm.

Note in all of the composite box plots, a mean and 95% confidence interval is not shown for males aged 17 as there was only two individuals in this group.

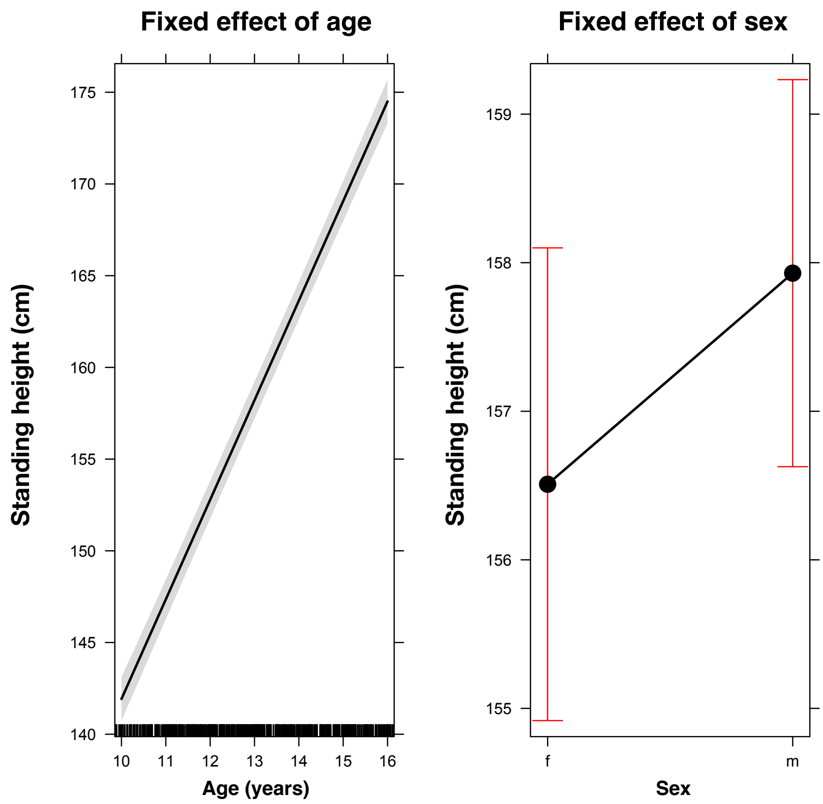


Figure 38: An example of the fixed effects seen in the model for standing height in the non-scoliotic cohort. The plot shows the fixed effect of age (thicker line) with the 95% confidence interval (grey funnel) on the left and the fixed effect of sex (with 95% confidence intervals for both male and female) on the right.

6.3.2 Results.

Standing and sitting height are measured in centimetres (cm). Measures of backlength and measures of width at the axillae and waist are shown in millimetres (mm). Weight is measured in kilogrammes (kg) and BMI (kg/m^2). Table 14 shows the significance of both age and sex on the measured parameters. Figures 39 to 45 show composite box plots and spaghetti plots for males and females for each parameter measuring torso size. Figure 38 shows an example of the fixed effects of the linear mixed effect models used and this relates to the coefficients seen in Table 14.

Table 14: The significance and coefficients for the measures of torso size in the non-scoliotic cohort.. For an explanation of the terms in the table see section 6.3.1.

Parameter		Chi squared	p value	coefficient	standard error
Standing height	Age	509.07	< 0.001	5.41 cm/yr	0.19
	Sex	2.31	0.129	1.70 cm	1.03
Sitting height	Age	488.21	< 0.001	2.72 cm/yr	0.05
	Sex	5.73	0.017	-1.24 cm	0.51
Backlength	Age	497.33	< 0.001	19.91 mm/yr	0.38
	Sex	3.16	0.075	-6.34 mm	3.49
Total axillary width *	Age	386.19	< 0.001	14.24 mm/yr	0.37
	Sex	9.64	0.002	10.81 mm	3.31
Total waist width	Age	285.59	< 0.001	7.28 mm/yr	0.26
	Sex	12.48	< 0.001	10.02 mm	2.80
Weight	Age	416.59	< 0.001	5.31 kg/yr	0.13
	Sex	0.40	0.529	-0.71 kg	1.12
BMI	Age	201.55	< 0.001	0.76 $kg/m^2/yr$	0.04
	Sex	0.28	0.590	0.19 kg/m^2	0.36

* One outlier removed that was having undue effects. Removal normalised the data and revealed an acceptable residual and qq plot.

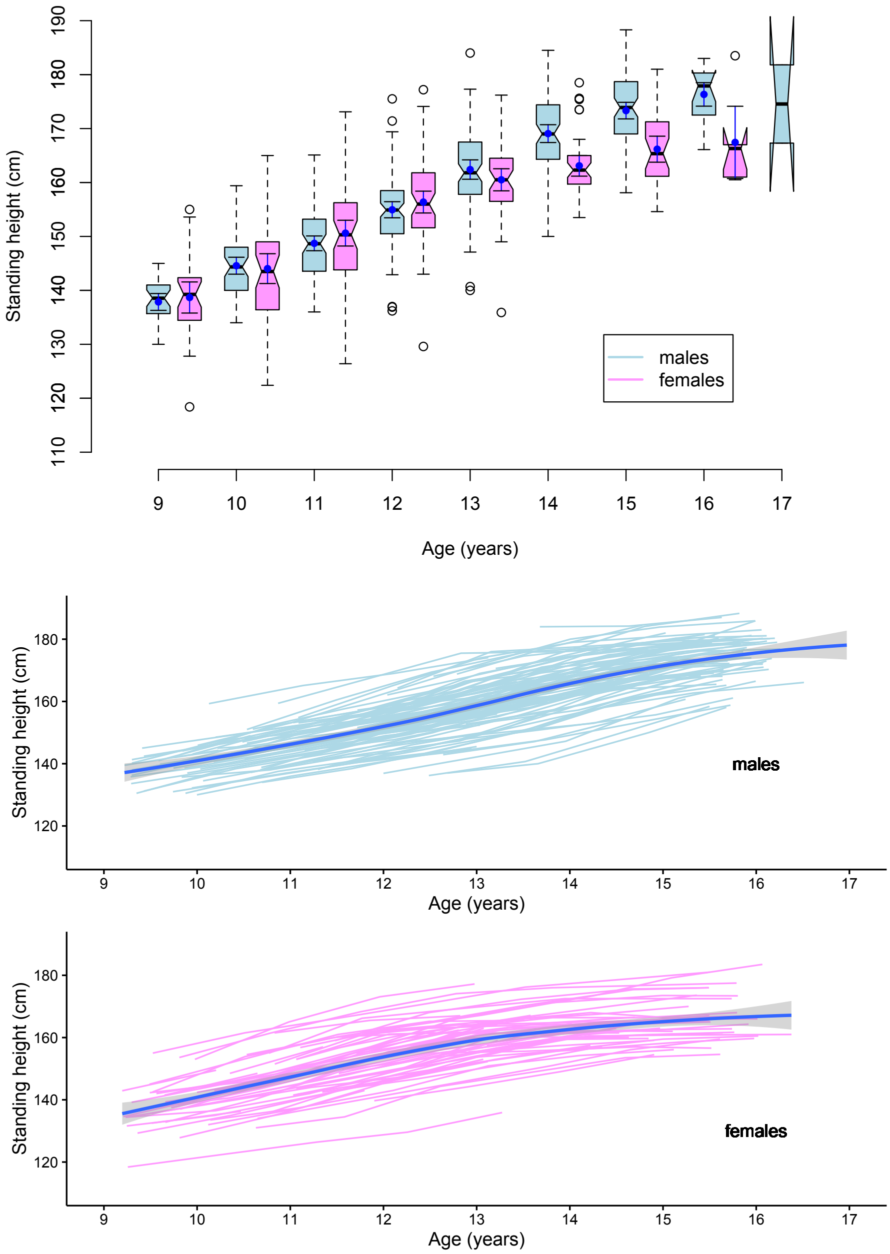


Figure 39: Standing height (cm) versus age (years) for males and females in the non-scoliotic cohort.

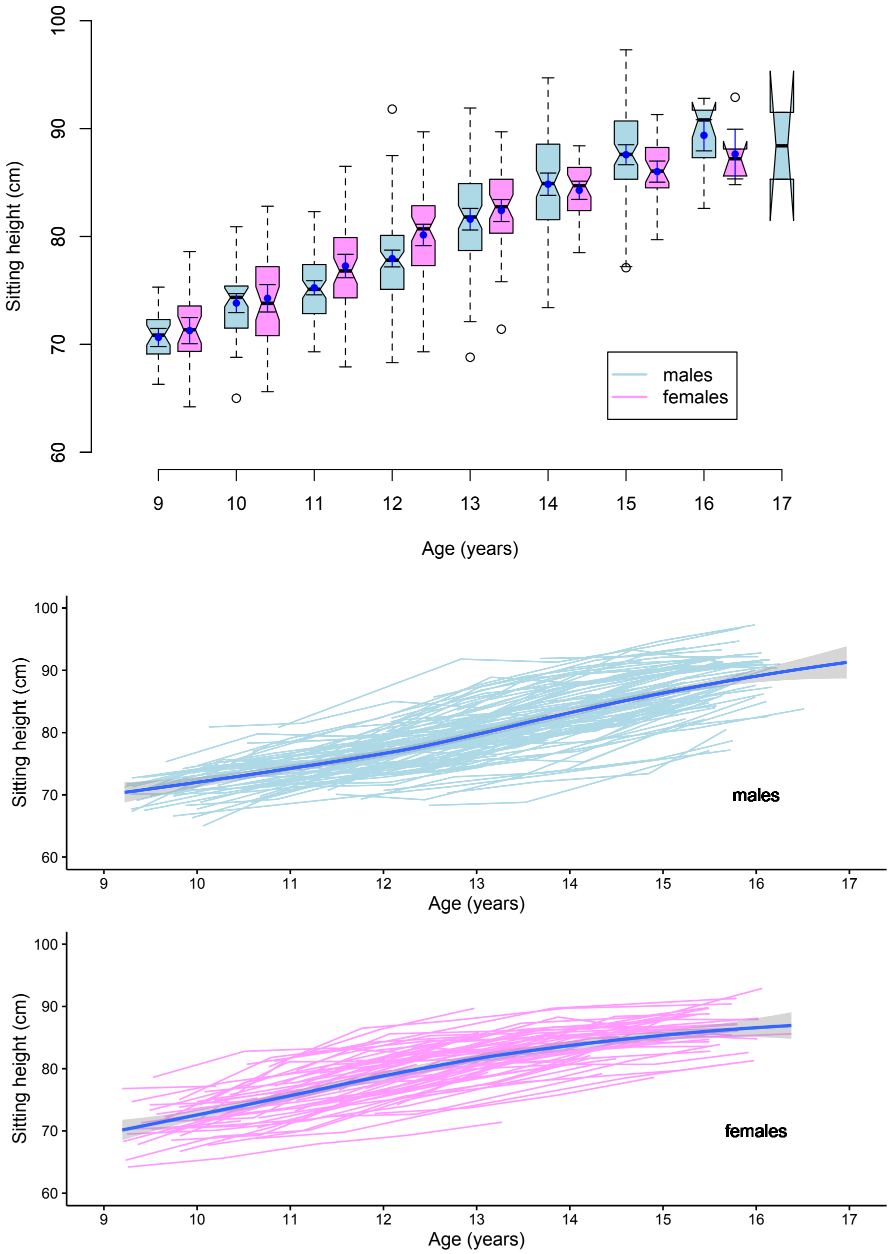


Figure 40: Sitting height (cm) versus age (years) for males and females in the non-scoliotic cohort.

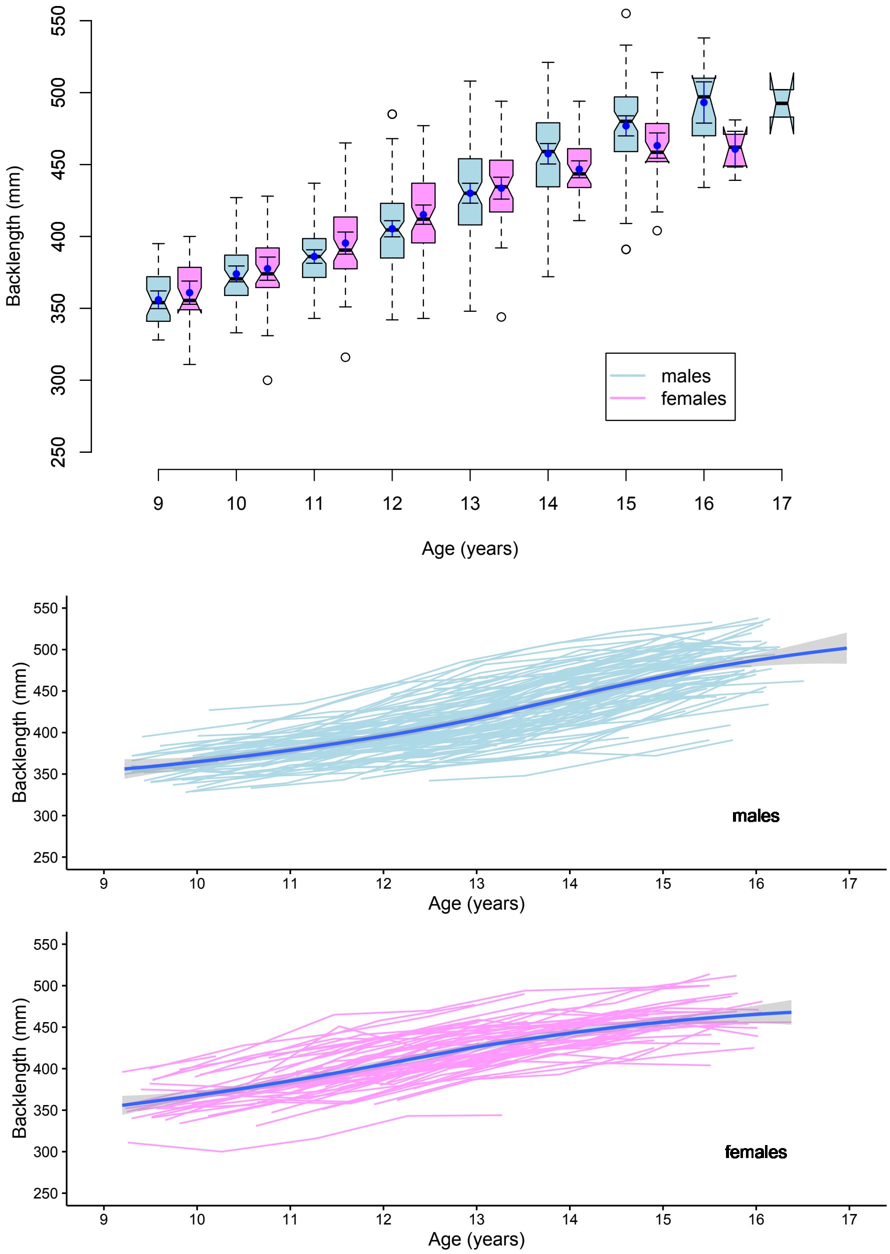


Figure 41: Backlength (mm) versus age (years) for males and females in the non-scoliotic cohort.

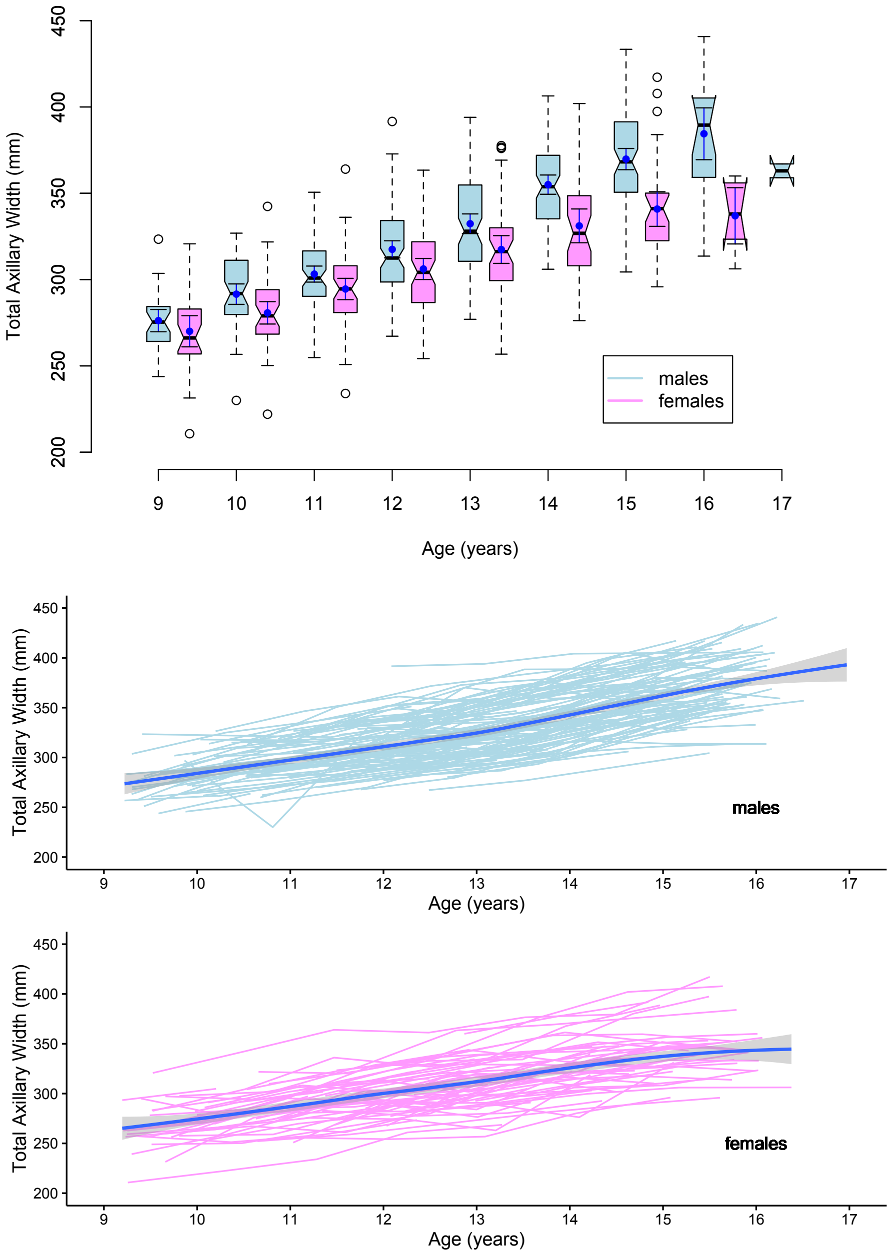


Figure 42: Total axillary width (mm) versus age (years) for males and females in the non-scoliotic cohort.

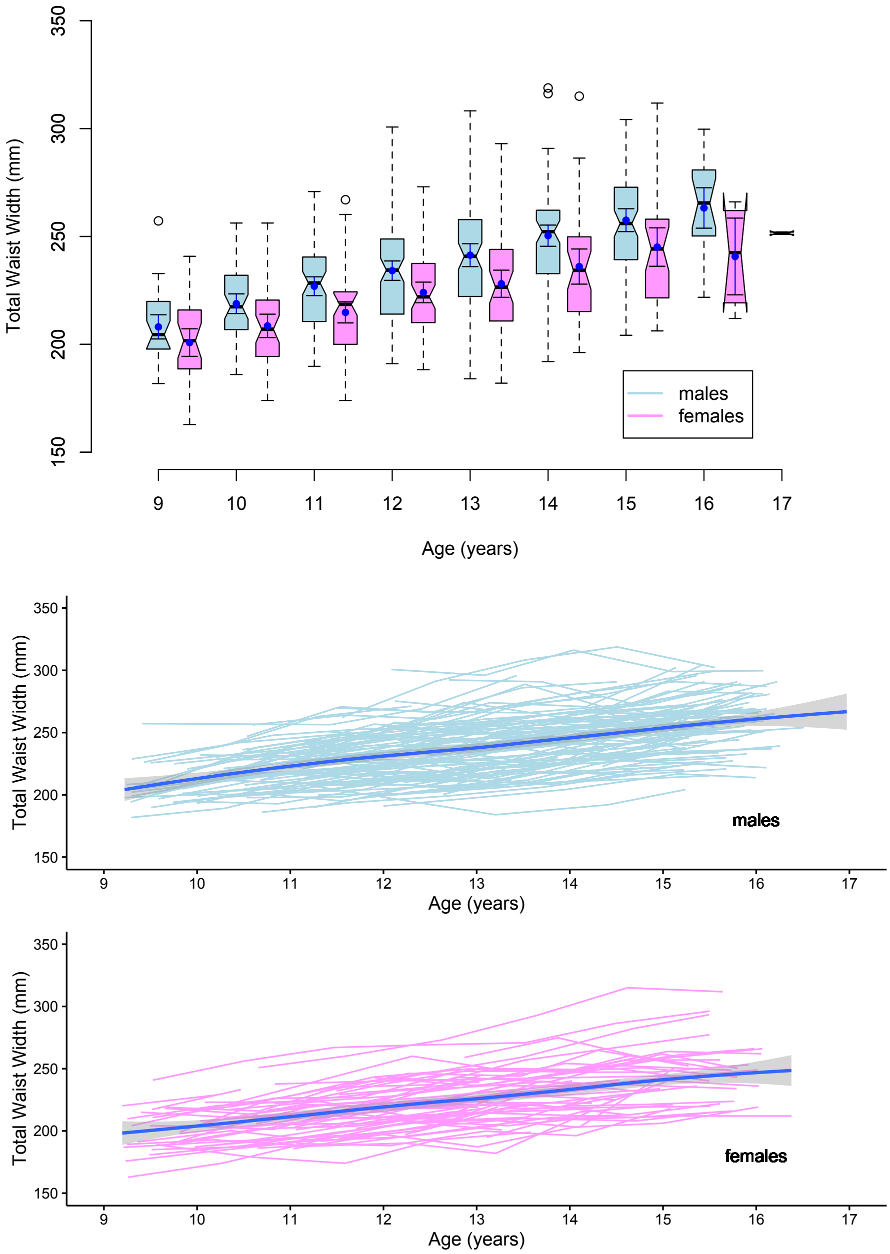


Figure 43: Total waist width (mm) versus age (years) for males and females in the non-scoliotic cohort.

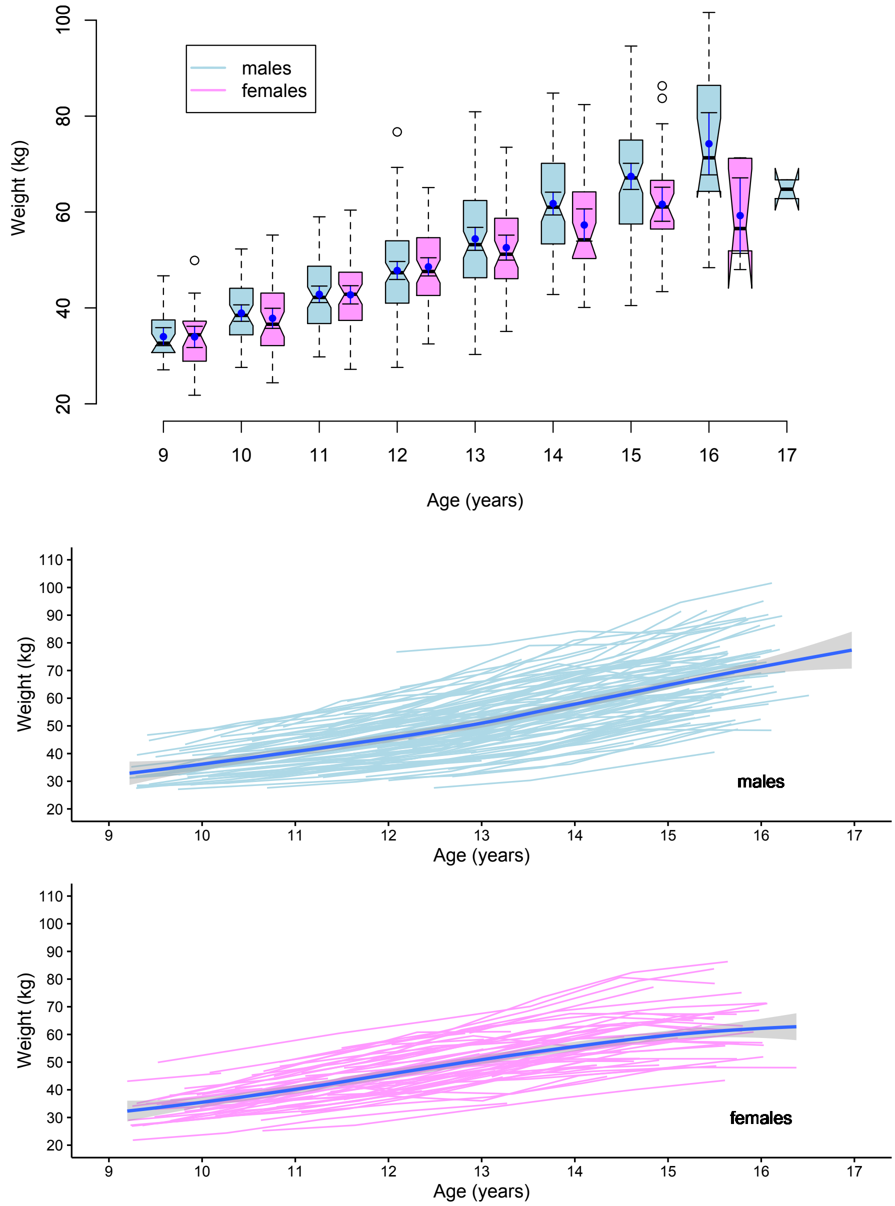


Figure 44: Weight (kg) versus age (years) for males and females in the non-scoliotic cohort.

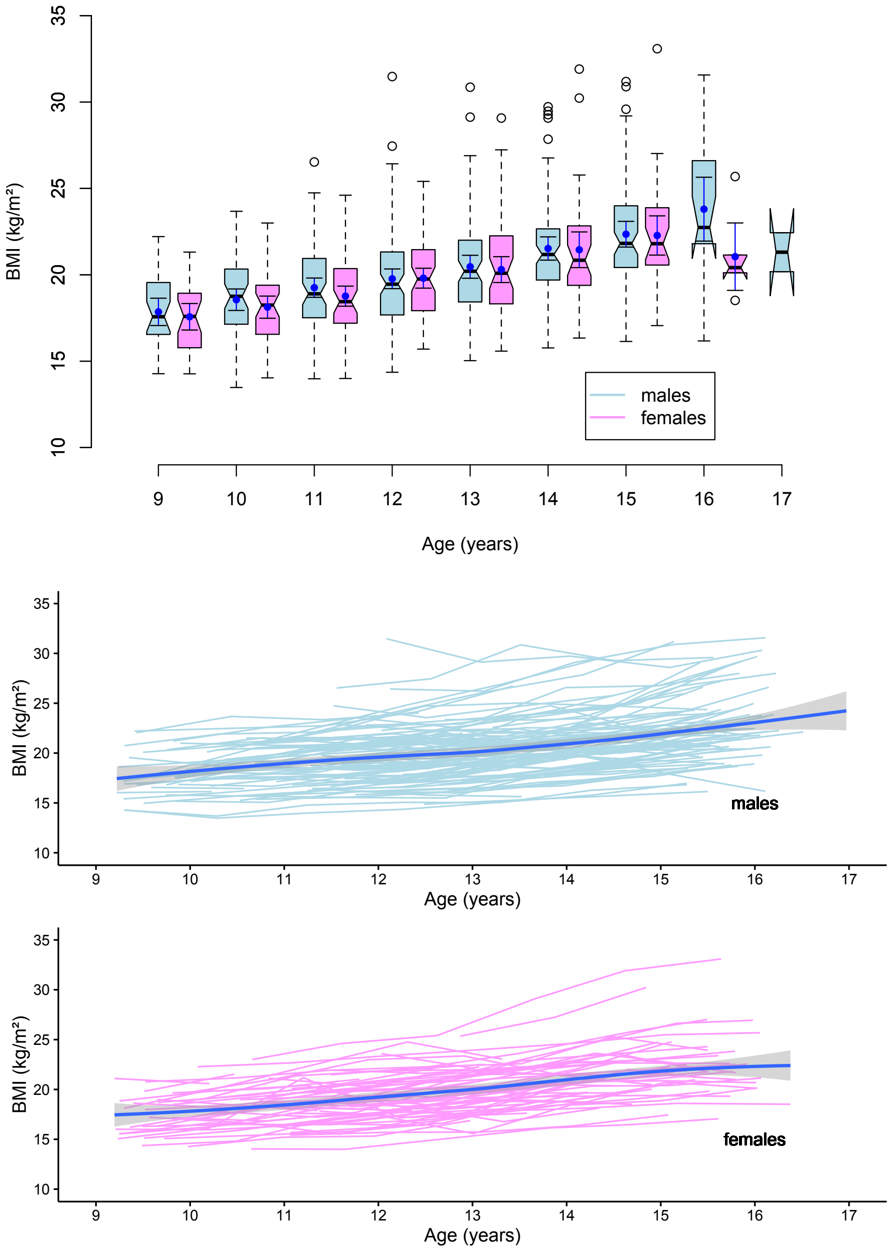


Figure 45: BMI (kg/m^2) versus age (years) for males and females in the non-scoliotic cohort.

6.3.3 Conclusions.

There is an increase in all measures with the ageing of the child both for males and females. Males are generally bigger in all parameters than females. Females seem to slow or halt in the increase of a parameter from around the age of 15 years whereas in males this either does not occur or the slowing is less marked. Table 14 shows the significance of the effects of age and sex on the measured parameters. Age is statistically significant in all parameters with all parameters increasing with age. Sex on the other hand is only a statistically significant factor with parameters of torso width and sitting height. Males are wider than females when measured at the axillae and waist.

6.4 Measures of torso asymmetry.

6.4.1 Methods.

A similar graphical and statistical analysis with composite box plots and spaghetti plots has been used for the parameters that are measures of torso asymmetry as seen for the parameters of measures of torso size (Figure 27 and Table 4 in the Methods chapter). The parameter presented on the y axis for all of the plots is the difference between the position of the torso point on the right hand side of the body in 2D compared to the left. By design, if the right sided point was found to be further away from the midline in the x axis (for measures of width) or more superior in the y axis (for measures of height) than the left sided point, this was defined as a positive measure and vice versa. Thus, if the parameter shown is below the zero line, the left is either further away from the midline than the right or higher than the right and demonstrates some asymmetry in that parameter between the sides of the body. Statistical analysis was again performed using linear fixed effects modeling with the lme4 package [198] using the same fixed and random effects as before.

6.4.2 Results.

This section presents the results for the torso points, demonstrated in Figures 46 to 50 and Table 15. All results are in millimetres.

Table 15: The significance and coefficients for the parameters of measures of torso asymmetry in the non-scoliotic cohort. For an explanation of the terms in the table see section 6.3.1. For the definitions of the parameters see Figures 27 and 28 and Table 4.

Parameter		Chi squared	p value	coefficient	standard error
ShDiffHt	Age	26.63	< 0.001	- 0.76 mm/yr	0.14
	Sex	2.68	0.102	- 1.43 mm	0.87
AxDiffHt *	Age	15.16	< 0.001	0.70 mm/yr	0.18
	Sex	3.09	0.079	- 1.85 mm	1.09
WaistDiffHt	Age	0.92	0.338	0.18 mm/yr	0.18
	Sex	0.001	0.971	0.04 mm	1.25
AxDiffOff	Age	3.71	0.054	0.34 mm/yr	0.18
	Sex	2.21	0.137	- 1.46 mm	0.98
WaistDiffOff	Age	2.69	0.101	0.45 mm/yr	0.27
	Sex	4.51	0.034	- 3.09 mm	1.44

* The model did not converge with either a random intercept and slope or random intercept only methodology. A linear model without mixed effects was used to give the coefficients and significance shown. The AIC was very similar in all.

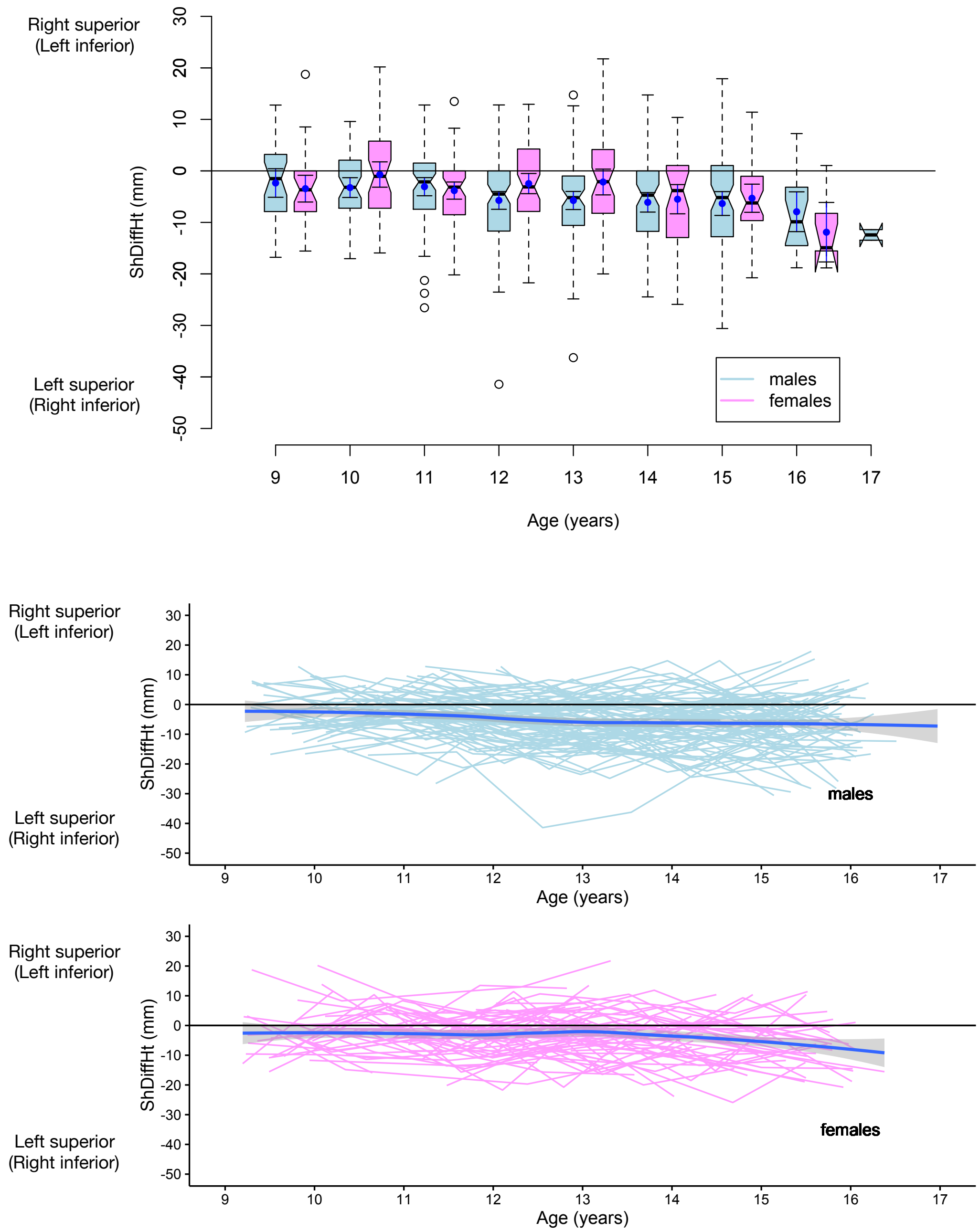


Figure 46: ShDiffHt in (mm) versus age (years) for males and females in the non-scoliotic cohort. ShDiffHt is the difference in vertical height between the shoulder points (see Figures 27 and 28 and Table 4).

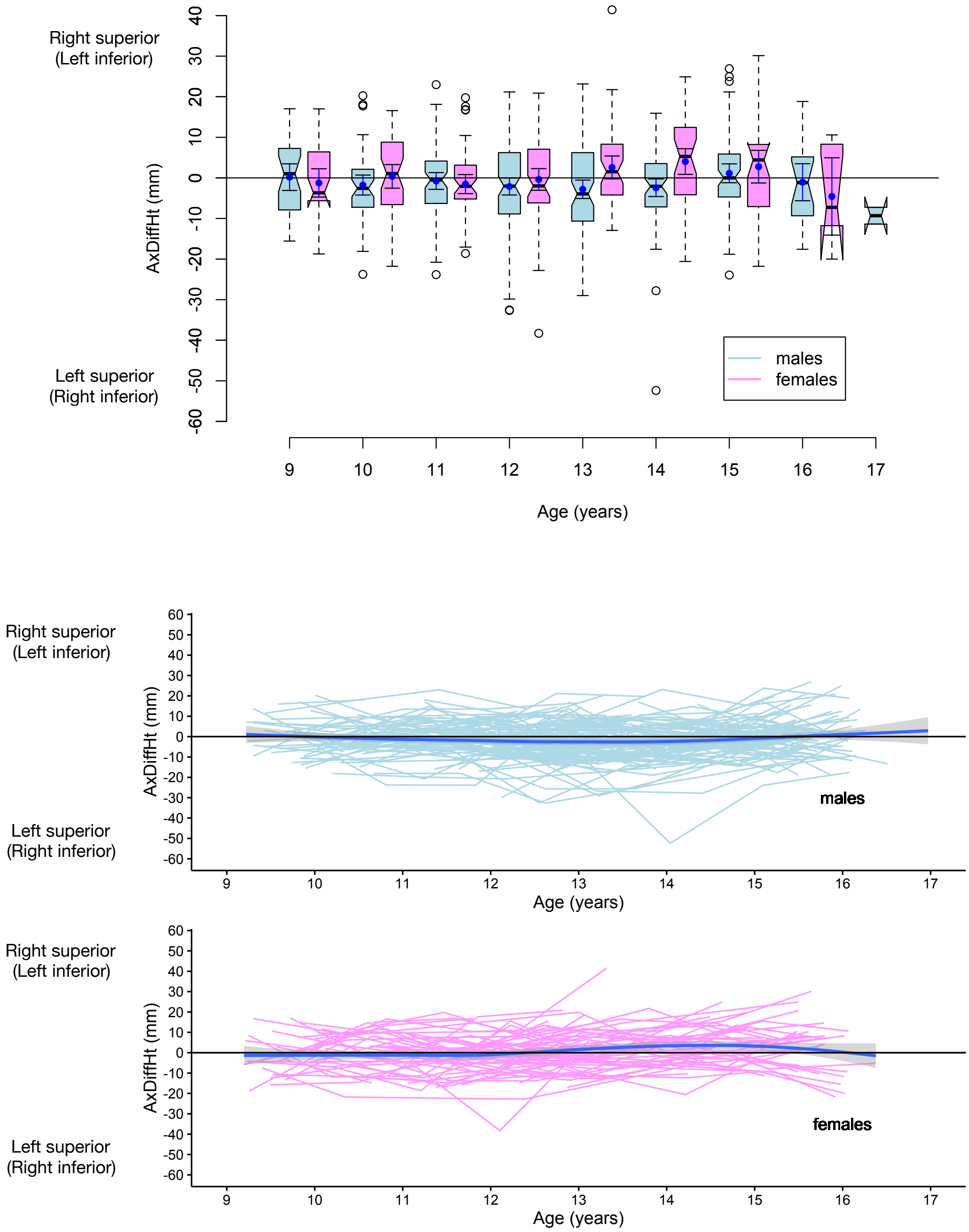


Figure 47: AxDiffHt in (mm) versus age (years) for males and females in the non-scoliotic cohort. AxDiffHt is the difference in vertical height between the axillary points (see Figures 27 and 28 and Table 4).

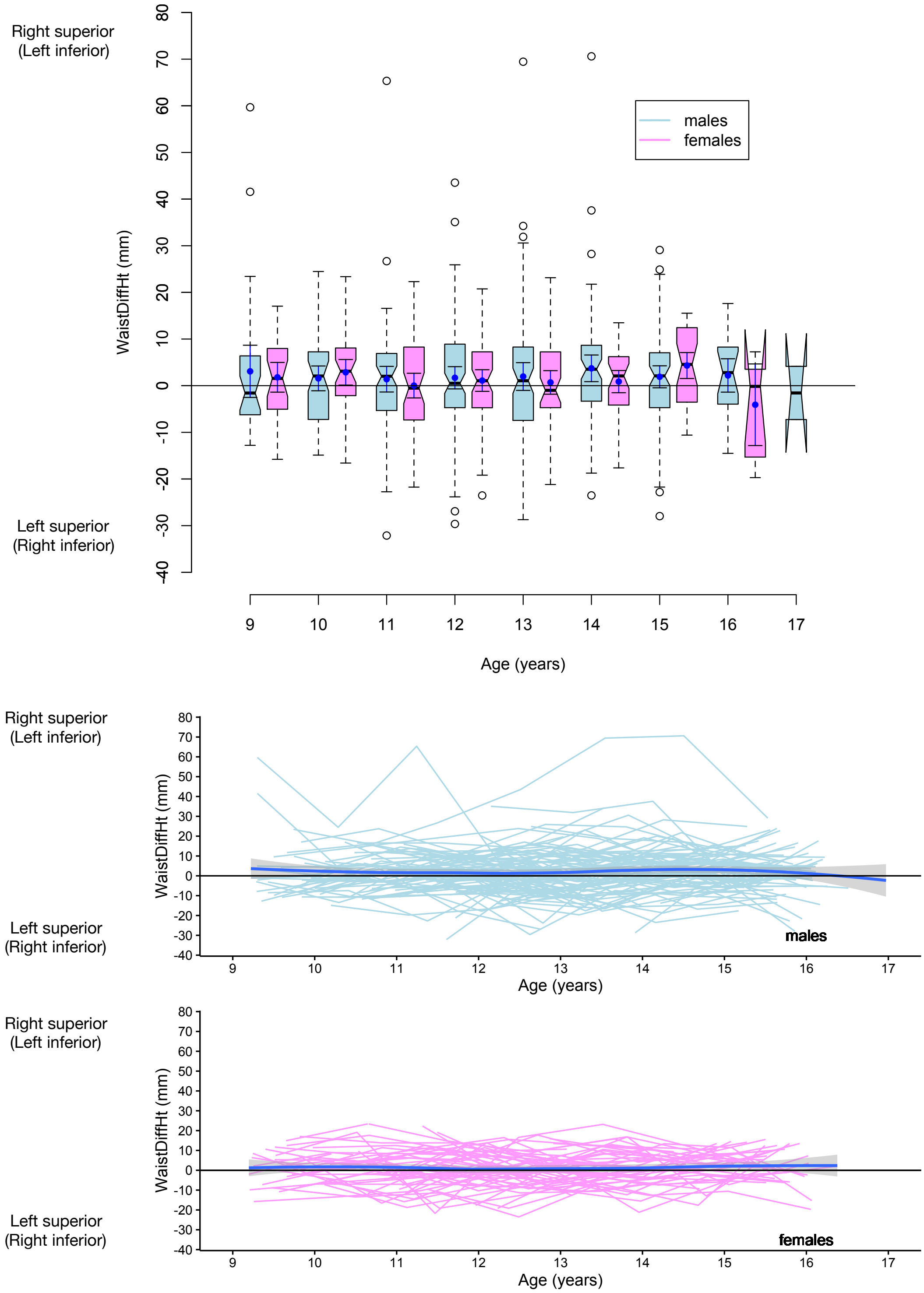


Figure 48: WaistDiffHt (mm) versus age (years) for males and females in the non-scoliotic cohort. WaistDiffHt is the difference in vertical height between the waist points (see Figures 27 and 28 and Table 4).

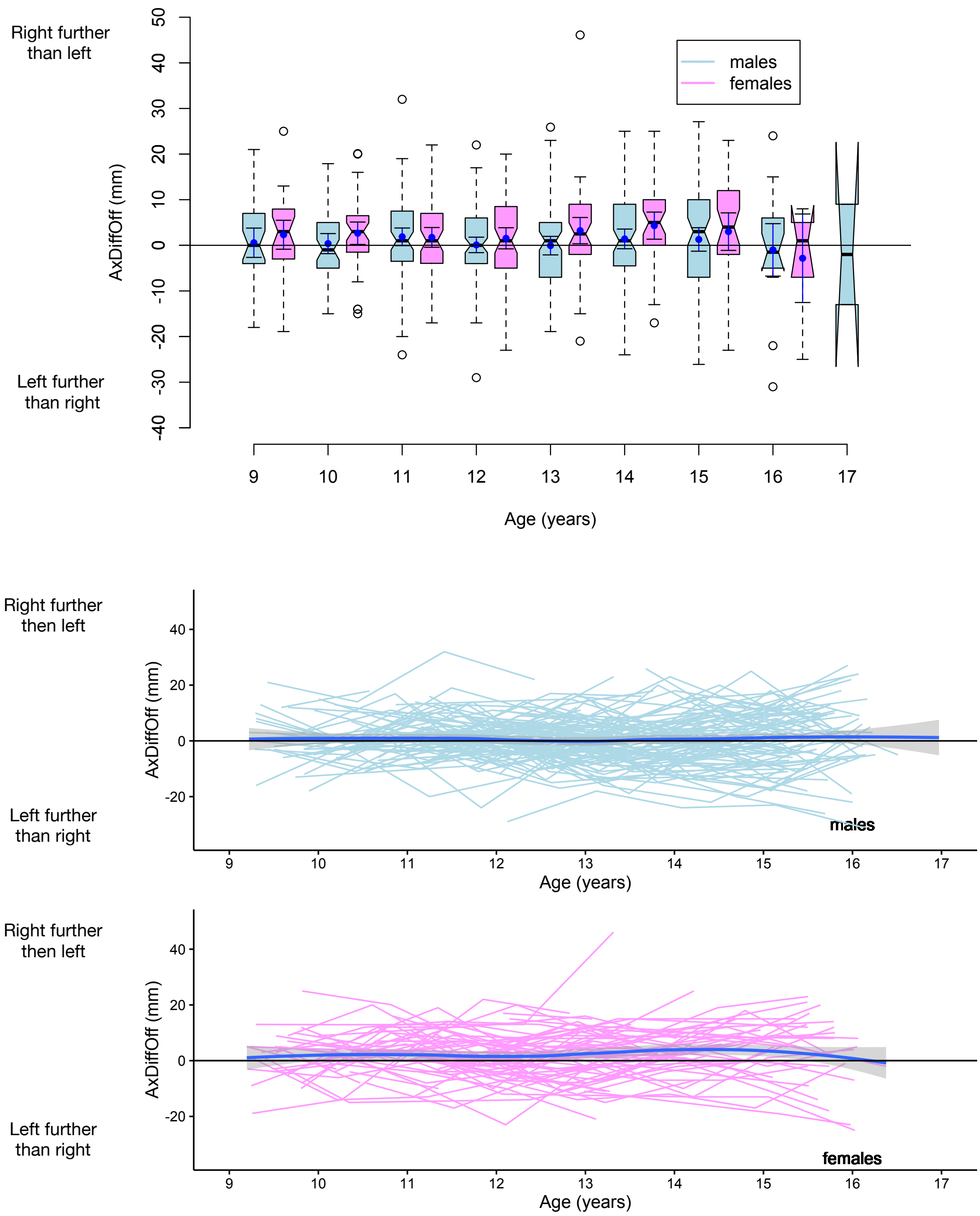


Figure 49: AxDiffOff (mm) versus age (years) for males and females in the non-scoliotic cohort. AxDiffOff is the difference in horizontal distance between the axillary points and the midline (see Figures 27 and 28 and Table 4).

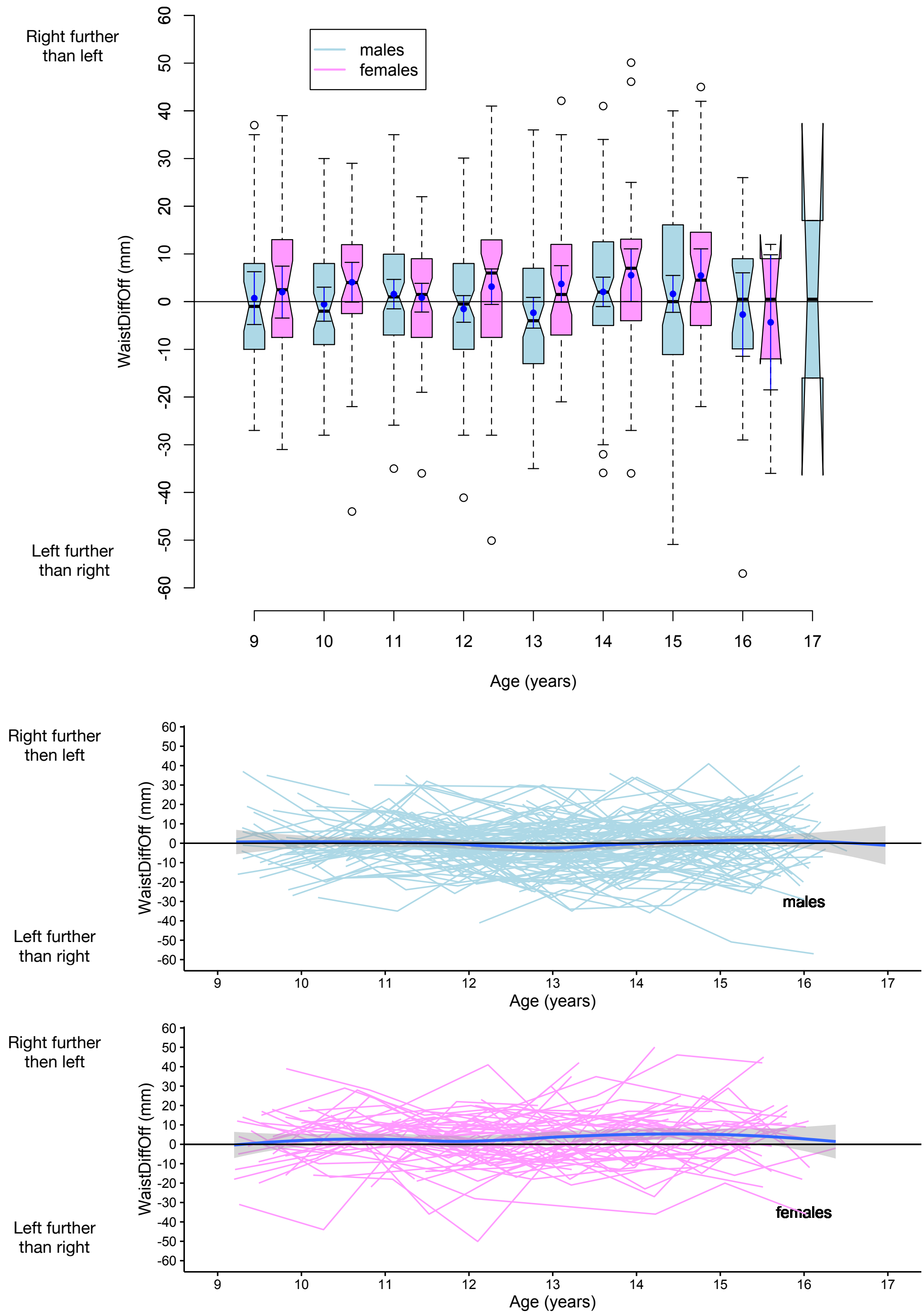


Figure 50: WaistDiffOff (mm) versus age (years) for males and females in the non-scoliotic cohort. WaistDiffOff is the difference in horizontal distance between the waist points and the midline (see Figures 27 and 28 and Table 4).

6.4.3 Conclusions.

In drawing conclusions for the parameters of measures of torso asymmetry, it is noted that all of the results for the mean values (as shown by the LOESS lines in the spaghetti plots) of the torso points are within a maximum of 10 mm from the zero line. This would suggest that, whilst there is asymmetry present between the left and right sides of the body, the mean difference for all parameters is less than 10 mm. This is unlikely to be clinically apparent or significant as shown by the paper by Akel et al [148] where a shoulder height difference of less than 10 mm was not appreciable to the participants of the study. Significant differences with regards to age are seen in ShDiffHt and AxDiffHt. Thus as the cohort ages, there is an increasing asymmetry in the difference in height of the shoulder and axillary points. There is also statistical significance seen in the asymmetry of the height of the axillary points and the distance from the midline of the waist points between males and females. The causes for these findings are not clear but they must be allowed for when reviewing the effects of scoliosis on torso shape. It is also of interest to note that there is a range of several centimetres for the spread of the data at any particular age in all of the parameters. This shows that, whilst the mean value is within 10 mm of symmetry, a population that regards themselves as without asymmetry may well have a measurable asymmetry of several centimetres. Again, this must be allowed for in future analysis in definitions of the extremes of range for normality.

6.5 Measures of spinal shape - coronal shape of the spine.

6.5.1 Methods.

When the data for the shape of the spine in the coronal plane was examined, it became apparent that there were two distinct patterns that emerged and these fitted with the common descriptions of the anatomical locations of scoliotic curves as defined by Lenke et al. [55]. This is a main (and thus larger) curve and a compensatory (and thus smaller) curve. Consequently both the main and compensatory curves are presented in composite box plots and spaghetti plots as used in previous sections. The magnitudes of the curves subdivided by the anatomical location (defined as PT – proximal thoracic, MT – main thoracic, TL – thoracolumbar, L – lumbar, based on the location of the apex of the curve [55]) for the main and compensatory curves are also shown as box-plots for males (light blue) and females (pink) separately.

6.5.2 Results.

Figures 51 to 54 show the anatomical distribution and magnitude of the curves seen for both the main and compensatory curves. Tables 16, 17 and 18 show the numbers in each anatomical group and the statistical significance of age and sex to the main and compensatory curves using mixed effect modeling with the lme4 package [198] with fixed and random effects as previously described.

Table 16: The number (and percentage) in each anatomical subtype of both the main and compensatory coronal curve for males in the non-scoliotic cohort.

Males	Proximal thoracic (PT)	Main thoracic (MT)	Thoraco-lumbar (TL)	Lumbar (L)	No curve
Main coronal curve	22 (4.27)	305 (59.22)	153 (29.71)	26 (5.05)	9 (1.75)
Compensatory coronal curve	64 (12.43)	85 (16.50)	46 (8.93)	136 (26.41)	184 (35.73)

Table 17: The number (and percentage) in each anatomical subtype of both the main and compensatory coronal curve for females in the non-scoliotic cohort.

Females	Proximal thoracic (PT)	Main thoracic (MT)	Thoraco-lumbar (TL)	Lumbar (L)	No curve
Main coronal curve	8 (2.53)	174 (55.06)	103 (41.14)	25 (7.91)	6 (1.90)
Compensatory coronal curve	58 (18.35)	60 (18.99)	39 (12.34)	79 (25.00)	80 (25.32)

Table 18: The significance and coefficients for the parameters of age and sex for main and compensatory coronal curves in the non-scoliotic cohort. For an explanation of the terms in the table see section 6.3.1.

Parameter		Chi squared	p value	coefficient	standard error
Main coronal curve	Age	8.99	0.003	0.36 °/yr	0.12
	Sex	1.84	0.175	- 0.82 °/yr	0.61
Compensatory coronal curve	Age	5.32	0.021	- 0.17 °/yr	0.07
	Sex	5.49	0.019	0.72 °/yr	0.31

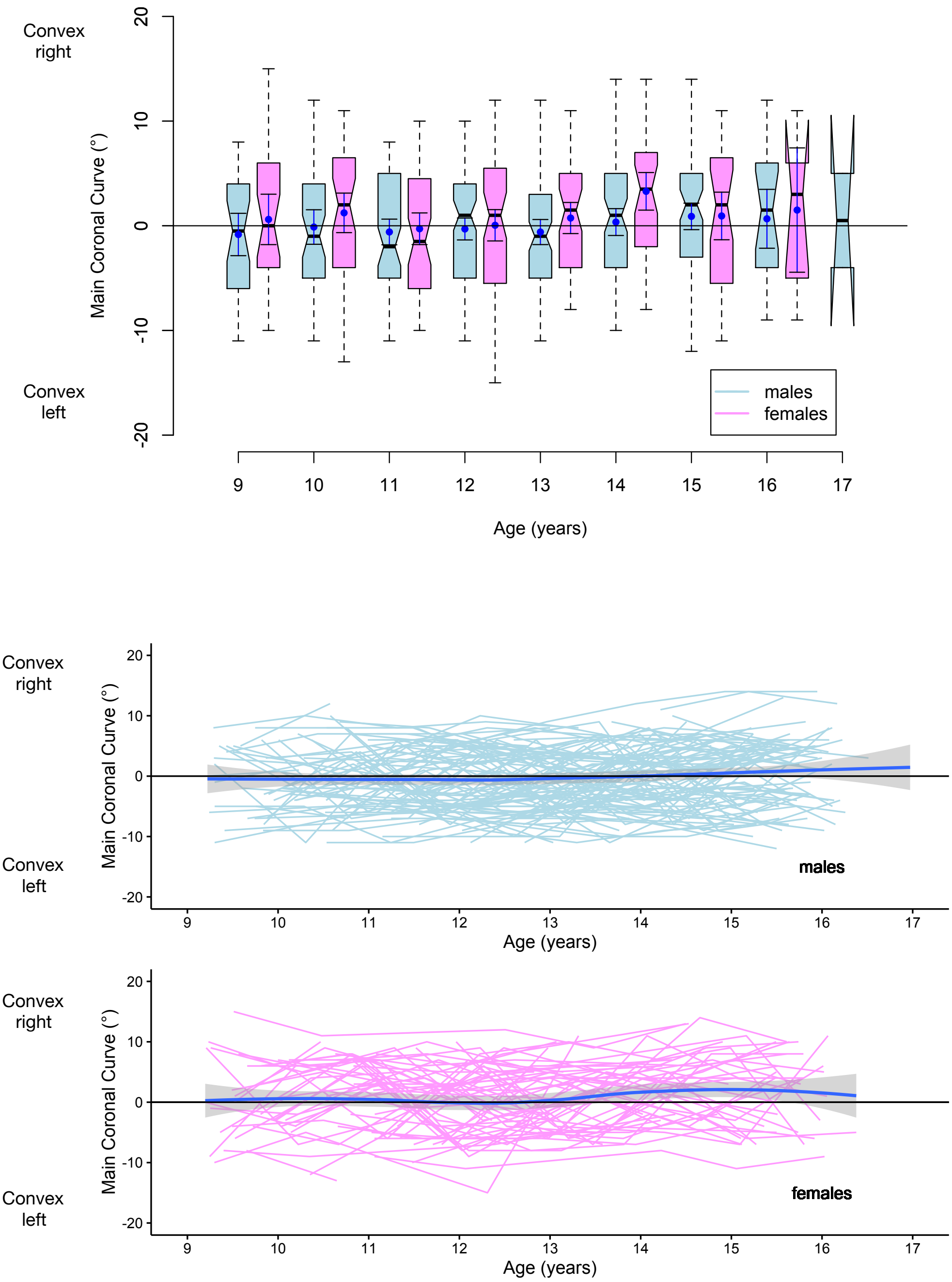


Figure 51: The main coronal curve (°) versus age (years) for males and females in the non-scoliotic cohort.

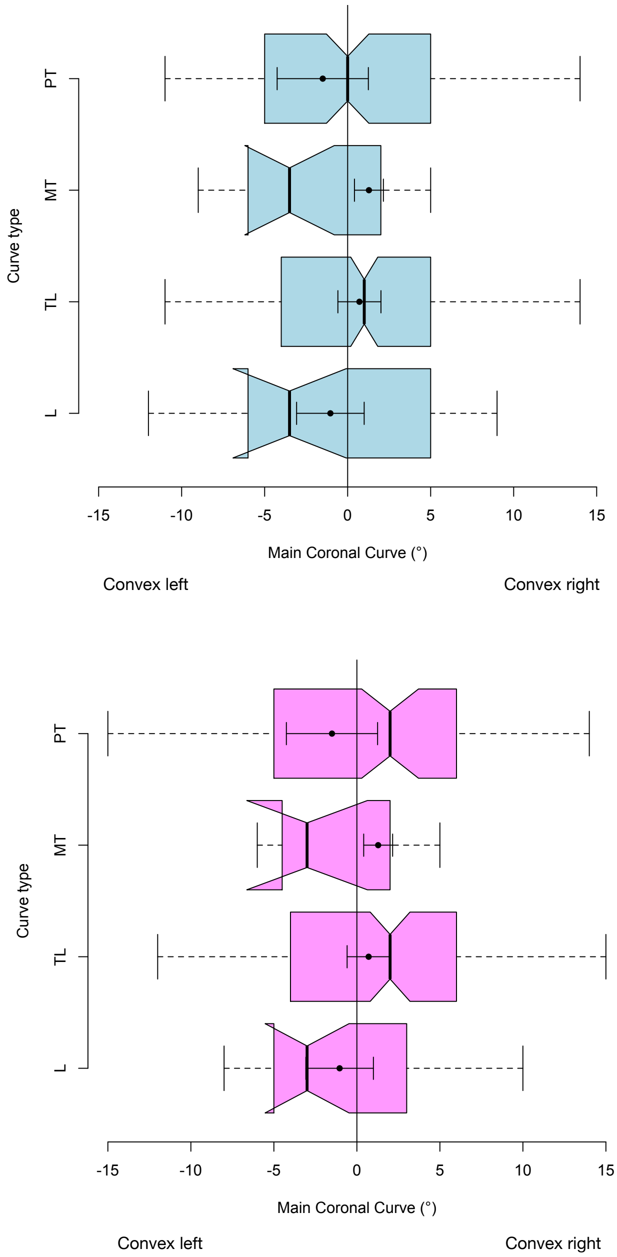


Figure 52: The anatomical distribution and size of the main coronal curve (°) for males and females in the non-scoliotic cohort.

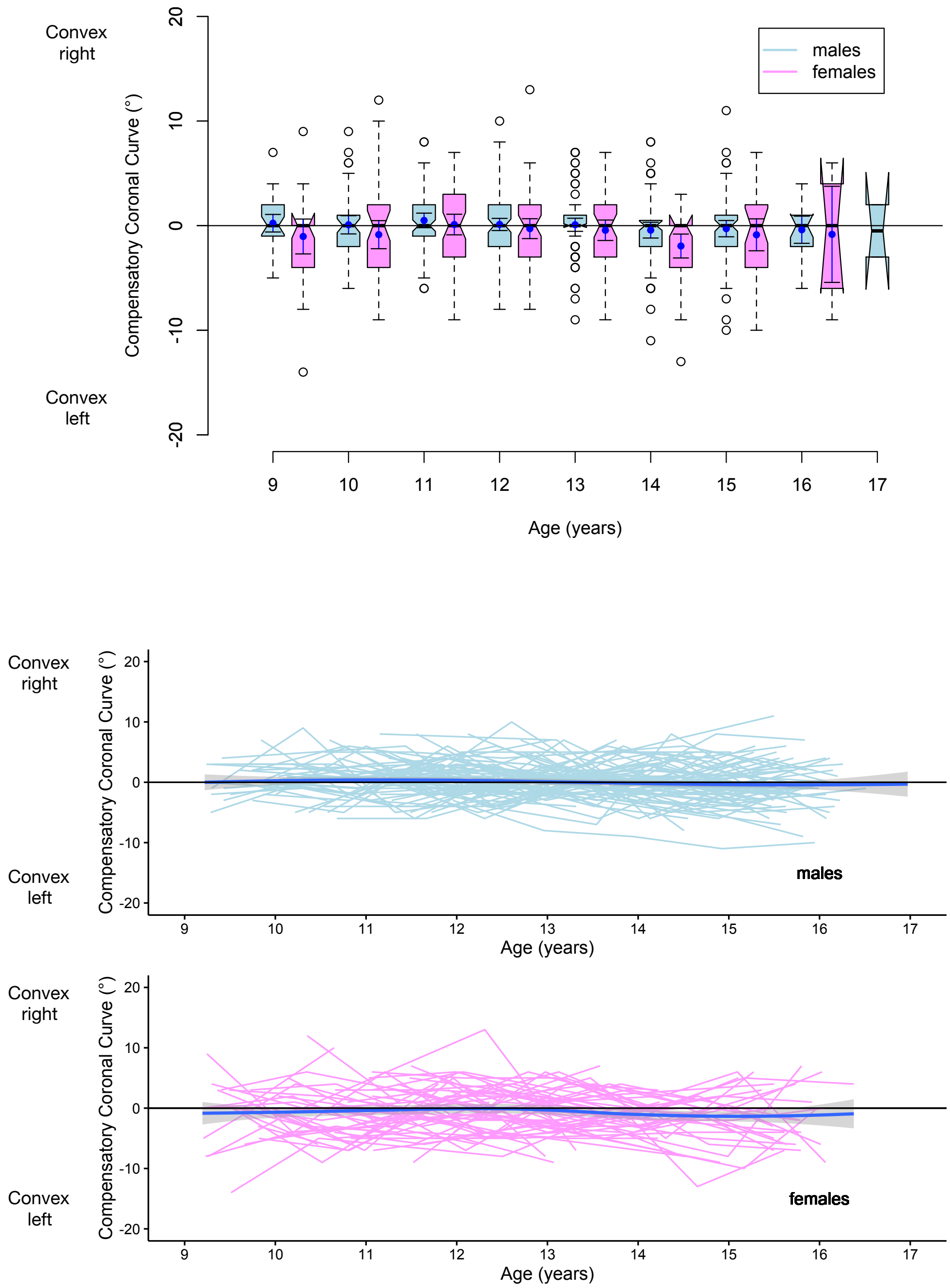


Figure 53: The compensatory coronal curve (°) versus age (years) for males and females in the non-scoliotic cohort.

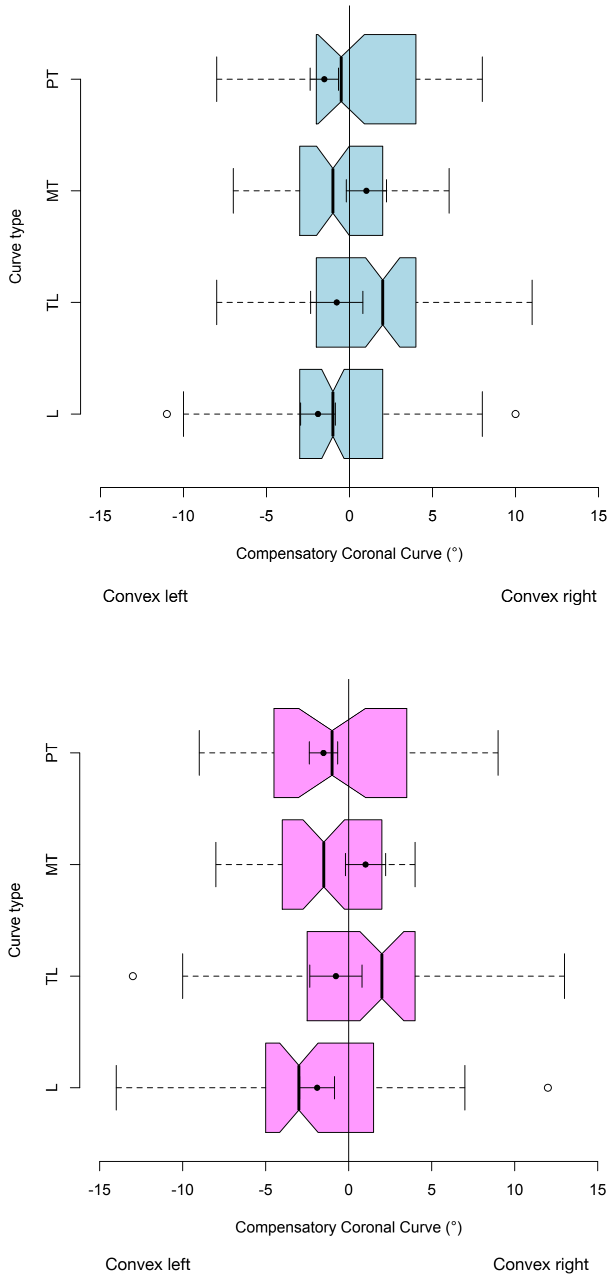


Figure 54: The anatomical distribution and size of the compensatory coronal curve (°) for males and females in the non-scoliotic cohort.

6.5.3 Conclusions.

The results show that there is a degree of curve in 98% of the cohort. The anatomical location of that curve varies. There are also main and compensatory curve patterns. Of interest is the direction of the convexity of the curve. Different to that of an AIS cohort [3], in the main thoracic curve the convexity is more frequently seen to the left, and in the thoracolumbar curve to the right although noting that the variability of the data indicates a mix of convexities to both sides for both of the main curve subtypes. The main and compensatory curves change with age. The compensatory curve is different for males and females although the main curve is not. The median value for all of the anatomical subtypes of curve, both main and compensatory, for both males and females are less than 10° so would not class as scoliosis [8] although there are some children in this cohort, that by this measure, have a small scoliosis. Thus a normal cohort of children will have a degree of coronal bend to their spine.

6.6 Measures of spinal shape – relationship between the coronal shape of the spine and the measures of torso asymmetry.

6.6.1 Methods.

The interactions between the coronal curves and the torso points are represented as data ellipses to show the bivariate nature of the data [202, 203]. The data are presented with curve size in degrees on the x-axis (convex to the right as a positive number and convex to the left as a negative number) and the torso point parameter on the y-axis (right side more superior than the left or further from the midline than the left as a positive number and right more inferior than the left or left further from the midline than right as a negative number). The solid red dot represents the mean value of the two parameters with the surrounding red ellipse as the 95% confidence interval of that mean. The box plots on the outside of the x-axis and y-axis give the distributions of the individual parameters (as in Figure 30). As previously, the data was divided according to the Lenke classification of AIS [55] as a main thoracic with compensatory thoracolumbar curve type and a main thoracolumbar with compensatory thoracic curve type. The data for males and females is combined for this analysis.

6.6.2 Results.

Figures 55 to 59 and 60 to 64 demonstrate the two predominate curve patterns, a main thoracic with compensatory thoracolumbar curve type and a main thoracolumbar curve with compensatory thoracic curve type. The torso parameters are plotted against the spinal curve located closest anatomically, i.e. ShDiffHt, AxDiffHt and AxDiffOff against the thoracic curve (main or compensatory) and WaistDiffHt and WaistDiffOff against the thoracolumbar curve (main or compensatory).

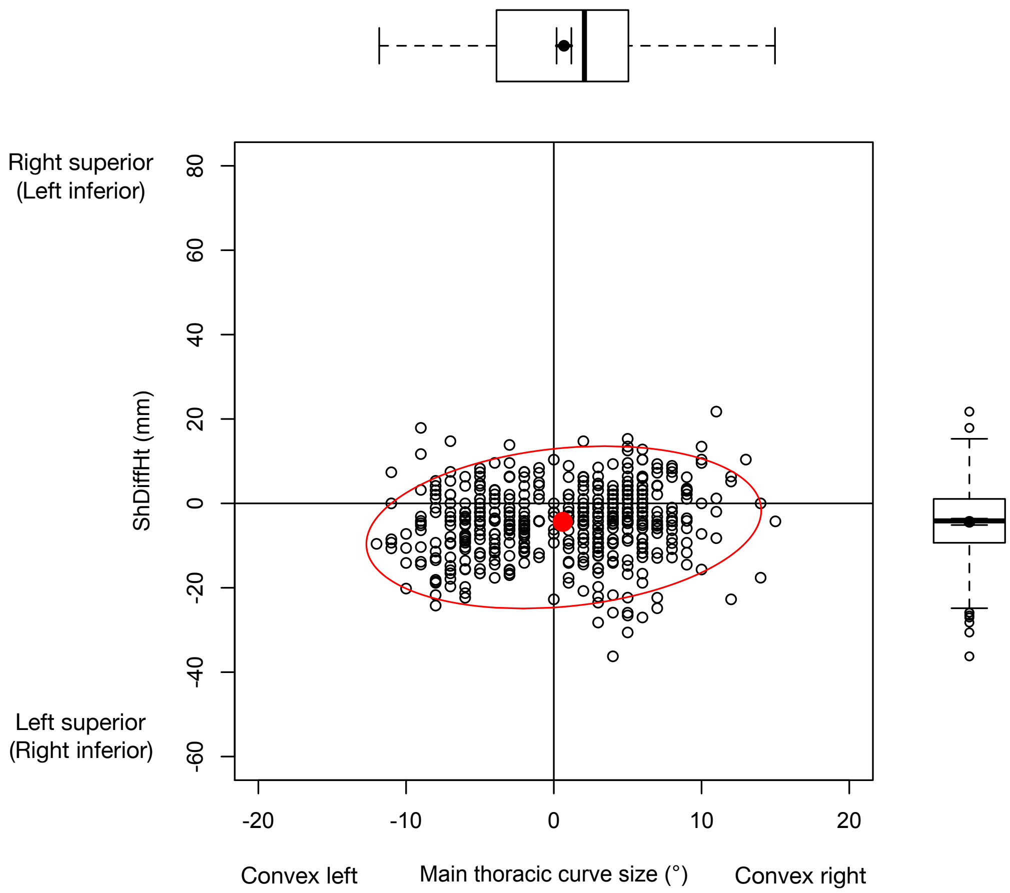


Figure 55: ShDiffHt (mm) versus curve size (°) for the main thoracic curve in the main thoracic curve group showing the mean value (red circle) and 95% confidence ellipse (red ellipse) in the non-scoliotic cohort. ShDiffHt is the difference in vertical height between the shoulder points (see Figures 27 and 28 and Table 4).

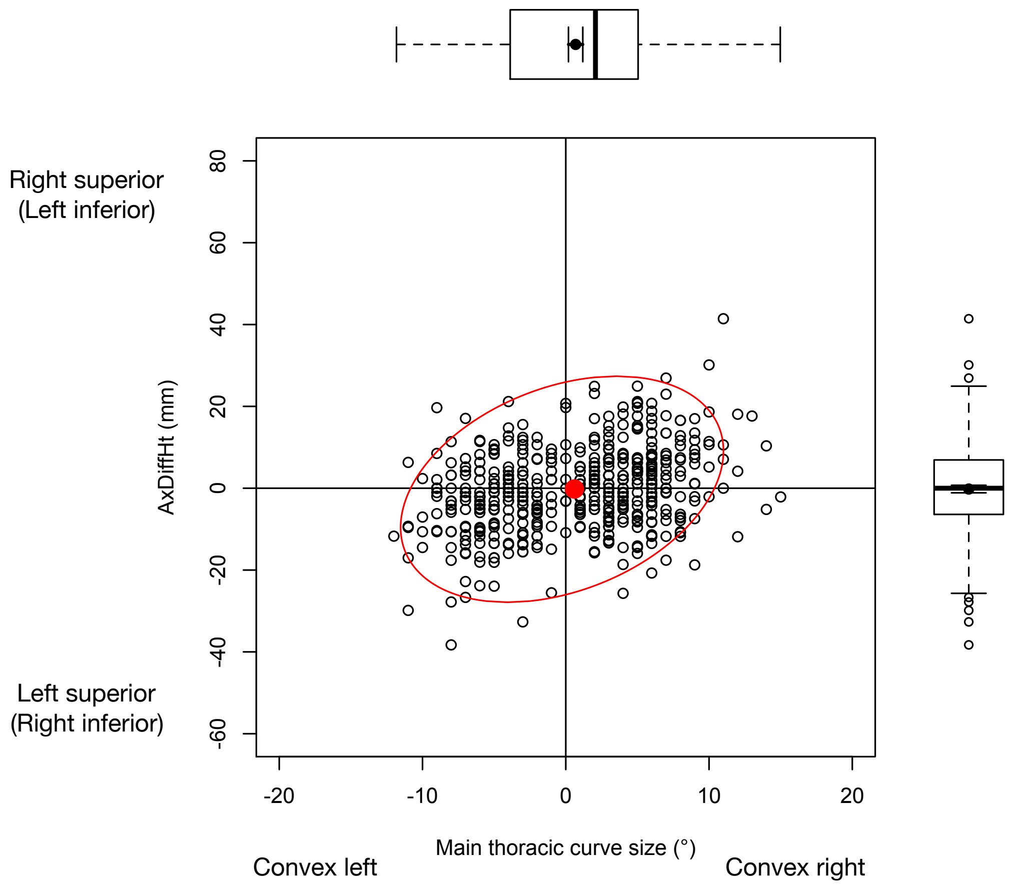


Figure 56: AxDiffHt (mm) versus curve size (°) for the main thoracic curve in the main thoracic curve group showing the mean value (red circle) and 95% confidence ellipse (red ellipse) in the non-scoliotic cohort. AxDiffHt is the difference in vertical height between the axillary points (see Figures 27 and 28 and Table 4).

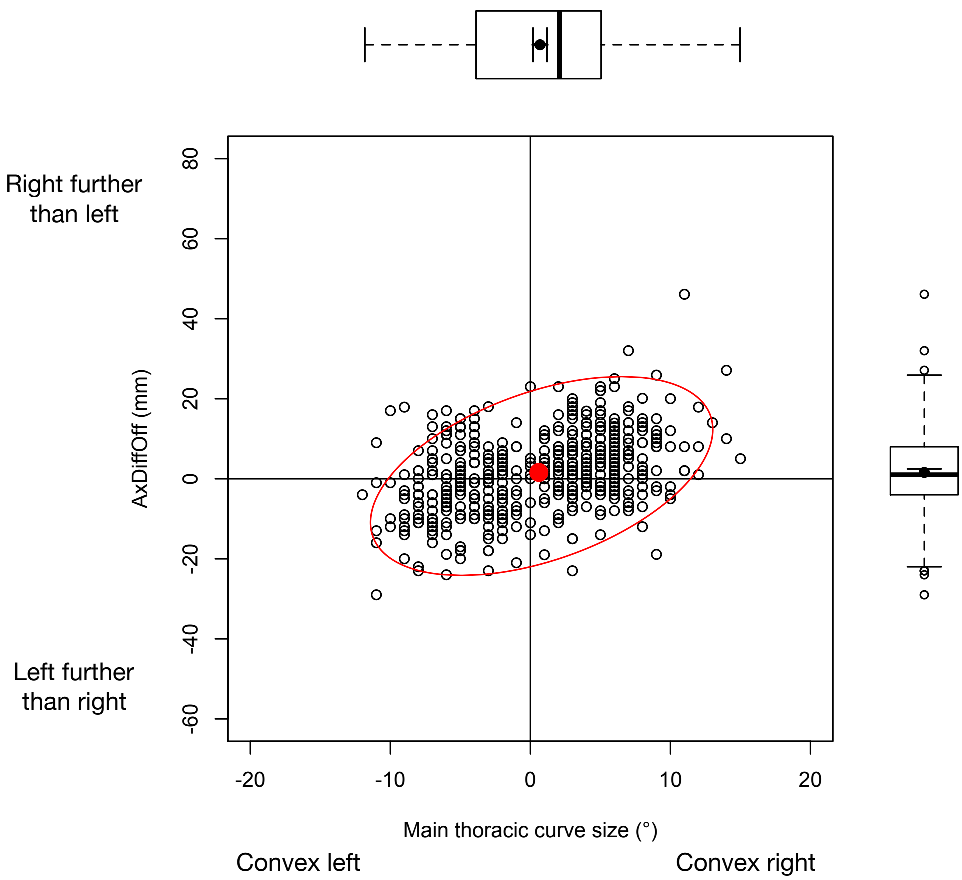


Figure 57: AxDiffOff (mm) versus curve size (°) for the main thoracic curve in the main thoracic curve group showing the mean value (red circle) and 95% confidence ellipse (red ellipse) in the non-scoliotic cohort. AxDiffOff is the difference in horizontal distance between the axillary points and the midline (see Figures 27 and 28 and Table 4).

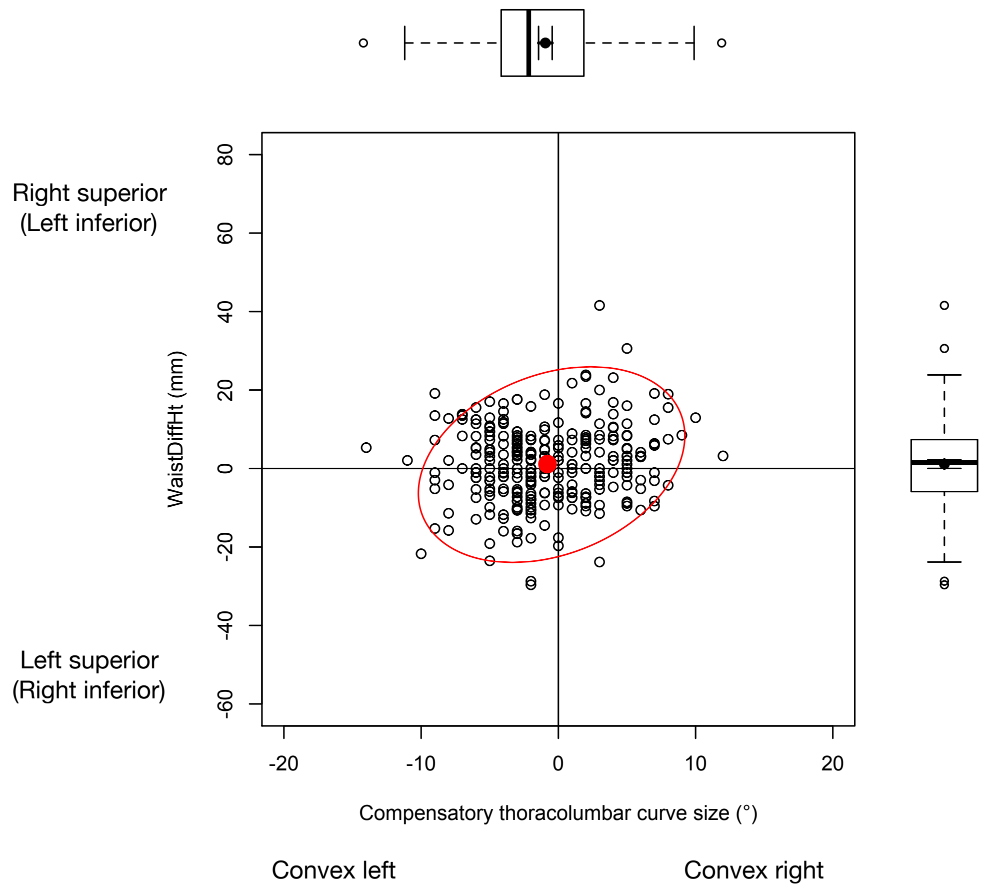


Figure 58: WaistDiffHt (mm) versus curve size (°) for the compensatory thoracolumbar curve in the main thoracic group showing the mean value (red circle) and 95% confidence ellipse (red ellipse) in the non-scoliotic cohort. WaistDiffHt is the difference in vertical height between the waist points (see Figures 27 and 28 and Table 4).

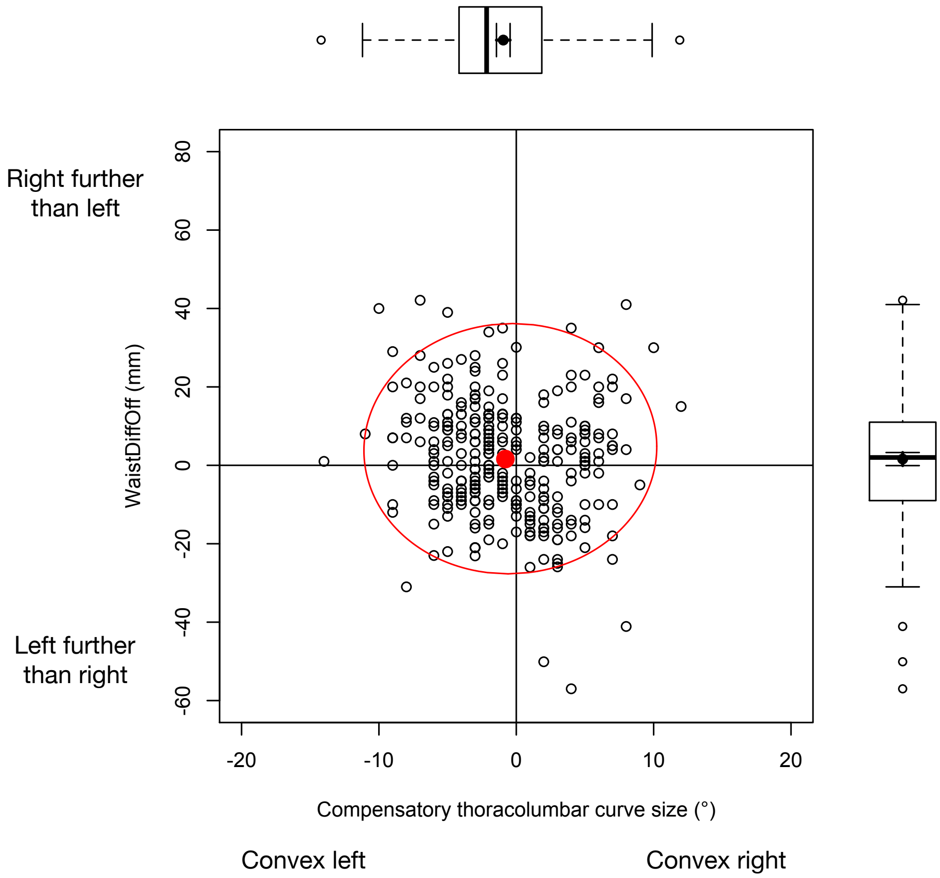


Figure 59: WaistDiffOff (mm) versus curve size (°) for the compensatory thoracolumbar curve in the main thoracic group showing the mean value (red circle) and 95% confidence ellipse (red ellipse) in the non-scoliotic cohort. WaistDiffOff is the difference in horizontal distance between the waist points and the midline (see Figures 27 and 28 and Table 4).

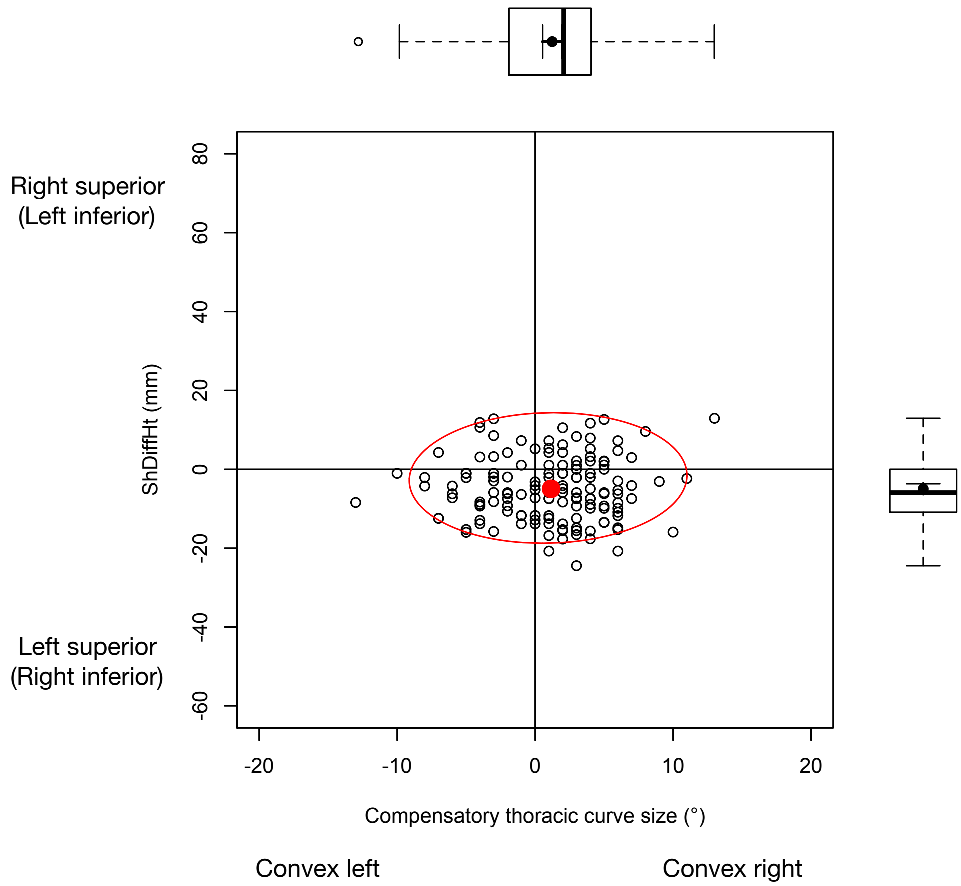


Figure 60: ShDiffHt (mm) versus curve (°) for the compensatory thoracic curve in the main thoracolumbar group showing the mean value (red circle) and 95% confidence ellipse (red ellipse) in the non-scoliotic cohort. ShDiffHt is the difference in vertical height between the shoulder points (see Figures 27 and 28 and Table 4).

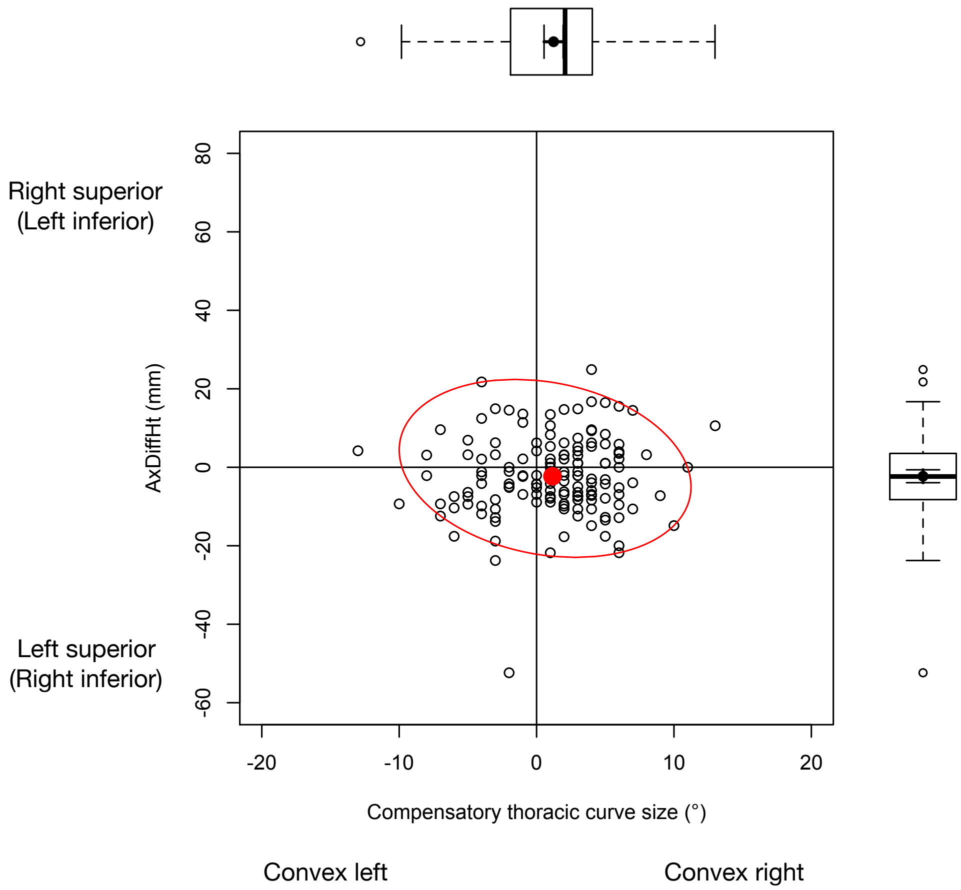


Figure 61: AxDiffHt (mm) versus curve size (°) for the compensatory thoracic curve in the main thoracolumbar group showing the mean value (red circle) and 95% confidence ellipse (red ellipse) in the non-scoliotic cohort. AxDiffHt is the difference in vertical height between the axillary points (see Figures 27 and 28 and Table 4).

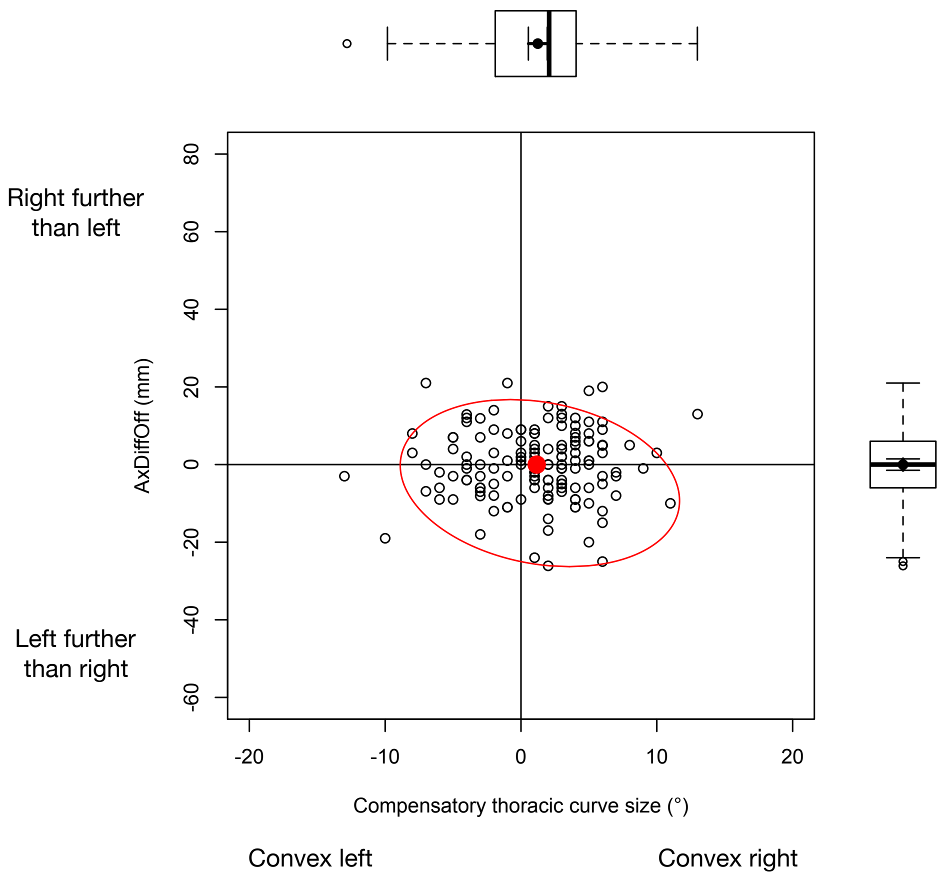


Figure 62: AxDiffOff (mm) versus curve size (°) for the compensatory thoracic curve in the main thoracolumbar group showing the mean value (red circle) and 95% confidence ellipse (red ellipse) in the non-scoliotic cohort. AxDiffOff is the difference in horizontal distance between the axillary points and the midline (see Figures 27 and 28 and Table 4).

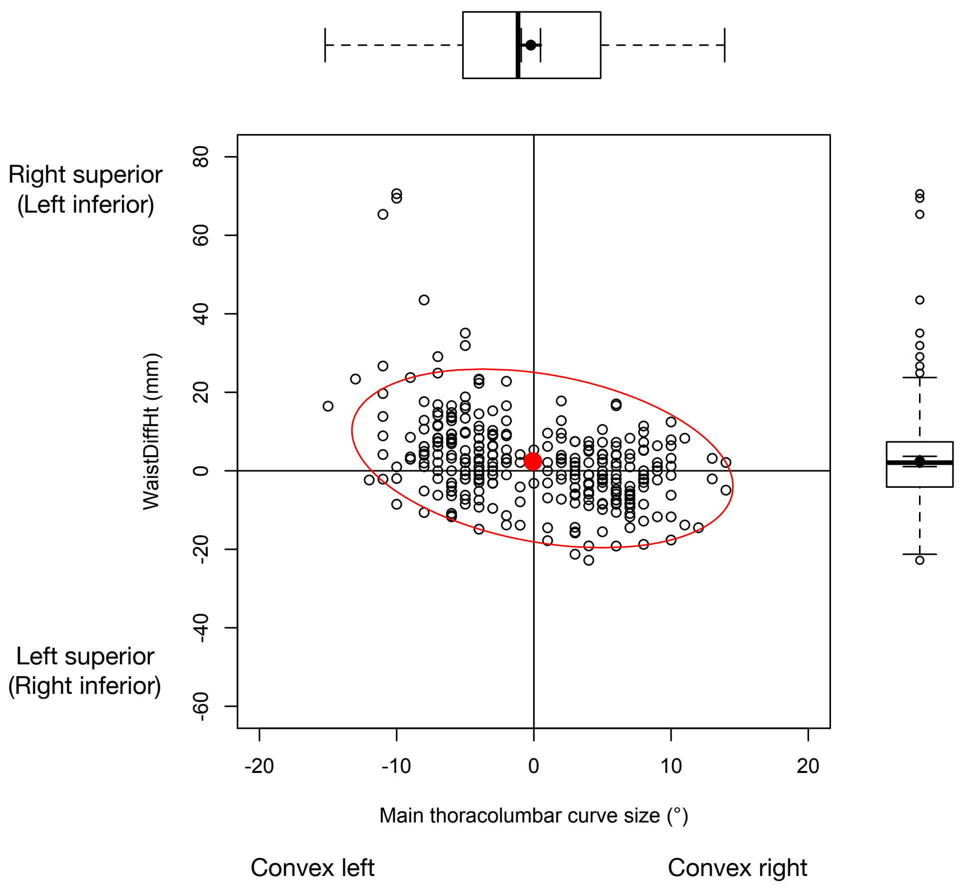


Figure 63: WaistDiffHt (mm) versus curve size (°) for the main thoracolumbar curve in the main thoracolumbar curve group showing the mean value (red circle) and 95% confidence ellipse (red ellipse) in the non-scoliotic cohort. WaistDiffHt is the difference in vertical height between the waist points (see Figures 27 and 28 and Table 4).

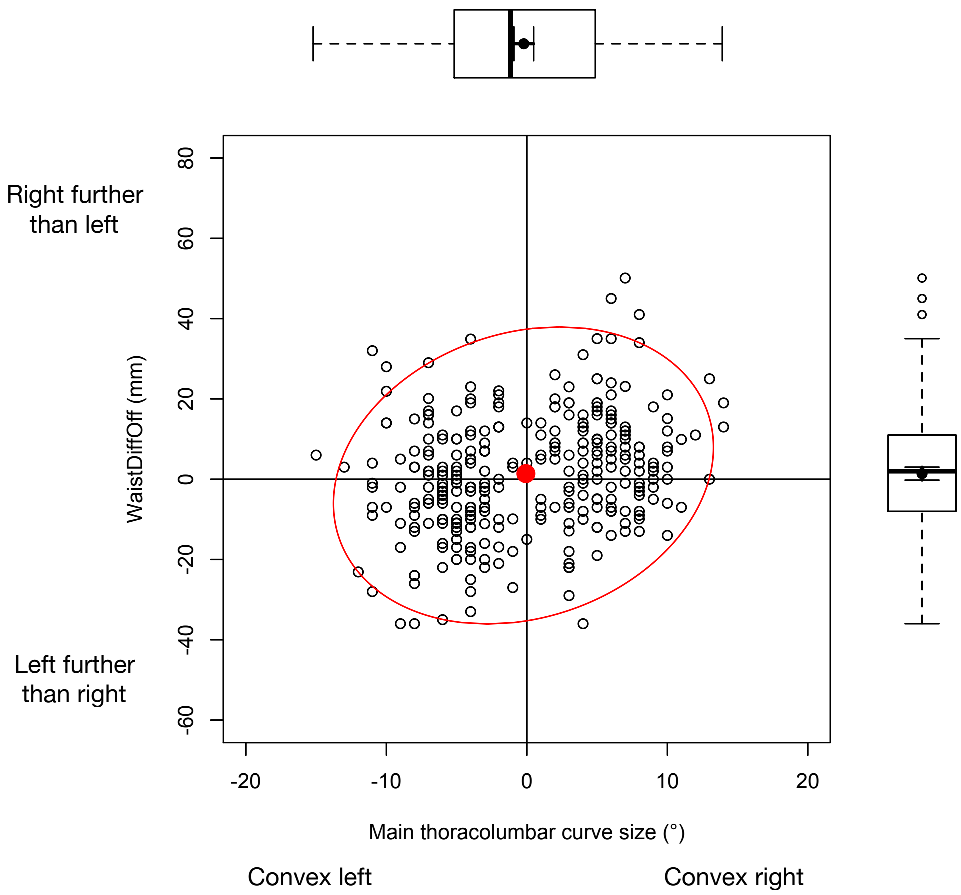


Figure 64: WaistDiffOff (mm) versus curve size (°) for the main thoracolumbar curve in the main thoracolumbar curve group showing the mean value (red circle) and 95% confidence ellipse (red ellipse) in the non-scoliotic cohort. WaistDiffOff is the difference in horizontal distance between the waist points and the midline (see Figures 27 and 28 and Table 4).

6.6.3 Conclusions.

The data ellipses show that with the two patterns of curve types there is a change in the asymmetry seen in the torso. This would seem reasonable as those parameters associated with the upper torso and shoulder girdle (ShDiffHt, AxDiffHt and AxDiffOff) would be affected by the shape of the thoracic spine and those parameters associated with the lower torso (WaistDiffHt and Waist DiffOff) would be affected by the shape of the thoracolumbar spine. The compensatory curves have a lesser effect on the surrounding torso as the curves are smaller. For some parameters, the effect of this is greater with increasing curve size and thus is seen more in the main rather than the compensatory curves. Particularly, the parameters of AxDiffOff and AxDiffHt increase as the thoracic curve increases. Interestingly this is not seen in ShDiffHt to the same degree and this may represent the anatomical relationship of the shoulder girdle as a mobile structure around the underlying torso. There is a greater variability in the data for WaistDiffOff than seen for other parameters for reasons that are not clear.

6.7 Measures of spinal shape – sagittal parameters of the spine (kyphosis and lordosis).

6.7.1 Methods.

The same methodology of composite box plots and spaghetti plots are used here for the plotting of kyphosis and lordosis. Further plots of the LOESS lines and confidence interval funnels only for both males and females are also plotted. Again, statistical significance is assessed using linear mixed effect modeling with the previously noted fixed effects of age and sex and the random effects of repeat measures.

6.7.2 Results.

Figure 65 relates to kyphosis and Figure 66 relates to lordosis. Figure 67 shows only the LOESS lines and 95% confidence interval funnels for kyphosis and lordosis. Table 19 shows the levels of statistical significance for the effects of age and sex on kyphosis and lordosis.

Table 19: The significance and coefficients for the parameters of age and sex for kyphosis and lordosis in the non-scoliotic cohort. For an explanation of the terms in the table see section 6.3.1.

Parameter		Chi squared	p value	coefficient	standard error
Kyphosis	Age	8.33	0.004	0.39 °/yr	0.14
	Sex	3.03	0.082	1.79 °	1.02
Lordosis	Age	23.68	< 0.001	0.84 °/yr	0.17
	Sex	6.76	0.009	- 3.02 °	1.16

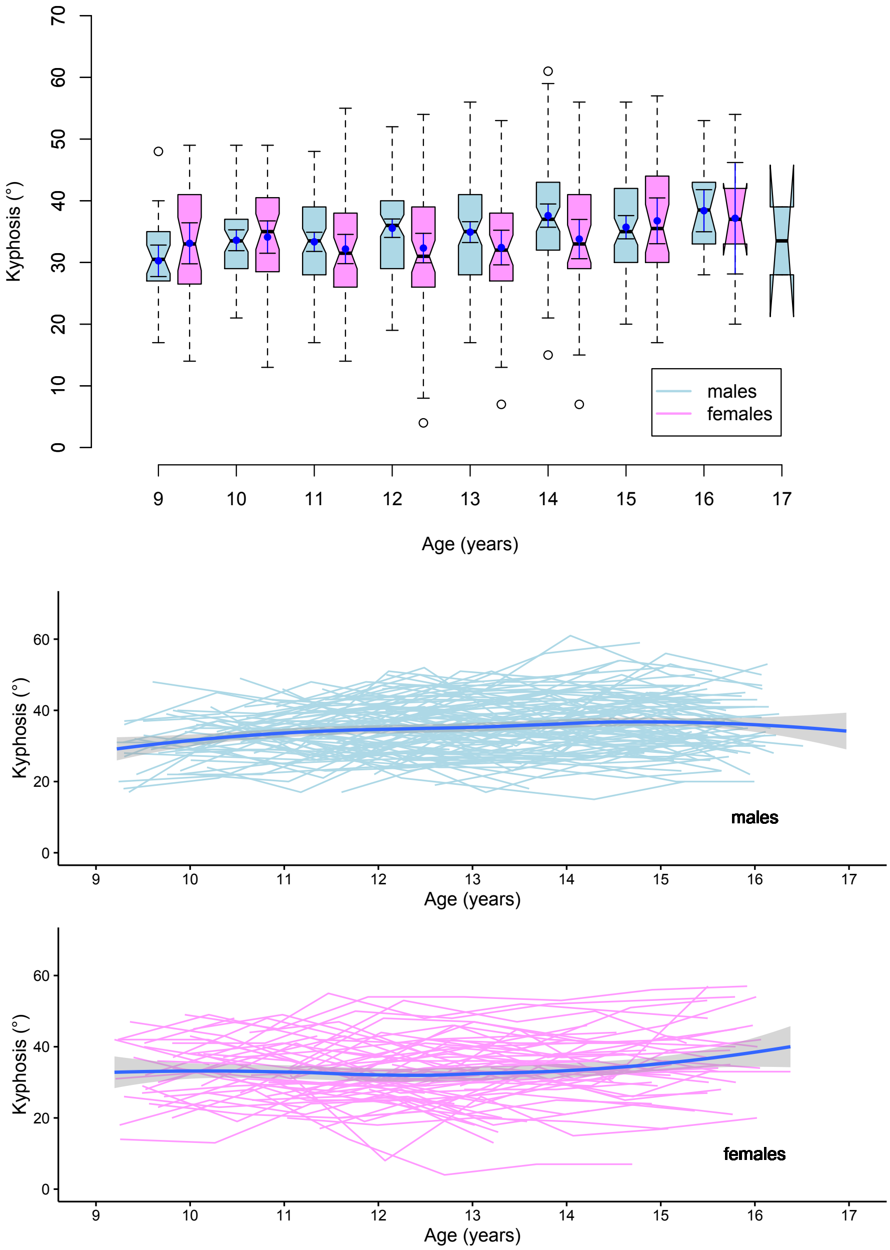


Figure 65: Kyphosis (°) versus age (years) for males and females in the non-scoliotic cohort.

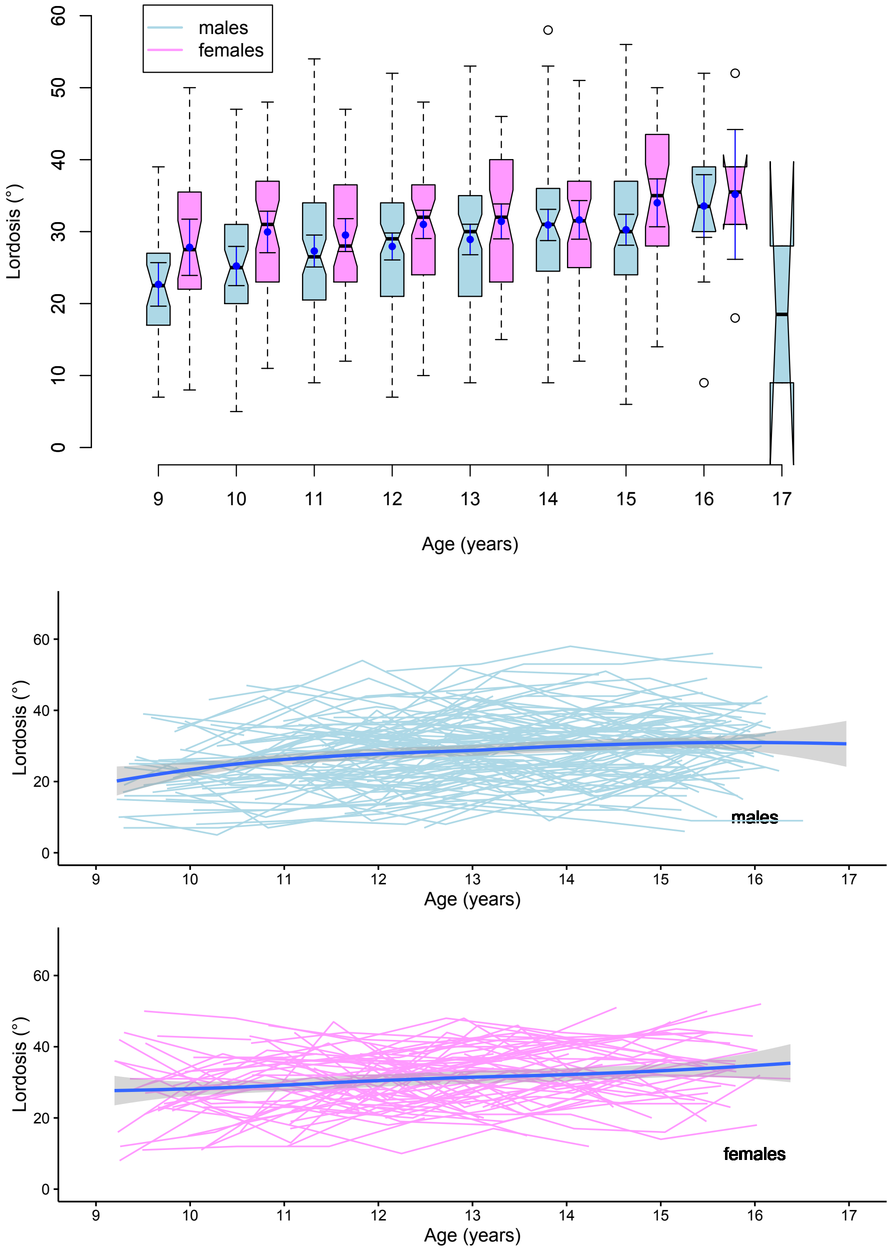


Figure 66: Lordosis (°) versus age (years) for males and females in the non-scoliotic cohort.

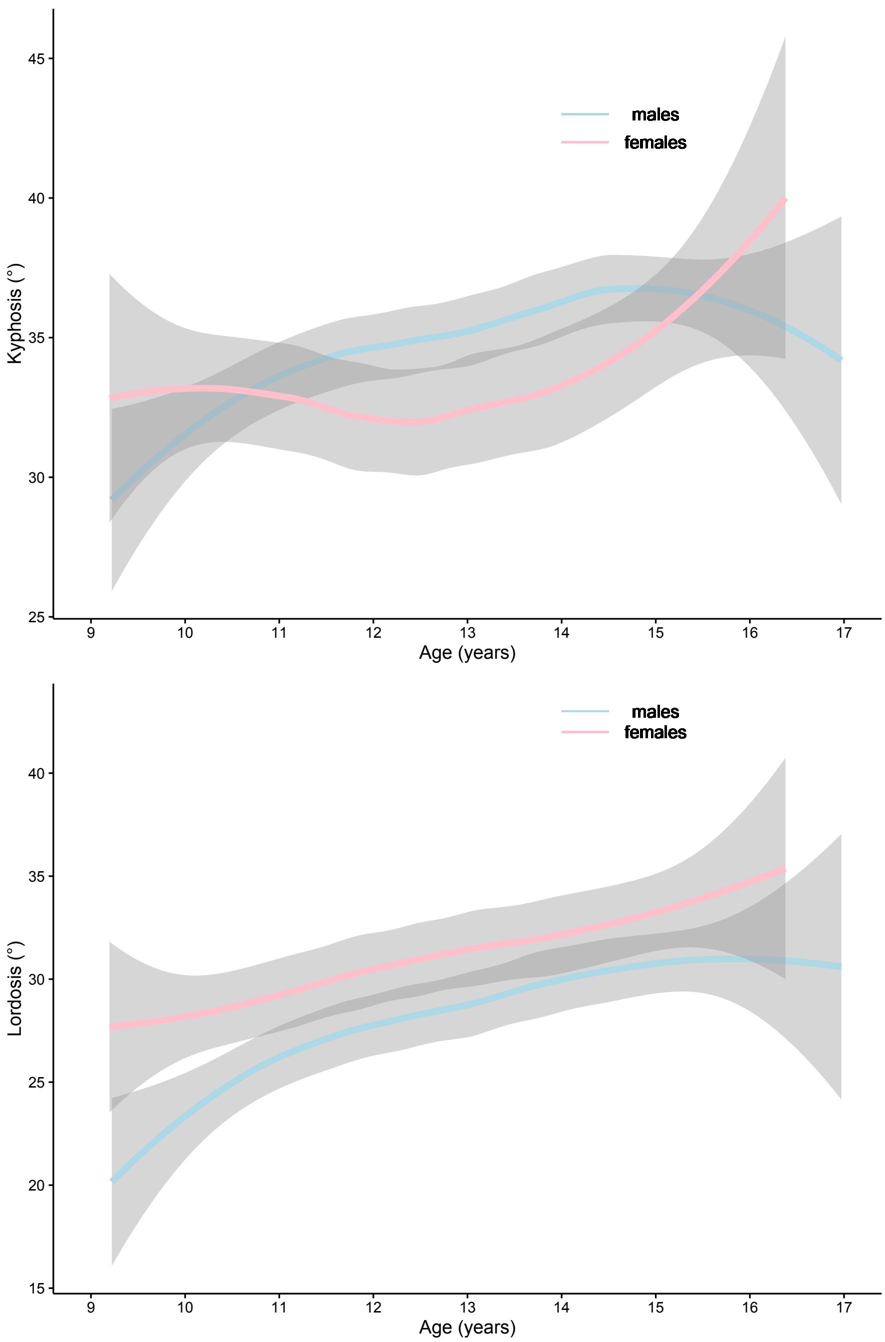


Figure 67: The mean LOESS line and 95% confidence intervals of kyphosis (°) and lordosis (°) versus age (years) for males and females in the non-scoliotic cohort.

6.7.3 Conclusions.

These results show that both kyphosis and lordosis increase over the ages measured for both males and females, in a linear fashion for lordosis but not for kyphosis. For males, kyphosis increased from age 9, decreasing again towards the older ages. Whereas for females, kyphosis fell below the starting mean value over the mid portion of the ages measured (11 to 14 years) before increasing again to be greater than the males at the older ages. However this was not significant. Lordosis is significantly larger in females than males across all ages. Again, there is a range of values within the group around the mean value.

6.8 Measures of 3D shape.

6.8.1 Methods.

The parameters that measure 3D shape are subdivided into those that document the position of the most prominent point over the left and right sides of the back, and volumetric asymmetry (VA). The most prominent points are documented in Figure 29 and Table 5 in the Methods chapter. VA is defined as per Figure 26 in the Methods chapter. Composite box plots and spaghetti plots are used for the individual parameters of the most prominent points and were then analysed for any relationship with age and sex using linear mixed effect modeling.

6.8.2 Results – most prominent points.

Table 20 shows the statistical significance of the effects of age and sex for the parameters MaxDiffOff, ScapDiffHt and ScapDiffDepth. Figures 68, 69 and 70 show composite box-plots and spaghetti plots for these parameters.

Table 20: The significance and coefficients for the parameters of age and sex of the most prominent points on the left and right sides of the back in the non-scoliotic cohort. For an explanation of the terms in the table see section 6.3.1. For the definitions of the parameters see Figure Figure 29 and Table 5.

Parameter		Chi squared	p value	coefficient	standard error
MaxDiffOff *	Age	8.86	0.003	0.67 mm/yr	0.22
	Sex	1.99	0.159	- 0.17 mm	1.18
ScapDiffHt **	Age	3.12	0.077	0.32 mm/yr	0.18
	Sex	0.001	0.973	- 0.04 mm	1.25
ScapDiffDepth	Age	10.46	0.001	0.31 mm/yr	0.09
	Sex	1.45	0.228	0.78 mm	0.65

* Undue effects from multiple outliers. The data was simplified and outliers greater than ± 2 standard deviations from the mean were excluded. This resulted in a model with an acceptable residual and qq plot.

** One outlier removed that was having undue effects. Removal normalised the data and revealed an acceptable residual and qq plot.

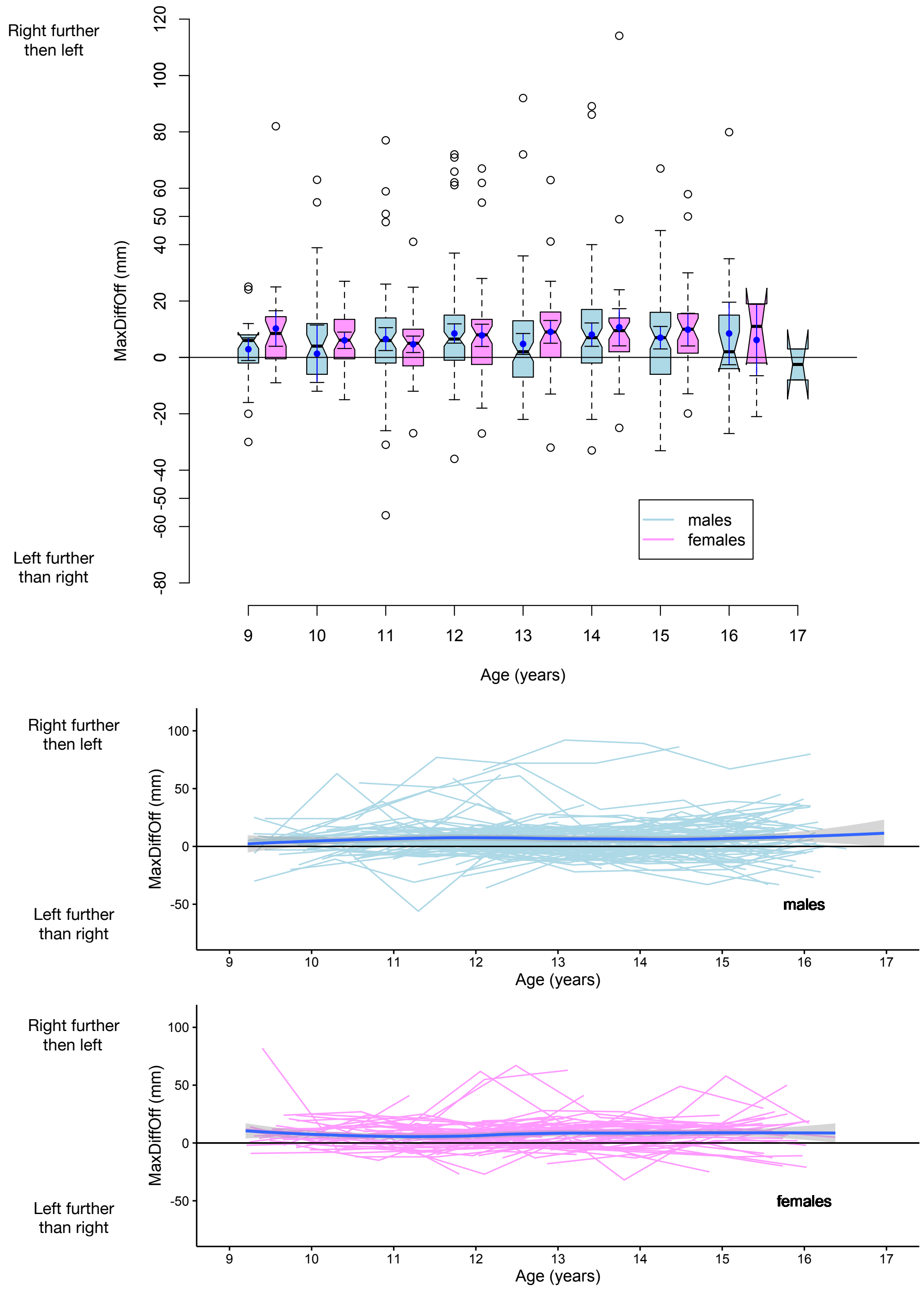


Figure 68: MaxDiffOff (mm) versus age (years) for males and females in the non-scoliotic cohort. MaxDiffOff is the difference in horizontal distance from the midline of the most prominent points (x coordinates) (see Figure 29 and Table 5.)

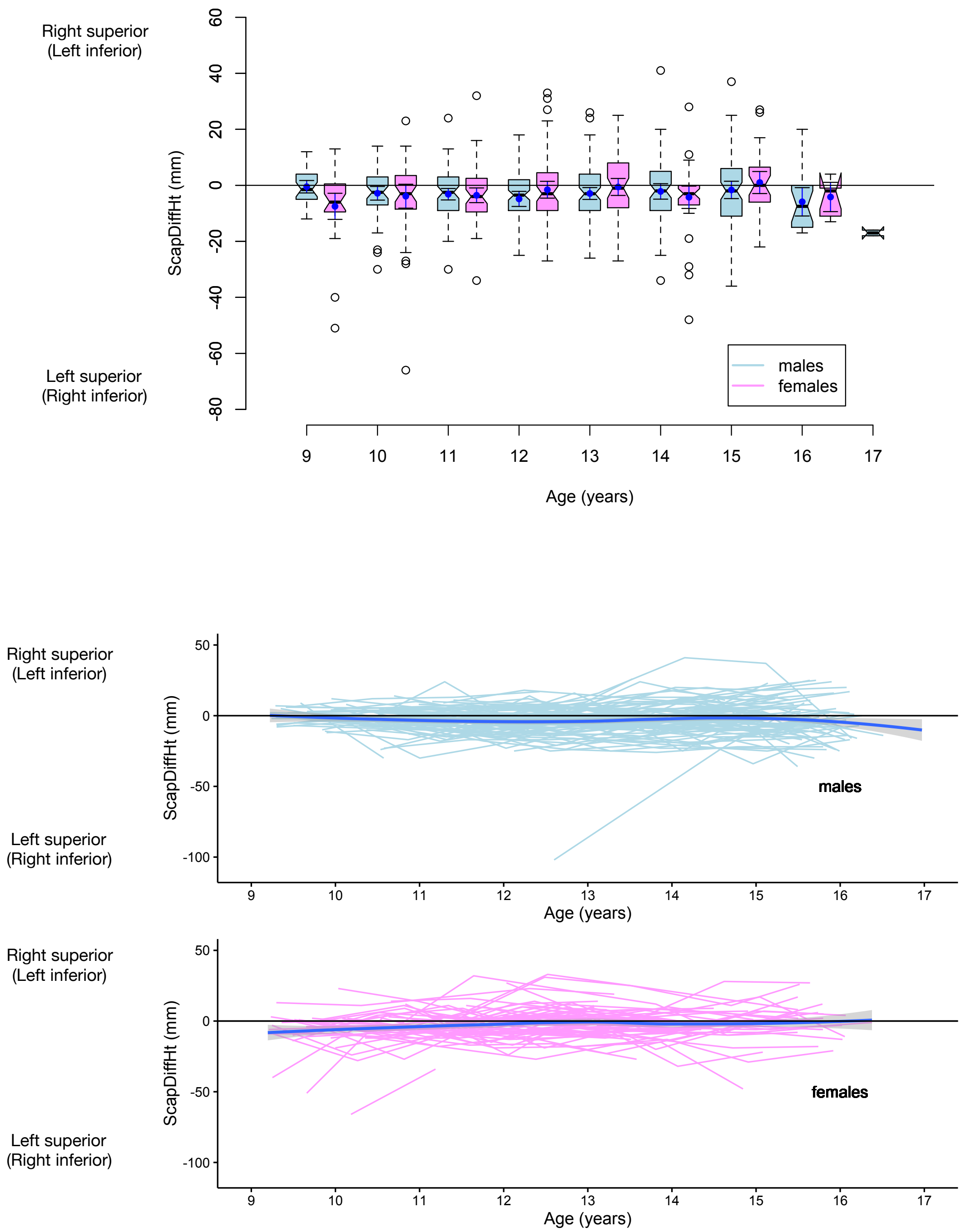
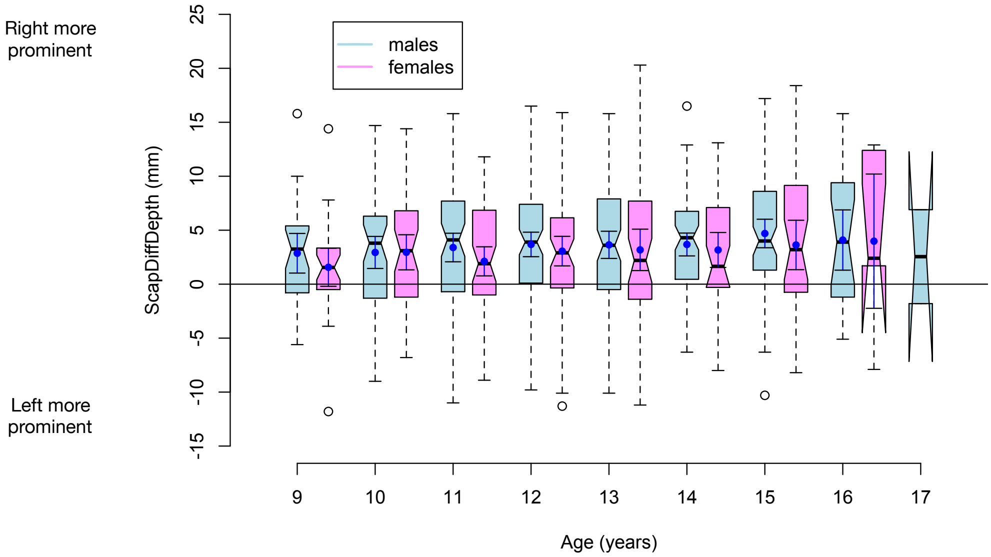
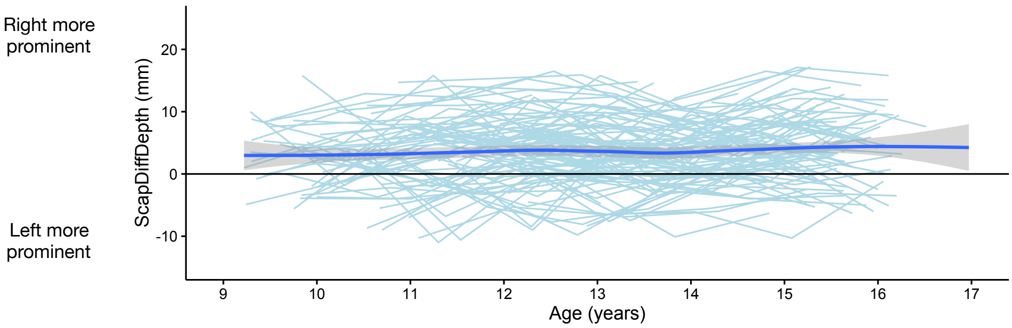


Figure 69: ScapDiffHt (mm) versus age (years) for males and females in the non-scoliotic cohort. ScapDiffHt is the difference in vertical height of the most prominent points (y coordinates) (see Figure 29 and Table 5.)





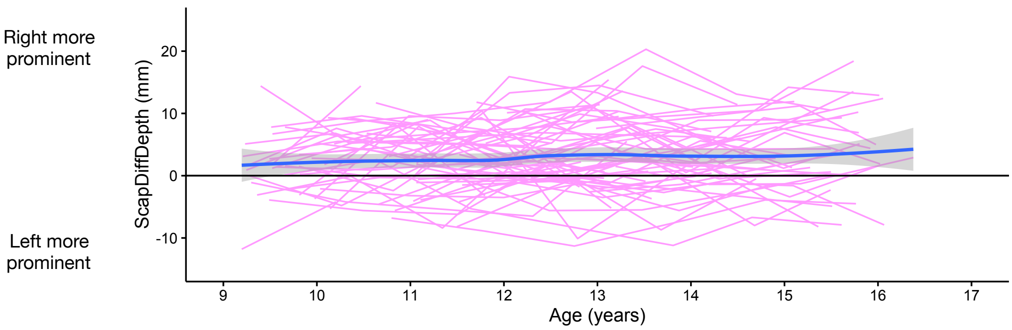


Figure 70: ScapDiffDepth (mm) versus age (years) for males and females in the non-scoliotic cohort. ScapDiffDepth is the difference in prominence of the most prominent points (z coordinates) (see Figure 29 and Table 5.)

6.8.3 Conclusions – most prominent points.

These results show that there is a difference between the most prominent point over the left and right side of the back with the right further from the midline, more inferior and further from the coronal plane (more prominent) than the left. There is no significant difference for sex. However, both MaxDiffOff and ScapDiffDepth are significant for age, where the difference between the sides is larger with increasing age showing that the right side becomes further away from the midline and more prominent than the left as the cohort aged. It will have been noted that there a number of outliers that seem at odds with the rest of the data presented. This may be because the most prominent points are simply the points furthest from the coronal plane through the subject, rather than any fixed anatomical landmarks. Consequently, as there is movement of the underlying skeleton, in this case the scapula, the most prominent point may change in position. It is likely that these outliers, whilst reflecting the position of the most prominent points at the moment in time that the image was captured, are not representative of the most prominent points for the majority of the time for that individual.

6.8.4 Results – volumetric asymmetry.

There were three distinct patterns of volumetric asymmetry seen in this cohort. There was either a single prominence on the right or left side (volr or voll only) of the torso or there were two prominences both right and left (volr and voll). When there were two prominences, either the right or the left was observed superiorly on the back with the other side sited inferiorly. Table 21 displays the distribution of these patterns of VA in the male and female group. Figures 71 to 78 show the VA against age and Table 22 shows the statistical significance for age and sex for the three different patterns of VA seen. Note that there were no 17 year olds in the group with only left sided VA.

Table 21: The number and percentage of the difference patterns of VA seen in the male and female groups in the non-scoliotic cohort. Volumetric asymmetry is a unit-less parameter that describes the difference in 3D volume between each side of the torso (see Figure 26).

	volr only	voll only	volr and voll
Male	263 (51%)	60 (12%)	192 (37%)
Female	139 (44%)	54 (17%)	123 (39%)

Table 22: The significance and coefficients for the parameters of age and sex of VA over the back in the non-scoliotic cohort. For an explanation of the terms in the table see section 6.3.1. Volumetric asymmetry is a unit-less parameter that describes the difference in 3D volume between each side of the torso (see Figure 26).

Parameter		Chi squared	p value	coefficients	standard error
volr only *	Age	32.43	<0.001	0.72	0.12
	Sex	0.92	0.338	- 0.72	0.76
voll only *	Age	0.58	0.45	0.123	0.16
	Sex	0.04	0.84	- 0.145	0.71
volr (as part of volr and voll) *	Age	13.51	< 0.001	0.401	0.11
	Sex	< 0.001	0.988	0.008	0.55
voll (as part of volr and voll) *	Age	1.08	0.299	0.061	0.06
	Sex	1.40	0.237	- 0.332	0.28

* Model simplified to random intercept only as there was a failure to converge with both random intercept and slope.

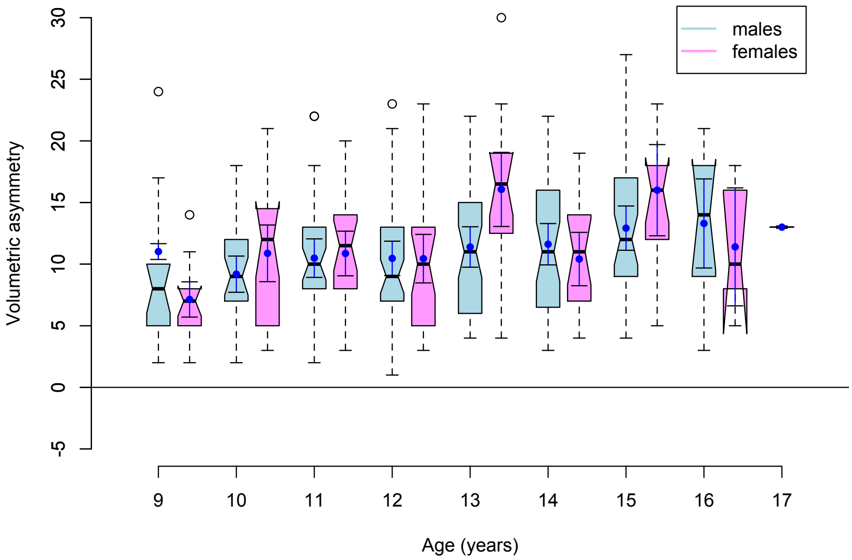


Figure 71: VA versus age (years) for males and females in the subgroup with only right sided VA (volr only) in the non-scoliotic cohort. Volumetric asymmetry is a unit-less parameter that describes the difference in 3D volume between each side of the torso (see Figure 26).

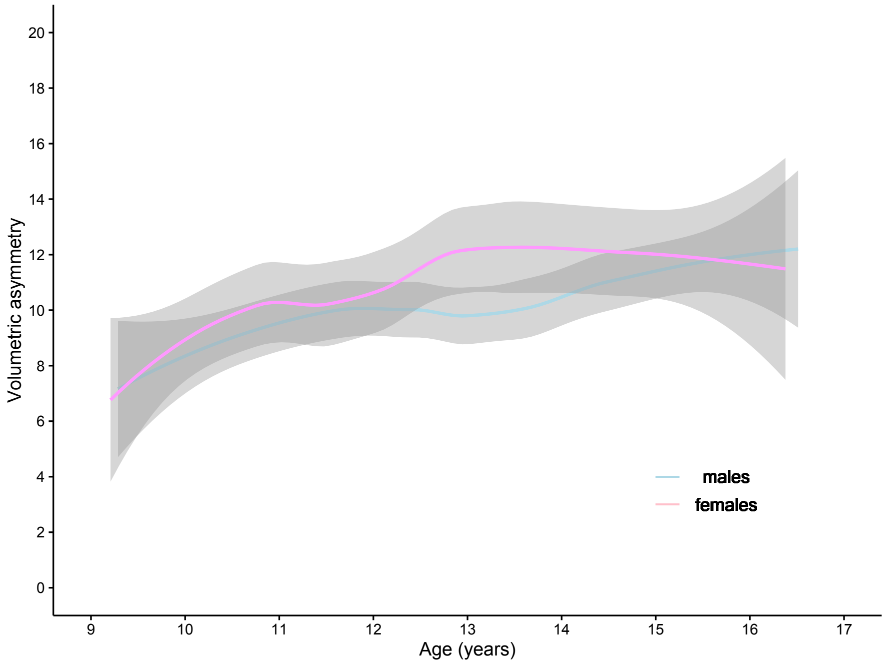


Figure 72: The LOESS line and 95% confidence interval of VA against age (years) in the subgroup with only right sided VA (volr only) for males and females in the non-scoliotic cohort. Volumetric asymmetry is a unit-less parameter that describes the difference in 3D volume between each side of the torso (see Figure 26).

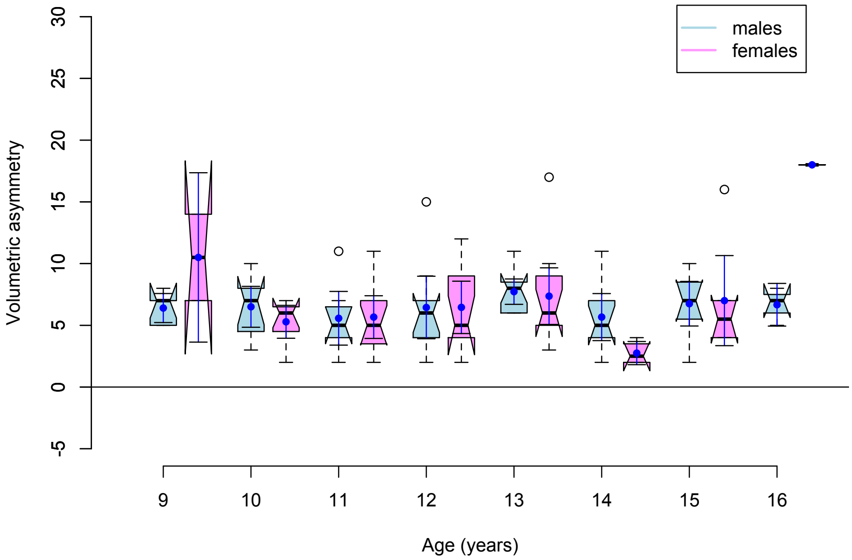


Figure 73: VA versus age (years) for males and females in the subgroup with only left sided VA (voll only) in the non-scoliotic cohort. Volumetric asymmetry is a unit-less parameter that describes the difference in 3D volume between each side of the torso (see Figure 26).

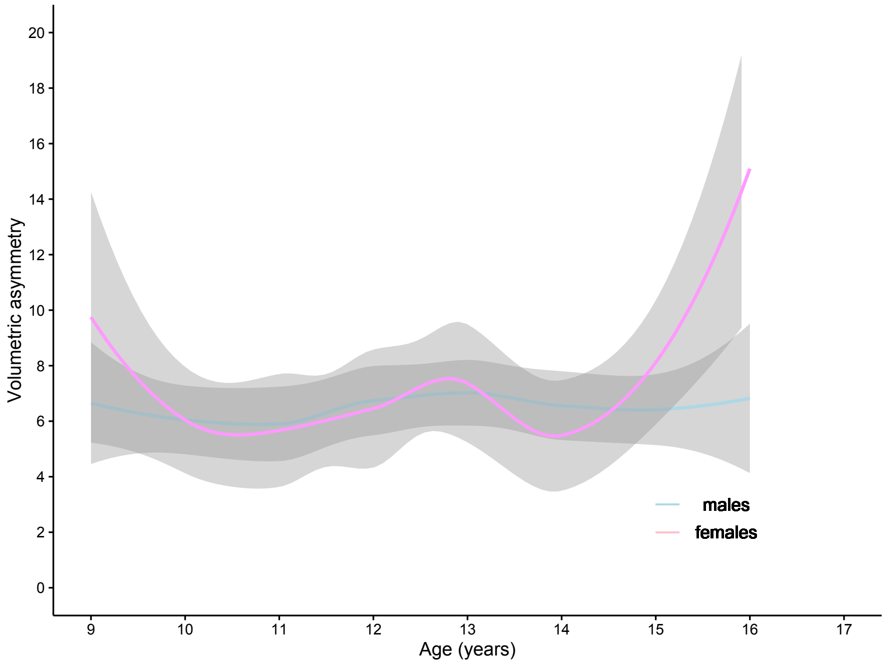


Figure 74: The LOESS line and 95% confidence interval of the VA against age (years) in the subgroup with only left sided VA (voll only) for males and females in the non-scoliotic cohort. Volumetric asymmetry is a unit-less parameter that describes the difference in 3D volume between each side of the torso (see Figure 26).

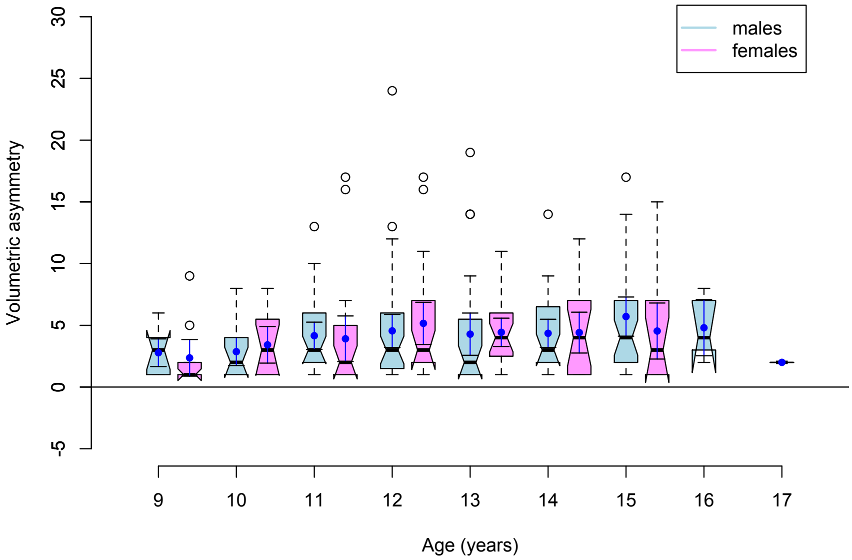


Figure 75: Right sided VA versus age (years) for males and females in the subgroup with both right and left sided VA (volr and voll) in the non-scoliotic cohort. Volumetric asymmetry is a unit-less parameter that describes the difference in 3D volume between each side of the torso (see Figure 26).

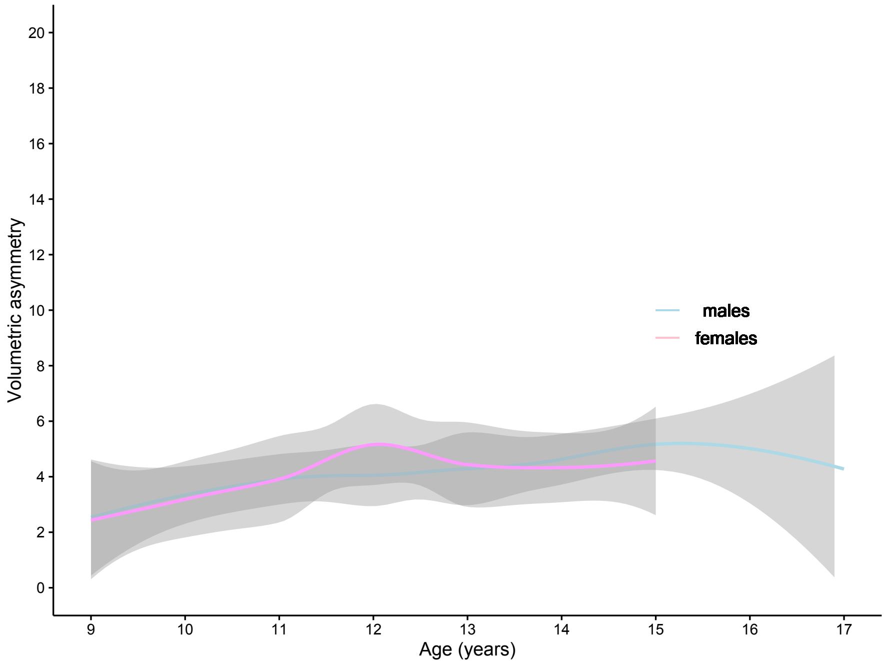


Figure 76: The LOESS line and 95% confidence interval of the VA against age (years) in on the right side of the torso in the subgroup with both right and left sided VA (volr and voll) for males and females in the non-scoliotic cohort. Volumetric asymmetry is a unit-less parameter that describes the difference in 3D volume between each side of the torso (see Figure 26).

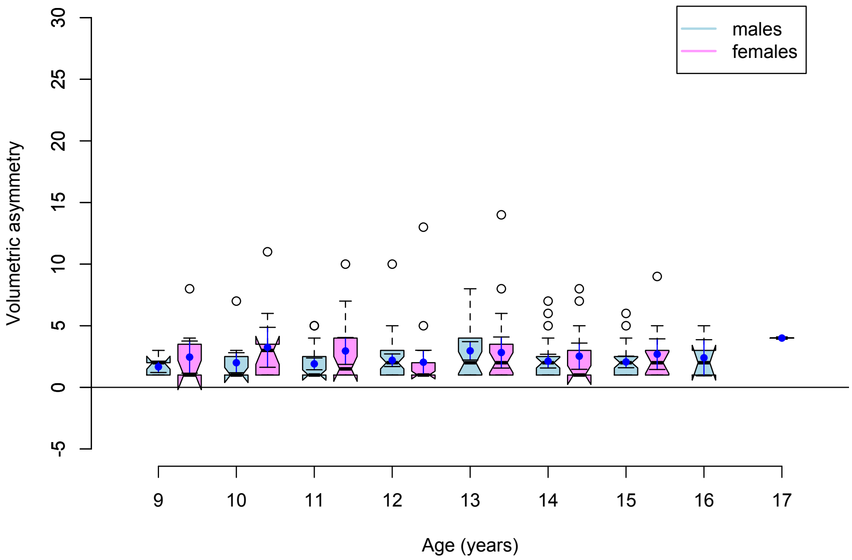


Figure 77: Left sided VA versus age (years) for males and females in the subgroup with both right and left sided VA (volr and voll) in the non-scoliotic cohort. Volumetric asymmetry is a unit-less parameter that describes the difference in 3D volume between each side of the torso (see Figure 26).

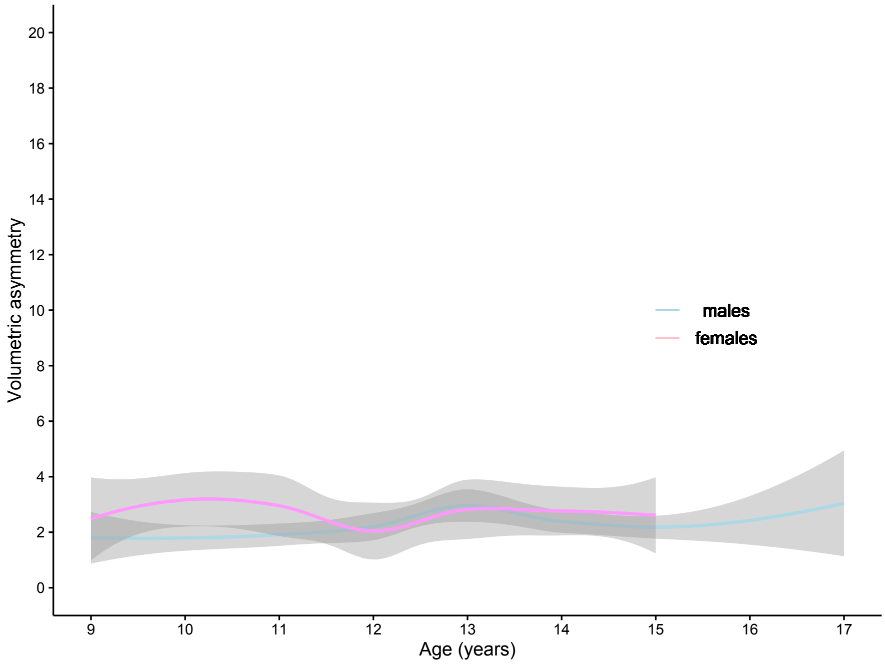


Figure 78: The LOESS line and 95% confidence interval of the VA against age (years) on the left side of the torso in the subgroup with both right and left sided VA (volr and voll) for males and females in the non-scoliotic cohort. Volumetric asymmetry is a unit-less parameter that describes the difference in 3D volume between each side of the torso (see Figure 26).

6.8.5 Conclusions – volumetric asymmetry.

These results show that there are three different patterns of VA seen in the normal cohort, namely right only (volr only), left only (voll only) and both right and left (volr and voll). If right or left only, then the magnitude of VA is larger than the equivalent in the combined right and left group. Interestingly, with increasing age, the right side, both as right only and right as part of combined right and left, shows increasing VA. Sex does not seem to make any difference to VA in any combination.

6.9 Power analysis.

A power analysis was performed based on the MCID reported by Akel et al [148]. Akel reported the difference in shoulder height in their cohort, in children who believed their shoulders to be equal in height and symmetrical, as 7.5 ± 5.8 mm. Using this figure, with a pre-defined power of 80% and a pre-defined alpha value of 0.05, a power analysis comparing two independent samples was performed. The MCID from Akel et al [148] was used as the reference figure, as the technique for measurement described by Akel is exactly replicated in the methods described within this thesis (the measure of ShDiffHt see Figures 27 and 28 and Table 4). The data from Akel et al [148] is used as there is very little data that exists in the literature other than the measures of shoulder height difference from Akel to base a power calculation on.

Comparing the results for the measurement of ShDiffHt in the non-scoliotic group (4.4 ± 8.5 mm) within the parameters specified, 119 measures were required to adequately power this investigation. This figure is less than both the number of participants and the number of individual measures, allowing the conclusion that the investigation is adequately powered.

6.10 Overall discussion of parameters describing growth in the normal cohort.

6.10.1 Summary of findings.

These data sets measure length, width, weight, spinal curvature (in the coronal and sagittal plane) and measures of 3D asymmetry in the torso of a cohort of children who were measured on sequential occasions using a standardised technique. For all parameters measuring body growth (height, weight, BMI, sitting height and standing to sitting height ratio), as the child ages these parameters increase. This is in keeping with the published growth standards (as seen in Chapter 5). With regards to the parameters of torso asymmetry (ShDiffHt, AxDiffHt, AxDiffOff, WaistDiffHt, WaistDiffOff), the mean values and surrounding variability show the range of the data. Thus this gives the range of normality given that all of the participants had no history or evidence on examination of a spinal deformity. When combined with information on spinal shape as data ellipses, the relationship of anatomical torso points and the coronal shape of the spine is demonstrated. Again, there is a range of results around a perfectly straight and perfectly symmetrical spine which represents the variability in a non-scoliotic population. The change of kyphosis and lordosis, particularly with reference to the differences seen between males and females is of note. The general reduction in the amount of kyphosis in females, being less than males, combined with a greater and increasing lordosis in females compared to males is a key finding. The measures of 3D shape, namely the most prominent points and VA, show differences in that the most prominent points are reasonably static and do not change whereas VA increases, particularly in those with a predominantly right sided asymmetry to the torso.

With regards to the distribution of the data, an inspection of the box and whisker plots shows that the mean and median values are very similar in all composite box plots other than for VA, apart from at the older ages where there are a smaller number of participants suggesting a normal distribution of the data. This is not seen to the same extent for the composite box plots for VA and thus the data has some skew in it. The LOESS line plots are more illustrative of the trend in these cases.

6.10.2 Relevance of findings.

This is a true longitudinal design and represents standards for normality in all of the parameters which can be used for future work. It is of note that the statistically significant differences between males and females in the parameters presented in this chapter were in width with males being wider at both the axillae and the waist, also in sitting height with males being taller than females. This is in agreement with the biacromial distance published by Malina [90] and is to be expected given the anatomical proximity of the acromion and the superior end of the posterior axillary fold. It is inappropriate to compare the bicristal distance of Malina [90] to the waist width presented here as they are anatomically different measures.

Concept of symmetry. The concept of symmetry with regards to the torso is reasonably straightforward. With a line of symmetry running in a straight line down the length of the spine in the coronal plane, any anatomical point on one side of the torso, reflected around that line of symmetry would fall on the same anatomical point on the other side of the torso. The anatomical points marked as the shoulder, axillae and waist would be at the same vertical level and the axillae and waist would be the same distance from the midline. The data suggests, however, that in those without spinal or thoracic deformity, there is a range of differences away from perfect symmetry with regards the shoulder, axillae and waist. Again the results define what is normal for future studies in this area. As the methodology was the same as that of Akel et al [148], a direct comparison can be made between the results of the current study and those of Akel. The average height difference of the shoulders quoted by Akel was 7.5 ± 5.8 mm. In the data set presented here the average for the whole cohort (not subdivided for age or sex) was 4.5 ± 8.5 mm. A weakness of the paper by Akel is the grouping together of all ages and both sexes. The current data shows that shoulder height does change with age and this finer detail is lost when all ages are reviewed as one. This issue of interpretation is also seen in other papers where age is not treated as a continuous variable and/or the study design is cross sectional but where the authors imply that the results can be viewed in a longitudinal manner [101, 102, 144, 145]. As can be seen from the data presented in this current work, the loss of true longitudinal study design leads to a loss of understanding of any true change that occurs with growth.

The torso points. The torso points documented here show that whilst there is a range across the value of perfect symmetry, the mean value was within 1 cm of perfect symmetry (the greatest deviation seen in ShDiffHt). There were significant differences seen for age in ShDiffHt and AxDiffHt and for sex in WaistDiffOff. Whilst these comparisons were statistically significant, a maximum difference of 1 cm is unlikely to be clinically significant as evidenced by the Akel paper [148] where all participants thought their shoulders were level. The observation from this current data is that ShDiffHt increases with age in both males and females with the right shoulder becoming lower than the left (Figure 46). This is difficult to explain but would still be within the limits of physiological asymmetry for shoulder height put forward by Vercauteren et al [144] as less than 10 mm.

It is not possible to directly compare the WaistDiffOff and WaistDiffHt data presented with the literature. The closest similar measure is that of Vercauteren [144] (although the measure is different as it is an asymmetry in the measure of the depth of the waist triangles from a vertical line drawn lateral to the body). Vercauteren defines physiological and postural asymmetry as a difference of less than or equal to 15 mm. Whilst the mean value for WaistDiffOff is within this value, the range of the data exceeds this. This reflects the greater ambiguity over the position of the waist, especially in those without deformity as, unlike with the axilla or shoulder, there is not a well defined anatomical point (e.g. the superior end of the posterior axillary fold) on which to base the measurement. This issue is particularly seen in young males before their growth spurt where the torso can look like a cylinder between the axillae and the iliac crests with little discernible waist to measure.

The main and compensatory coronal curve data shows that in the majority of the cohort there is a small amount of curve, either as a single curve or as double curves. At the extremes of the range, some of the points are greater than the definition of scoliosis (an angulation of greater than 10°) [8] but the majority are within this cut off and would not be classed as scoliosis. The main and compensatory curve data confirms the definition of Kane (that states that scoliosis is a coronal curve of a Cobb angle of greater than 10°) [8] and highlights that the spine in the coronal plane is not straight in the majority. In a similar way to the patterns of curve seen in AIS [55], most curves seen were main thoracic with compensatory thoracolumbar curve and main thoracolumbar with compensatory thoracic curve.

The interactions of the coronal curve type and the anatomically close torso points show that, in a normal cohort without spinal deformity, there is an association between the shape of the spine and shape of the torso and this establishes a normal range for future work. What is of particular note, is the difference that an increasing thoracic curve has on the shoulder points and the corresponding axillae points (Figures 55, 56 and 57). As the curve size increases, convex to the right, the right axilla point becomes higher (AxDiffHt) and further from the midline (AxDiffOff) than the left (and vice versa for convex to the left). This is not seen for the shoulder points (ShDiffHt) where the vertical height of the left and right shoulder points remains similar despite the underlying shape of the spine. It is likely that this occurs due to the anatomical relationship of the shoulder girdle and underlying thoracic cage. The only skeletal attachment of the shoulder girdle complex to the thoracic cage is via the sternoclavicular joint. Through positioning of the shoulder girdle around the thoracic cage via this attachment, it is possible to keep the hands an equal distance from the floor and hence the shape of the ellipse of ShDiffHt (Figure 55). The effect of compensatory curve in both curve patterns is less marked on the anatomically proximal torso points with the data ellipse being more circular in shape.

Measures of spinal shape – sagittal parameters of the spine (kyphosis and lordosis). The sagittal measures of kyphosis and lordosis differ from those published in the literature previously as the results here are represented in a truly longitudinal fashion. Both kyphosis and lordosis increase with age (Figures 65 to 66). Lordosis increases in an approximately linear fashion and there is a statistically significant difference for sex with females having a larger lordosis than males at all ages (Figure 67). The change in kyphosis is not linear with a decrease for females below the starting value of kyphosis for age 9 between the ages of 11 and 14 years with a minimum value at age 12.5 years, and then an increase following that. Males on the other hand have an increase in kyphosis over the same period and decrease again later on. These differences are not significant. The pattern of the change in kyphosis (Figure 67) does appear similar to that reported by Willner et al [27] and casts doubt on the results of Carr et al [145] and Giglio and Volpon [166]. Other literature, reporting results with methods where all ages are grouped together or where no distinction is made for the difference caused by sex [29, 159, 161, 162, 163, 164, 165] does not show these differences and suggests that these approaches are too broad and lose important detail. The statistical significance of sex in the measurement of lordosis, with females having a greater lordosis than males is of note. One of the suggested mechanisms for the development of AIS, put forward by the Utrecht group [204, 205], is where the initiating factor is a posteriorly directed load across the facetal articulations, a biomechanically disadvantageous situation that leads to reduced segmental stability. Given the greater lordosis and the smaller kyphosis when comparing females to males, it follows that there will be more levels in the spine where there are posteriorly directed forces. This adds to the possible reasons why AIS may be more common in females than males [206, 207].

Most prominent points. The most prominent points over the back are the points that are furthest from the coronal plane. In essence they are the 'points that stick out the most' and are usually found in the region of the scapula. These points form part of the DAPI assessment of posterior shape [106]. The most prominent points are represented in 3D, with parameters describing the position of the points in 3D also. The parameter MaxDiffOff (Figure 68) is the difference in the distance of the points laterally from the midline in the coronal plane (x coordinate). The parameter ScapDiffHt (Figure 69) is the difference in the position of the points in a superior-inferior direction in the coronal plane (y coordinate). The parameter ScapDiffDepth (Figure 70) is the difference between the prominence of the points away from the coronal plane, or height of the points (z coordinate). In the symmetrical back all of these parameters would equal zero as the right and left sides of the back would be identical in shape. The data presented here shows the mean value is close to zero for all three parameters. However in both MaxDiffOff and ScapDiffDepth the right side is further from the midline and more prominent than the left. For ScapDiffHt, there is a trend for the left to be slightly superior to the right. There is no difference between males and females for any of the three parameters. There is however a significant change for age for the parameters MaxDiffOff and ScapDiffDepth showing that as both males and females age, the difference between the right and left points increases in both distance from the midline and prominence from the coronal plane.

Volumetric asymmetry. Volumetric asymmetry (VA) is a unit-less parameter that indicates the volume of any prominence of the back, indicating the size of the rib hump or lumbar hump. Whilst this concept is similar to the most prominent points over the back, it is different as the most prominent points are a single point in three dimensions whereas VA is a size measure of a three dimensional volume. The work of Nissinen et al [149, 150, 151] has shown that, in a normal cohort, there is a spectrum of differences in the shape of the back that changes with age. The data presented here show that, in the normal cohort without spinal deformity, there is a subdivision into three distinct groups. There are those with only right sided VA and prominence, those with only left sided VA and prominence and those with both left and right sided VA and prominence. The results for the changes in VA with ageing for both males and females are shown in this way (Figures 71 to 78). The subgroup of right sided VA only is the most commonly seen in both males and females. The pattern of left sided VA only is the least frequently occurring in both males and females. In the right sided VA only group, VA increases with age and this is seen to a lesser extent in the right sided VA measure in those with right and left sided VA group. For left sided VA, both as the only prominence and as part of those with both left and right sided VA, there seems to be little change with age. This is in comparison with the only study measuring the same parameter [145] where VA was a parameter that only increased in males on the right hand side (Figure 12 in the Literature review).

The findings with regard to the most prominent points over the back and VA demonstrate that in a normal population without spinal deformity, the shape of the back is not symmetrical. There is no statistically significant difference with regards sex for any of the parameters but age does play a role and there is an increase with age for the parameters of MaxDiffOff, ScapDiffDepth and right sided only VA.

This chapter demonstrates the standards for growth in children without spinal or thoracic deformity during their adolescent growth. This is the first time that these parameters have been reported in a truly longitudinal fashion documenting the differences of sex and the changes with age. This provides important normative values for future work.

7 Pre-operative to post-operative change.

7.1 Introduction.

The aims of scoliosis surgery are the prevention of progression in the size of the scoliosis, correction of the spinal shape and a safe neurological outcome. The minimum acceptable outcome of a scoliosis operation is the prevention of further progression without any attempt in the correction of spinal shape. On its own, this minimum is a very uncommon event; it is usually related to the inability to effect a correction because of difficulties in the monitoring of spinal cord function during the procedure. The vast majority of scoliosis operations aim to change the shape of the spine and torso to give a symmetrical torso with the recreation of a normal spinal shape, that is no scoliosis and normal amounts of thoracic kyphosis and lumbar lordosis.

This chapter details the 2D changes that occur in the spine and torso with surgery for AIS on the scoliotic cohort. This is compared to the range of values for the same parameters that were obtained from the non-scoliotic cohort of children as documented in Chapter 6.

7.2 Methods.

Initial review of the data showed that the scoliotic curves seen in the cohort divided into two distinct groups along the lines described by Lenke et al [55], in a similar way to the curves seen in the non-scoliotic cohort. These were a main thoracic curve with a compensatory thoracolumbar curve and a main thoracolumbar curve with a compensatory thoracic curve.

From the subgroup who had this data recorded (as detailed in Section 4.1 and in Table 23), the parameters of standing height, weight, BMI, sitting height and sitting height to standing height ratio were plotted on to the appropriate UK-WHO standard

[82] and sitting height standard [83]. In this case, as the standards are sex specific, the data were analysed and are presented, subdivided into the main thoracic and main thoracolumbar curve patterns, for both males and females. In a similar way to the growth standards in the chapter assessing the non-scoliotic data on the UK-WHO growth standards, the number of data points above and below the median line and the 5th and 95th centiles were compared.

With the same methodology as in Chapter 6 on the shape of the normal child, the anatomical points seen in Figures 27 and 28 were identified. This shape analysis was done using combined data for the males and females due to the small numbers of males with scoliosis. Using these measured parameters, graphical and statistical descriptions of shape were created. For the torso points and coronal curve, in the main thoracic curve type, data ellipses [202, 203] were created to show the mean and 95% confidence interval. Given the smaller numbers in the main thoracolumbar group and the skewed data seen in the box and whisker plots, data ellipses for this group were created using the median and 95% centiles. In both cases the plots show the pre-operative data, post-operative data (data points, mean or median and the 95% confidence or centile ellipse) together with the mean and 95% confidence ellipse of the data from the normal cohort. The individual data sets are shown as box and whisker plots. Statistical analysis was undertaken to compare the distributions of the pre-operative data to the normal data, the post-operative data to the normal data and the pre-operative data to the post-operative data using Student's t test in the main thoracic group and Wilcoxon sum rank test in the main thoracolumbar group. Statistical significance was defined as $p < 0.05$.

Similar methodology was used in analysing the thoracic kyphosis and lumbar lordosis for both the main thoracic and main thoracolumbar groups. The most prominent points over the scapula were also analysed using this methodology. Composite ellipse diagrams with box and whisker plots are presented for MaxDiffOff, ScapDiffHeight and ScapDiffDepth.

Following the divisions seen in the groups for volumetric asymmetry (VA) in the non-scoliotic cohort, the data was analysed in the groups of those with right sided only VA, those with left sided only VA and those with both right and left sided VA, subdividing for the main thoracic or thoracolumbar curve pattern. The data were analysed with plots that show VA in the pre-operative and post-operative cohorts and the size of the change effected through the surgery.

7.3 Results.

7.3.1 Results - The analysis of standing height, weight, BMI, sitting height and sitting height to standing height ratio on growth standards.

These data were then plotted on to the UK-WHO growth standards for standing height, weight and BMI [82] and the Dutch standards for sitting height and sitting height to standing height ratio [83] (Figures 79 to 93). Summary statistics of the data are presented in Table 23 with statistical significance calculated using Student's t test. The main thoracic curve pattern males are shown in Figures 79 to 83, the main thoracic curve pattern females are shown in Figures 84 to 88 and the main thoracolumbar curve pattern females are shown in Figures 89 to 93. The distribution of the pre-operative and post-operative data is shown through the percentage of the data points below the 5th and 95th centiles and the median line and this is shown in Tables 24, 25 and 26.

Table 23: The mean, standard deviation and range for the pre-operative and post-operative values of standing height, weight, BMI, sitting height and sitting height to standing height ratio with the significance of the change caused by the surgery (males n=20, females n=153).

Parameter	Sex	Pre-operative mean value (SD, range)	Post-operative mean value (SD, range)	Significance
Standing height (cm)	Male	169.4 (9.4, 149.5 to 193.3)	175.3 (8.7, 156.6 to 200.1)	<0.001
	Female	157.9 (7.7, 134.0 to 175.3)	162.6 (6.6, 140.2 to 177.8)	<0.001
Weight (kg)	Male	58.4 (13.3, 34.8 to 91.0)	62.7 (16.3, 40.6 to 109.7)	0.003
	Female	51.7 (11.2, 29.1 to 90.4)	54.7 (10.9, 36.4 to 81.3)	<0.001
BMI (kg/m^2)	Male	20.3 (4.2, 15.6 to 30.3)	20.4 (4.9, 16.5 to 33.6)	0.820
	Female	20.7 (3.9, 14.1 to 34.3)	20.7 (3.8, 15.0 to 30.8)	0.971
Sitting height (cm)	Male	84.8 (5.5, 72.5 to 93.5)	88.6 (4.9, 78.0 to 97.0)	<0.001
	Female	80.1 (4.9, 67.3 to 91.8)	84.1 (4.1, 73.7 to 92.7)	<0.001
Sitting to standing height ratio	Male	0.50 (0.02, 0.46 to 0.53)	0.51 (0.02, 0.48 to 0.53)	0.071
	Female	0.51 (0.02, 0.47 to 0.57)	0.52 (0.01, 0.48 to 0.56)	<0.001

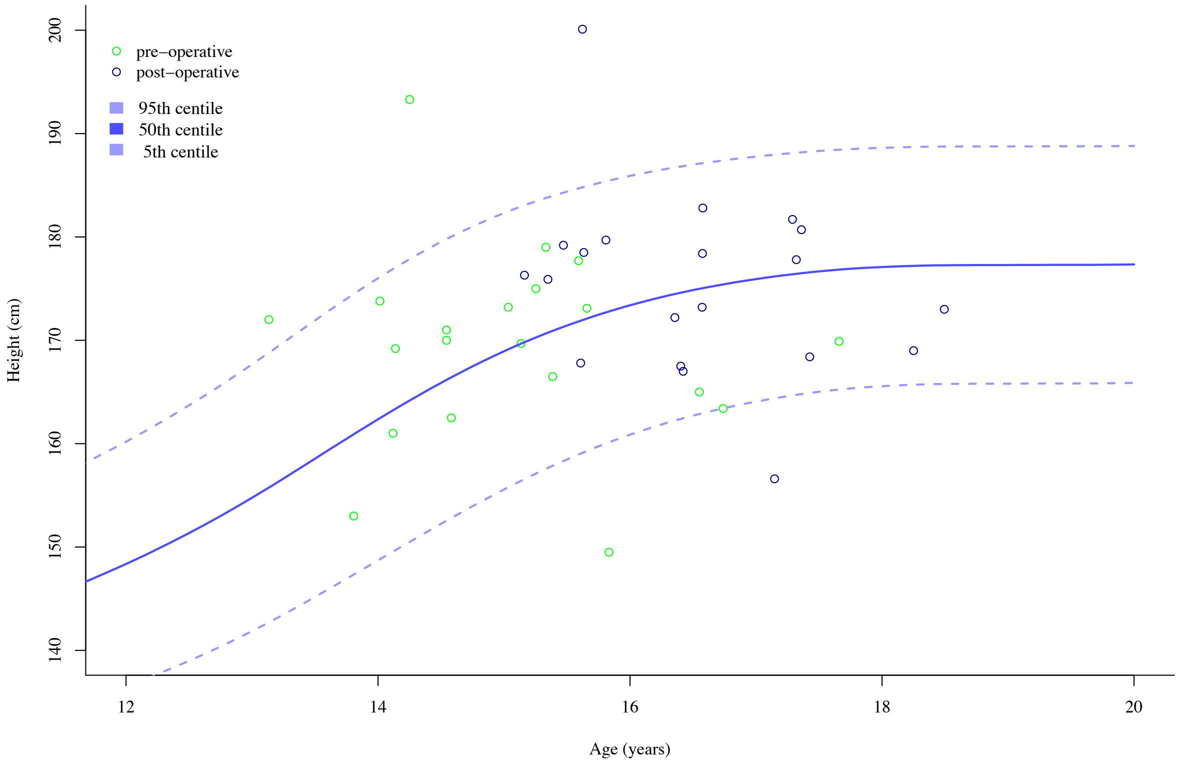


Figure 79: The distribution of the standing height (cm) versus age (years) in males with a main thoracic curve pattern on the UK-WHO growth standard redrawn from [82, 87]. For this plot n = 20.

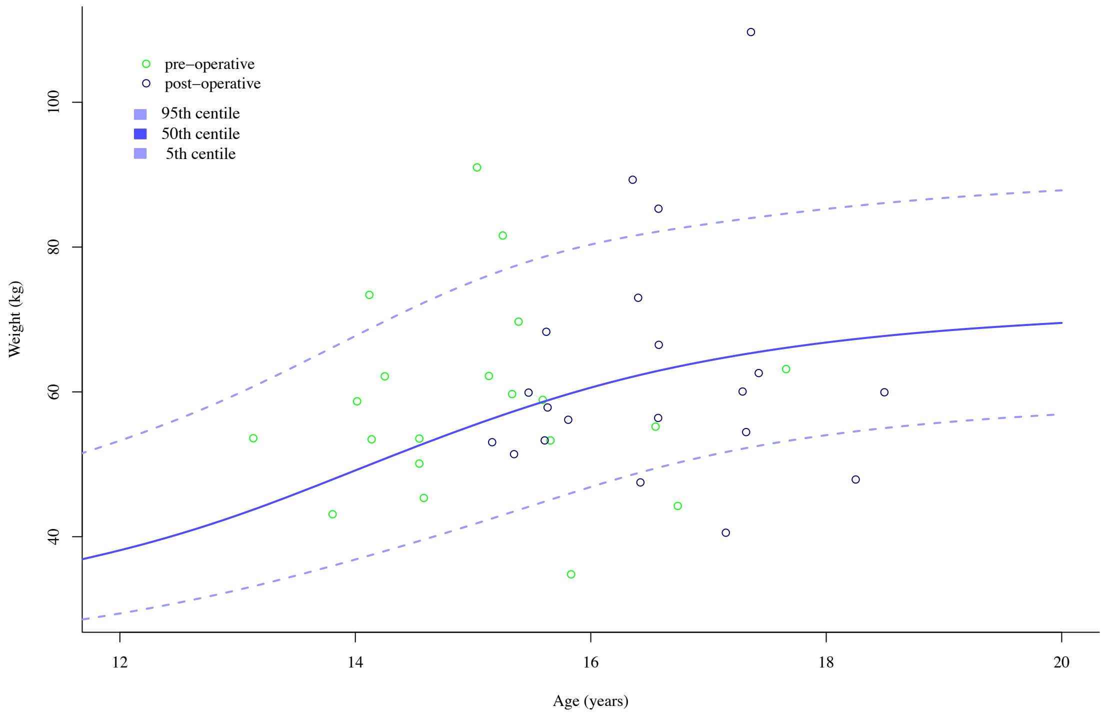


Figure 80: The distribution of the weight (kg) versus age (years) in males with a main thoracic curve pattern on the UK-WHO growth standard redrawn from [82, 87]. For this plot n = 20.

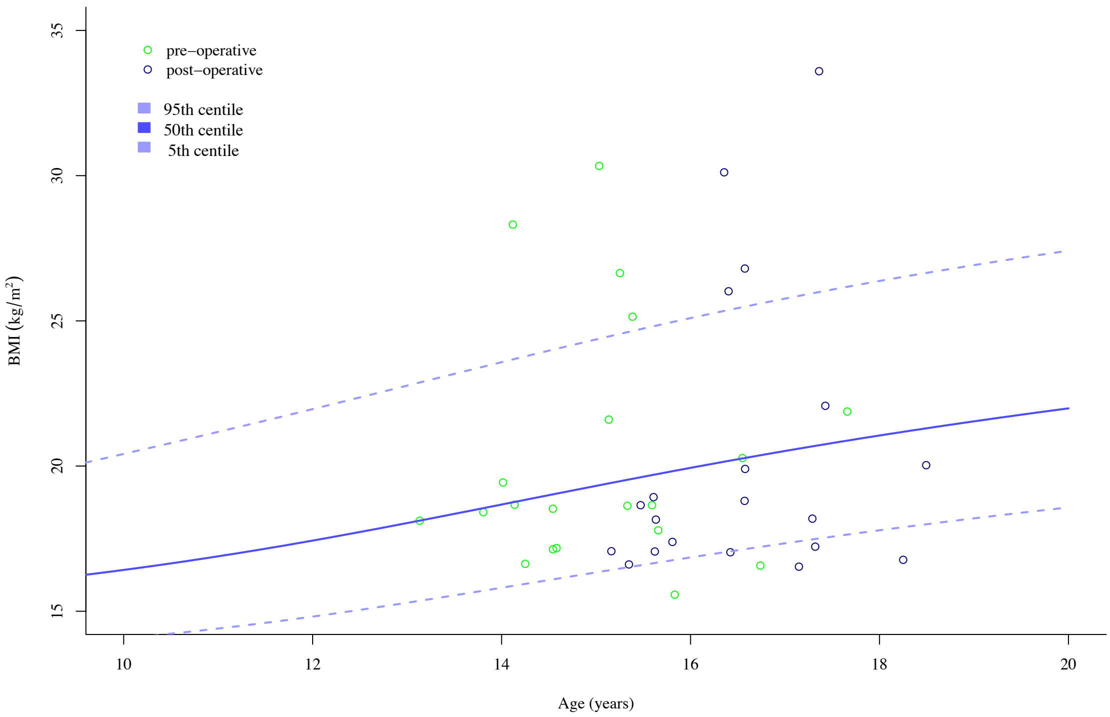


Figure 81: The distribution of BMI (kg/m^2) versus age (years) in males with a main thoracic curve pattern on the UK-WHO growth standard redrawn from [82, 87]. For this plot n = 20.

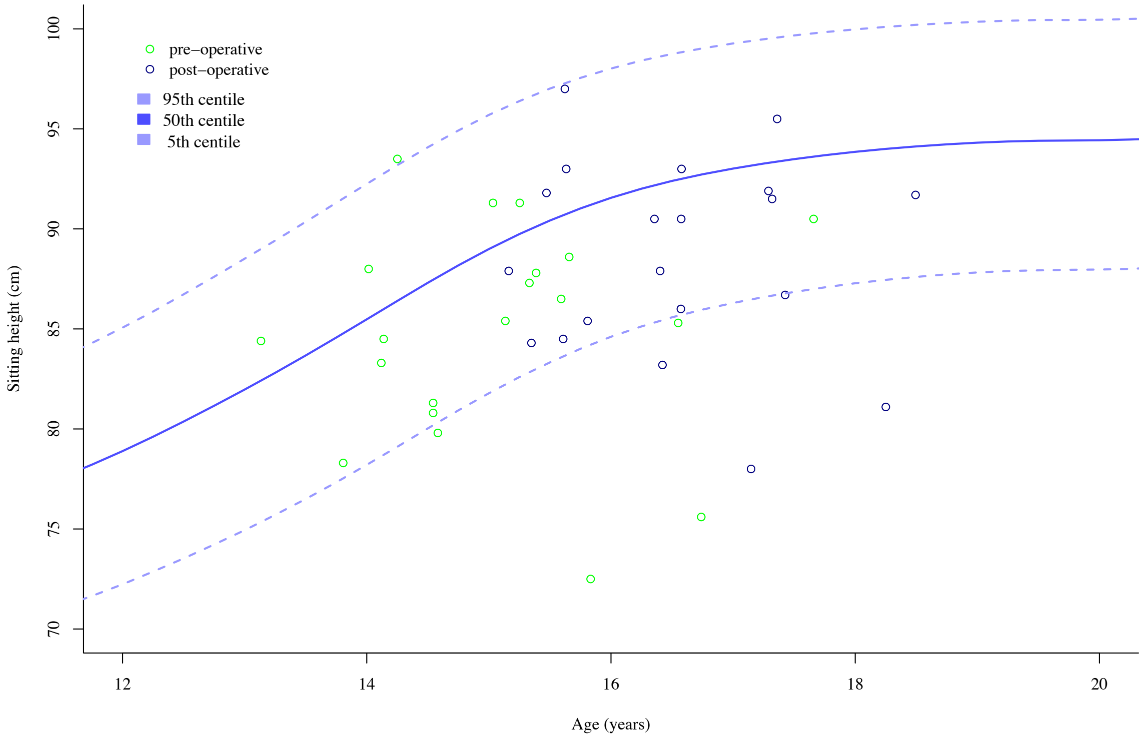


Figure 82: The distribution of the sitting height (cm) versus age (years) in males with a main thoracic curve pattern on the Dutch growth standard redrawn from [83, 87]. For this plot n = 20.

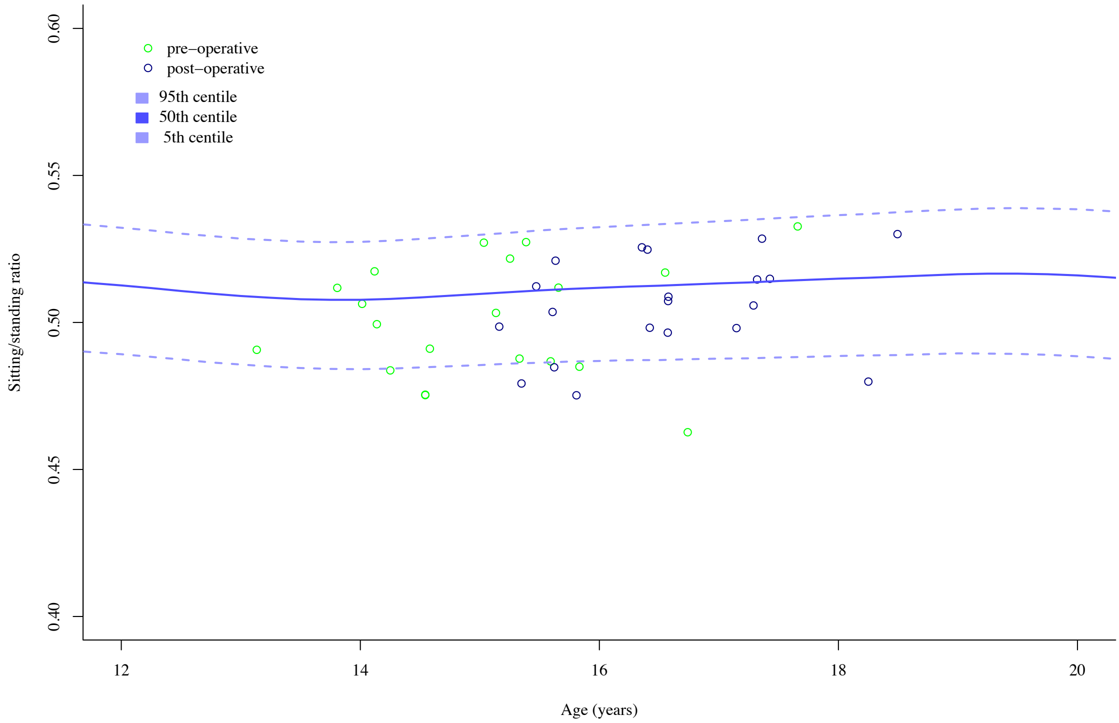


Figure 83: The distribution of the sitting height to standing height ratio versus age (years) in males with a main thoracic curve pattern on the Dutch growth standard redrawn from [83, 87]. For this plot n = 20.

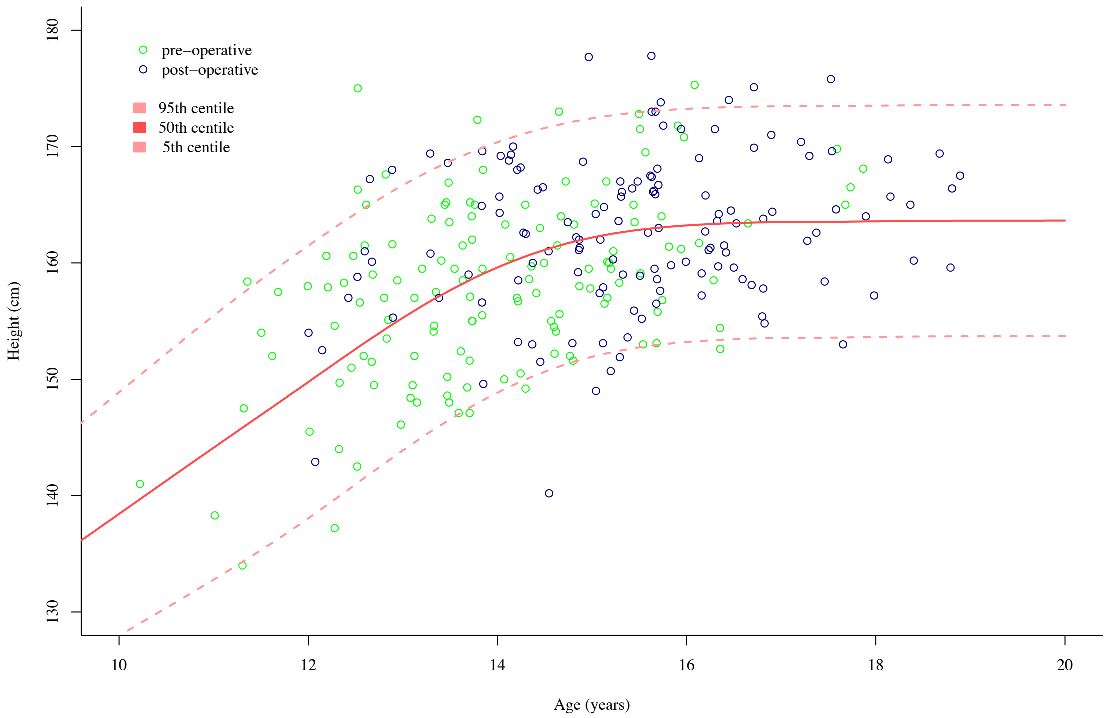


Figure 84: The distribution of the standing height (cm) versus age (years) in females with a main thoracic curve pattern on the UK-WHO growth standard redrawn from [82, 87]. For this plot n = 135.

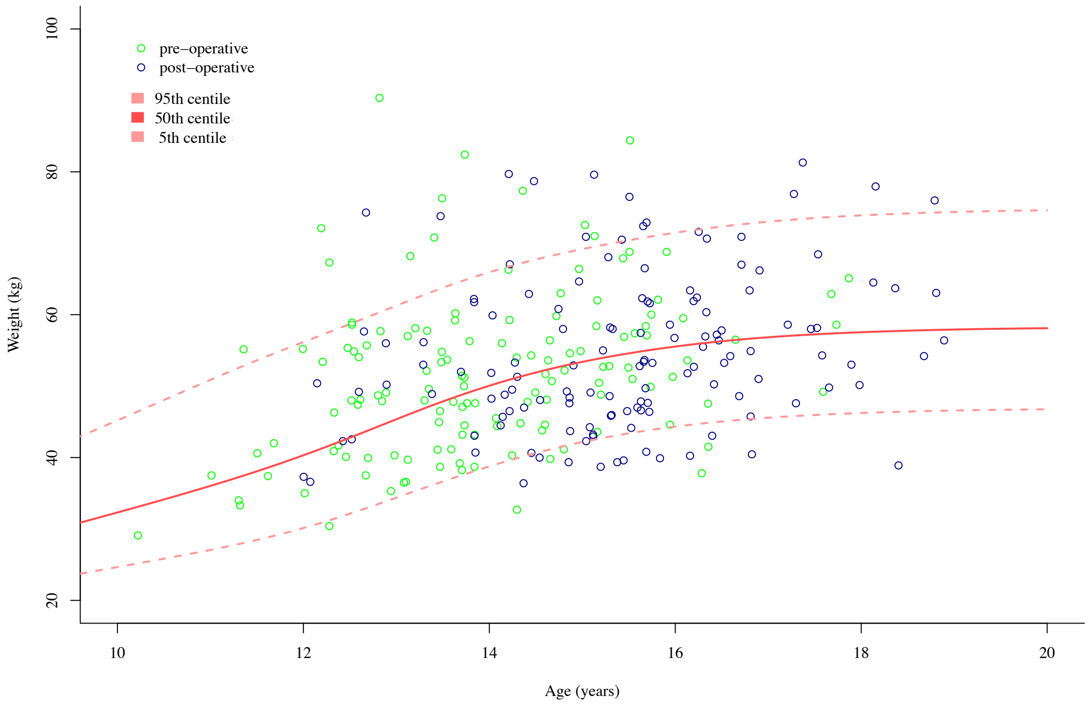


Figure 85: The distribution of the weight (kg) versus age (years) in females with a main thoracic curve pattern on the UK-WHO growth standard redrawn from [82, 87]. For this plot n = 135.

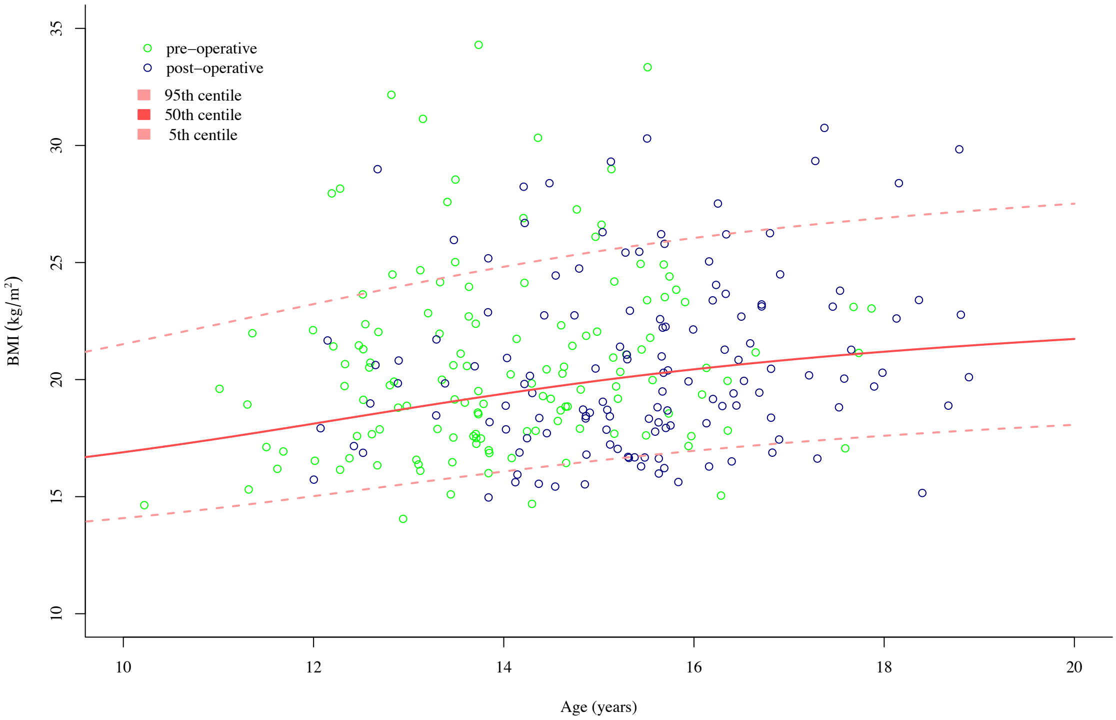


Figure 86: The distribution of BMI (kg/m^2) versus age (years) in females with a main thoracic curve pattern on the UK-WHO growth standard redrawn from [82, 87]. For this plot n = 135.

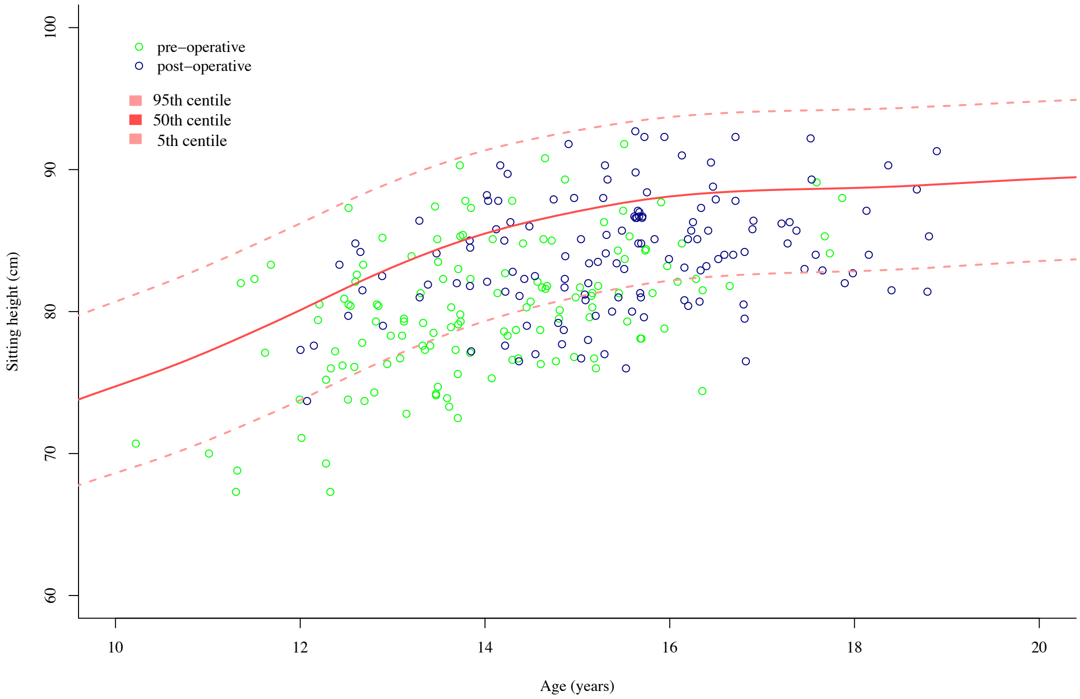


Figure 87: The distribution of the sitting height (cm) versus age (years) in females with a main thoracic curve pattern on the Dutch growth standard redrawn from [83, 87]. For this plot n = 135.

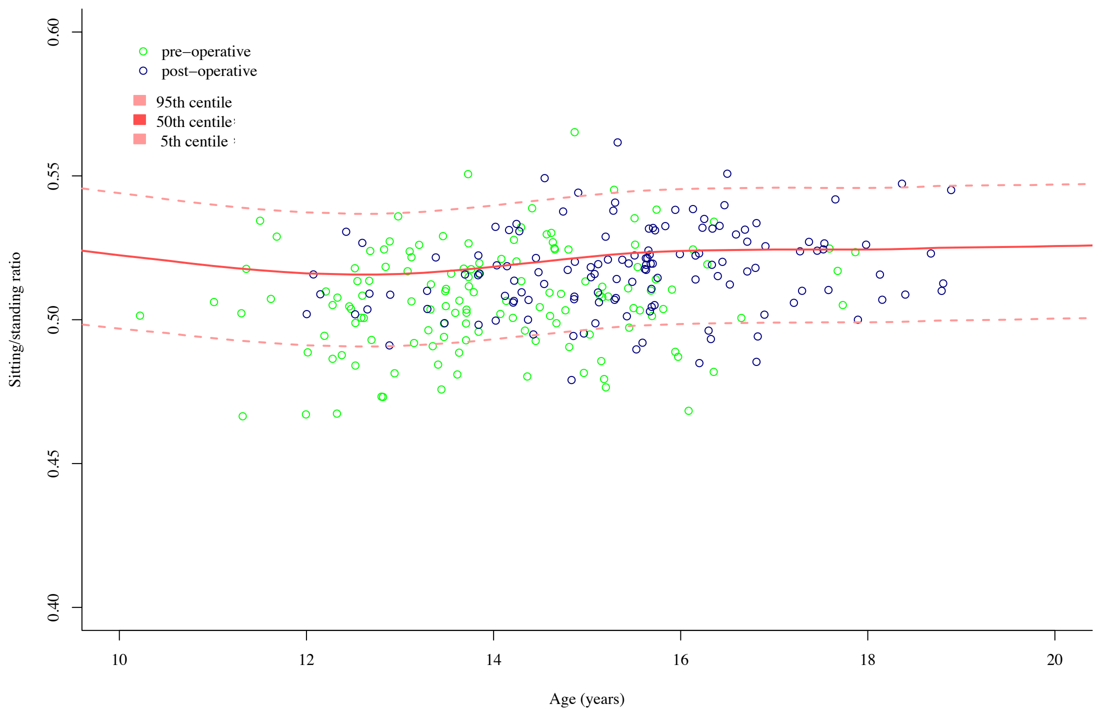


Figure 88: The distribution of the sitting height to standing height ratio versus age (years) in females with a main thoracic curve pattern on the Dutch growth standard redrawn from [83, 87]. For this plot n = 135.

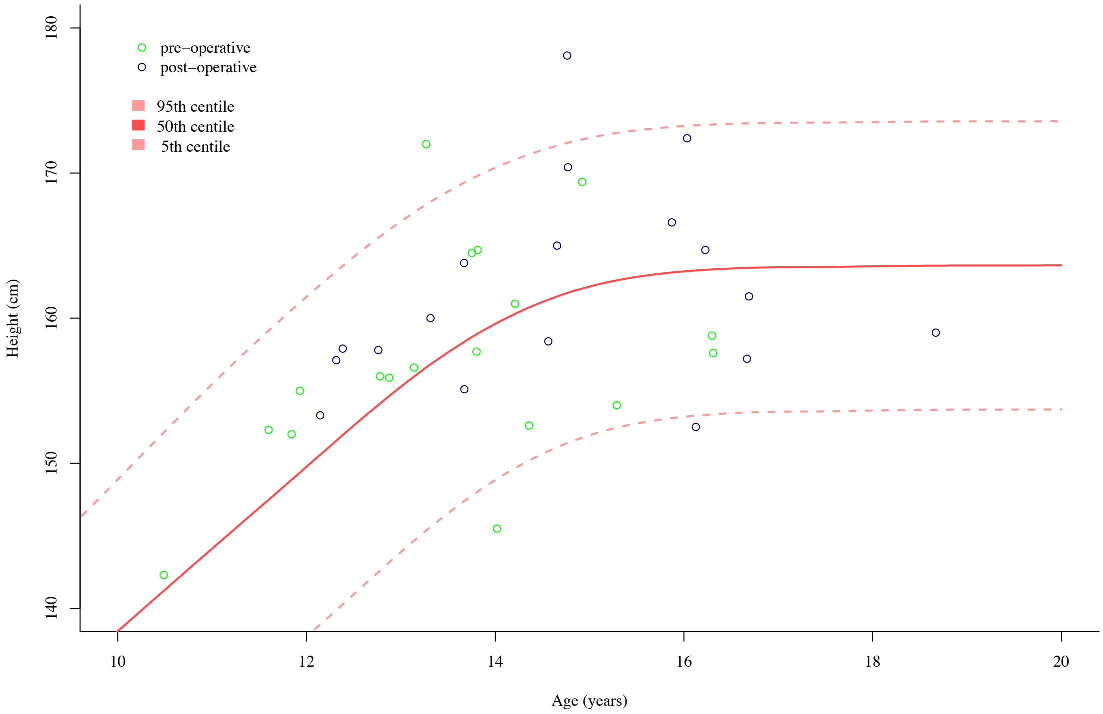


Figure 89: The distribution of the standing height (cm) versus age (years) in females with a main thoracolumbar curve pattern on the UK-WHO growth standard redrawn from [82, 87]. For this plot n = 18.

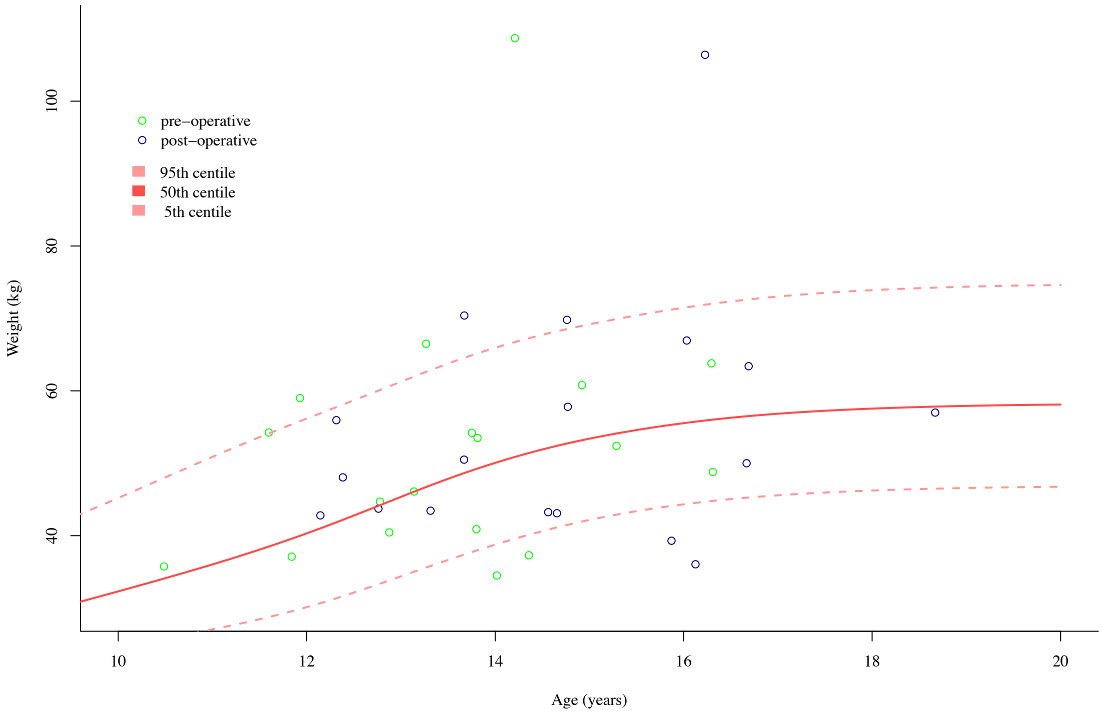


Figure 90: The distribution of the weight (kg) versus age (years) in females with a main thoracolumbar curve pattern on the UK-WHO growth standard redrawn from [82, 87]. For this plot n = 18.

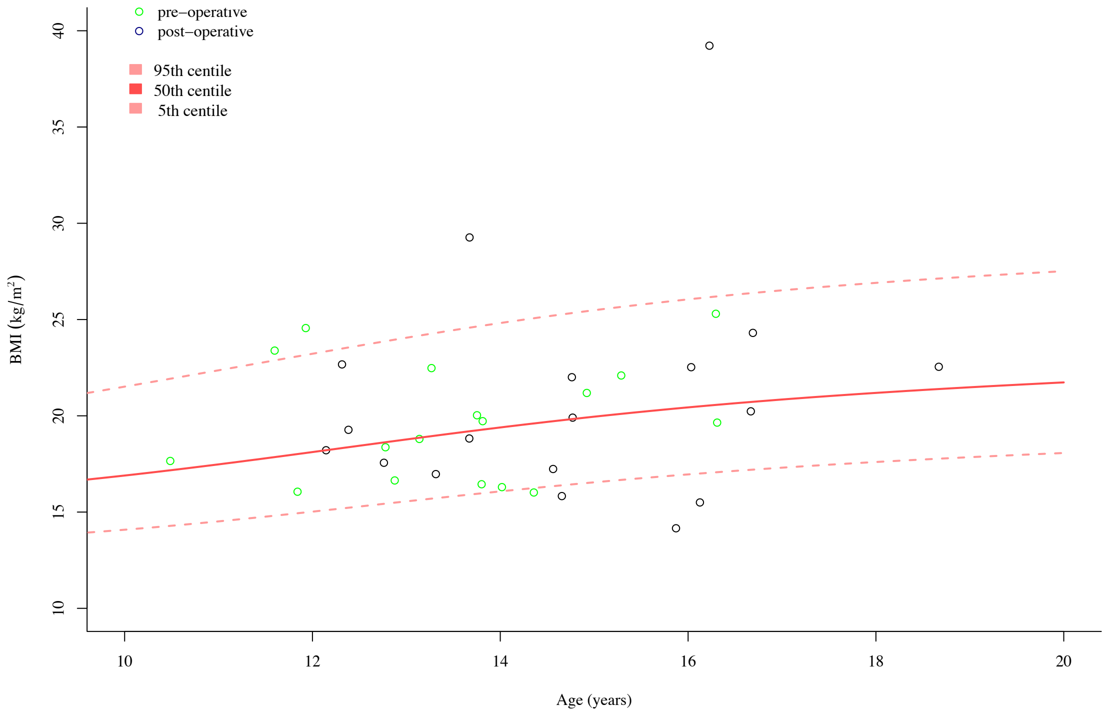


Figure 91: The distribution of BMI (kg/m^2) versus age (years) in females with a main thoracolumbar curve pattern on the UK-WHO growth standard redrawn from [82, 87]. For this plot n = 18.

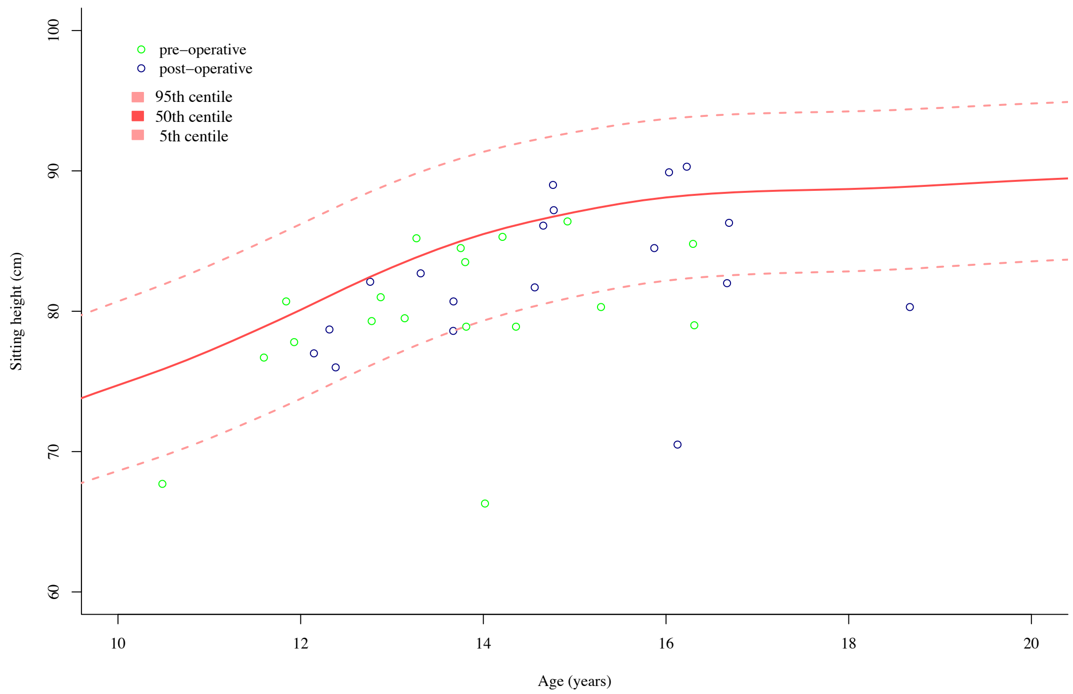


Figure 92: The distribution of sitting height (cm) versus age (years) in females with a main thoracolumbar curve pattern on the Dutch growth standard redrawn from [83, 87]. For this plot n = 18.

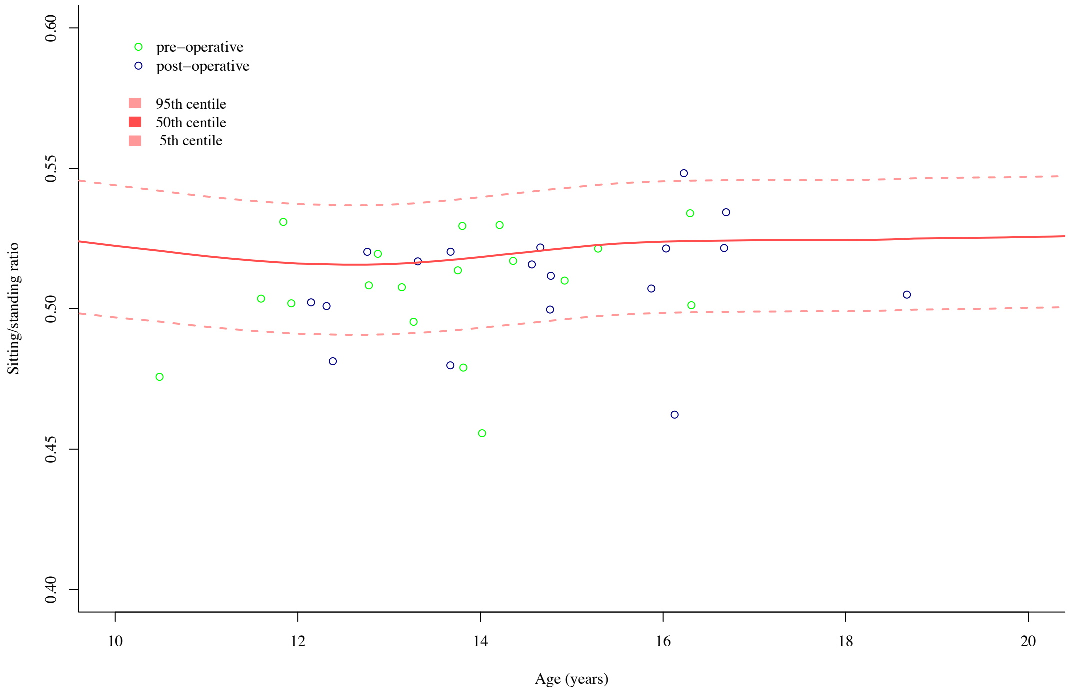


Figure 93: The distribution of the sitting height to standing height ratio versus age (years) in females with a main thoracolumbar curve pattern on the Dutch growth standard redrawn from [83, 87]. For this plot n = 18.

Table 24: The percentage (and number) of pre-operative and post-operative scoliotic data points referenced to the appropriate growth standard for males (n = 20) with a main thoracic curve pattern.

		% under the 5th centile	% under the median	% over the 95th centile
Standing height	Pre-operative	10 (2)	40 (8)	10 (2)
	Post-operative	5 (1)	45 (9)	5 (1)
Weight	Pre-operative	10 (2)	40 (8)	15 (3)
	Post-operative	15 (3)	65 (13)	85 (17)
BMI	Pre-operative	10 (2)	60 (12)	20 (40
	Post-operative	20 (4)	75 (15)	20 (4)
Sitting height	Pre-operative	20 (4)	75 (15)	5 (1)
	Post-operative	20 (4)	75 (15)	0 (0)
Sitting to standing height ratio	Pre-operative	25 (5)	60 (12)	0 (0)
	Post-operative	20 (4)	60 (12)	0 (0)

Table 25: The percentage (and number) of pre-operative and post-operative scoliotic data points referenced to the appropriate growth standard for females (n = 135) with a main thoracic curve pattern.

		% under the 5th centile	% under the median	% over the 95th centile
Standing height	Pre-operative	4 (5)	51 (69)	6 (8)
	Post-operative	4 (5)	43 (58)	7 (9)
Weight	Pre-operative	4 (6)	45 (61)	10 (13)
	Post-operative	9 (12)	51 (69)	11 (15)
BMI	Pre-operative	4 (5)	43 (58)	13 (17)
	Post-operative	14 (19)	52 (70)	11 (15)
Sitting height	Pre-operative	39 (53)	84 (114)	0 (0)
	Post-operative	23 (31)	78 (105)	0 (0)
Sitting to standing height ratio	Pre-operative	21 (28)	73 (98)	2 (3)
	Post-operative	7 (10)	69 (93)	4 (5)

Table 26: The percentage (and number) of pre-operative and post-operative scoliotic data points referenced to the appropriate growth standard for females (n = 18) with a main thoracolumbar curve pattern.

		% under the 5th centile	% under the median	% over the 95th centile
Standing height	Pre-operative	6 (1)	33 (6)	6 (1)
	Post-operative	6 (1)	33 (6)	6 (1)
Weight	Pre-operative	11 (2)	39 (7)	22 (4)
	Post-operative	11 (2)	44 (8)	17 (3)
BMI	Pre-operative	0 (0)	33 (6)	17 (3)
	Post-operative	17 (3)	39 (7)	17 (3)
Sitting height	Pre-operative	33 (6)	89 (16)	0 (0)
	Post-operative	17 (3)	78 (14)	0 (0)
Sitting to standing height ratio	Pre-operative	17 (3)	72 (13)	0 (0)
	Post-operative	17 (3)	67 (12)	6 (1)

7.3.2 Summary - The analysis of standing height, weight, BMI, sitting height and sitting height to standing height ratio on growth standards.

For all of the different measures of growth that can be compared to the UK-WHO and Dutch growth standards, there is a significant change between the pre-operative and post-operative measure other than for BMI in both males and females and sitting to standing height ratio in males. This will be a combination of change that occurs as a direct result of the surgery, and due to any growth that has occurred between the pre-operative and post-operative measures. The distribution of the data from the scoliotic cohort around the UK-WHO and Dutch growth standards is shown.

7.3.3 Results - The analysis of 2D torso points.

The torso points were used for the analysis of the 2D torso points as seen in Figures 27 and 28 and Table 4 in the Methods chapter. Table 27 shows the number of pre-operative and post-operative pairs that were included along with the distributions of ages and sex.

Table 27: The numbers, ages and sex of the pairs of pre-operative and post-operative patients available for the analysis of torso and spinal shape.

Curve pattern	Curve type	Number of pairs	Mean age (years)	SD (years)	Range (years)	Females	Males
main thoracic curve pattern	main thoracic curve	248	13.8	1.5	9.9 to 17.9	213	35
	compensatory thora-columbar curve	129	13.8	1.5	9.9 to 17.7	115	14
main thora-columbar curve pattern	main thora-columbar curve	41	14.4	1.7	10.5 to 17.0	37	4
	compensatory thoracic curve	25	1.8	1.8	10.5 to 16.9	22	3

Torso points in the pre-operative, post-operative and non-scoliotic cohorts. Figures 94 to 98 are ellipses documenting the different parameters of torso shape (ShDiffHt, AxDiffHt, AxDiffOff, WaistDiffHt and WaistDiffOff) for the main thoracic and compensatory thoracolumbar curve pattern. The parameter is referenced against the closest anatomical area of the spine (parameters of the shoulder girdle against the thoracic spine and the waist against the thoracolumbar spine). The pre-operative data is in green, the post-operative in blue and the non-scoliotic in red. The non-scoliotics are only shown as the mean and 95% confidence interval ellipse for clarity. The mean is the solid dot within the confidence interval of the same colour. The box plots with the matching colour represent the same parameter as the colour of the ellipse. The format of the box and whisker plot is defined in Figure 30. Table 28 shows the mean value of the parameter for the pre-operative and non-scoliotic cohorts and the statistical significance of the differences between them. Tables 29 and 30 show the same for the comparisons of the post-operative versus non-scoliotic and the pre-operative versus post-operative cohorts, respectively. This is repeated for the main thoracolumbar curve pattern in Figure 99 to 103 (with the non-scoliotic group in orange, the pre-operative group in dark green and the post-operative group in purple) and Tables 31, 32 and 33.

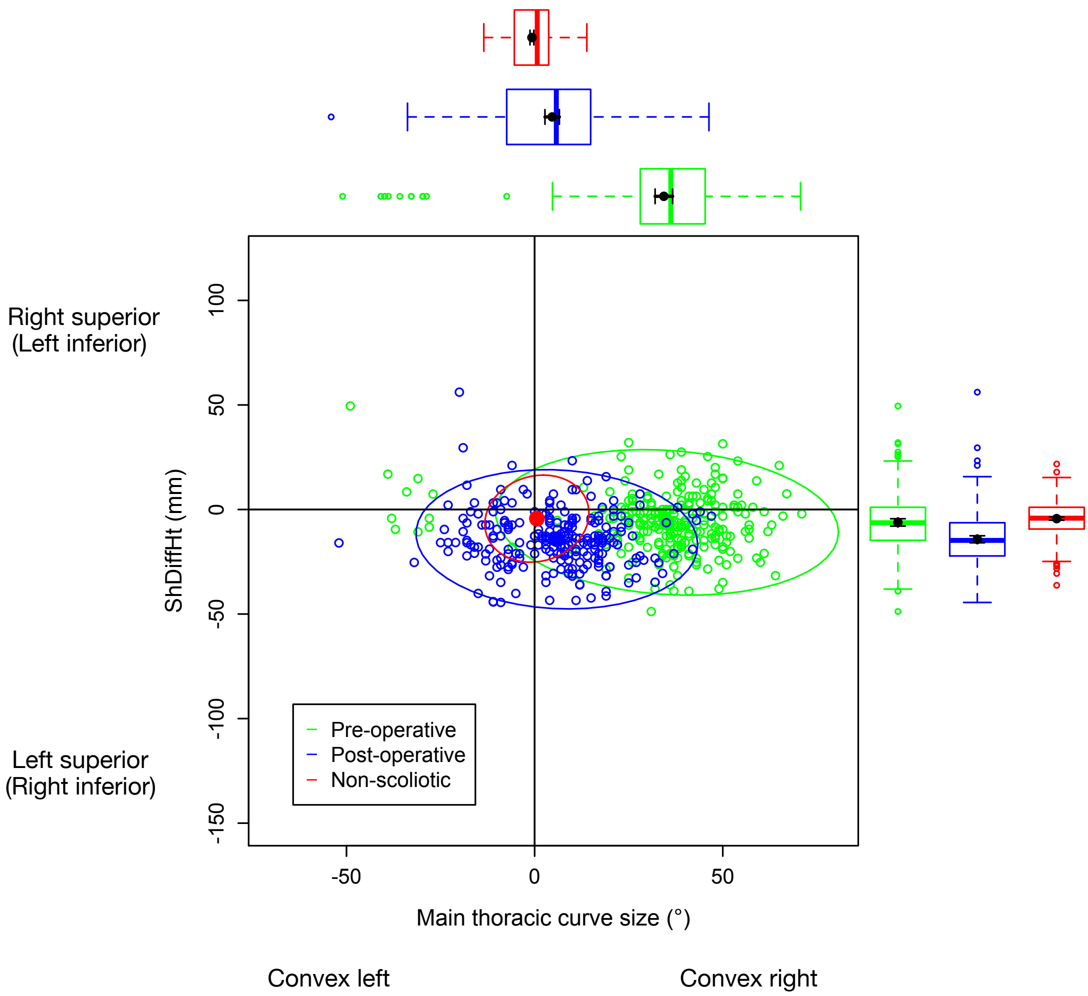


Figure 94: ShDiffHt (mm) versus curve (°) for the main thoracic curve in the main thoracic curve group for the non-scoliotic, pre-operative and post-operative cohorts. The mean values are the solid circles and the 95% confidence ellipses are in the matching colours. ShDiffHt is the difference in vertical height between the shoulder points (see Figures 27 and 28 and Table 4).

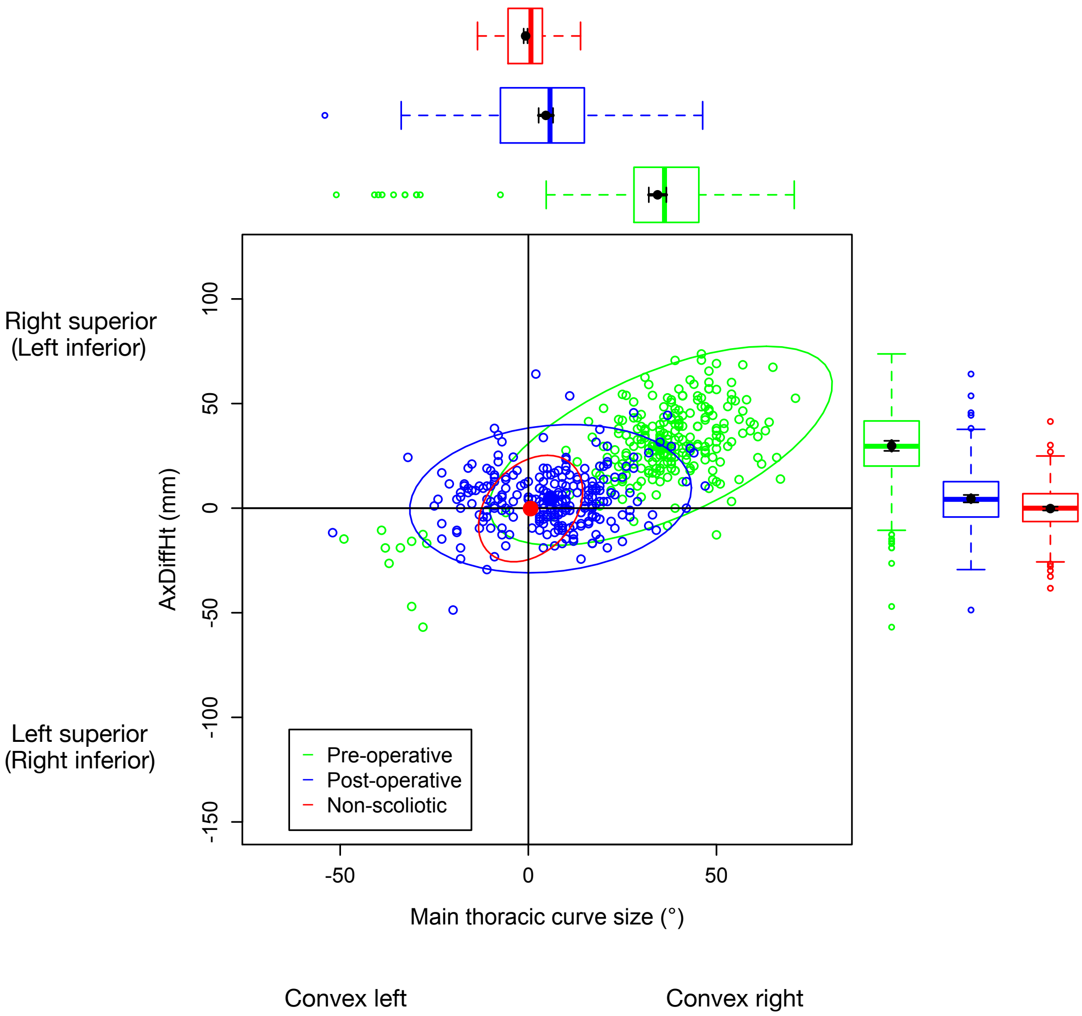


Figure 95: AxDiffHt (mm) versus curve (°) for the main thoracic curve in the main thoracic curve group for the non-scoliotic, pre-operative and post-operative cohorts. The mean values are the solid circles and the 95% confidence ellipses are in the matching colours. AxDiffHt is the difference in vertical height between the axillary points (see Figures 27 and 28 and Table 4).

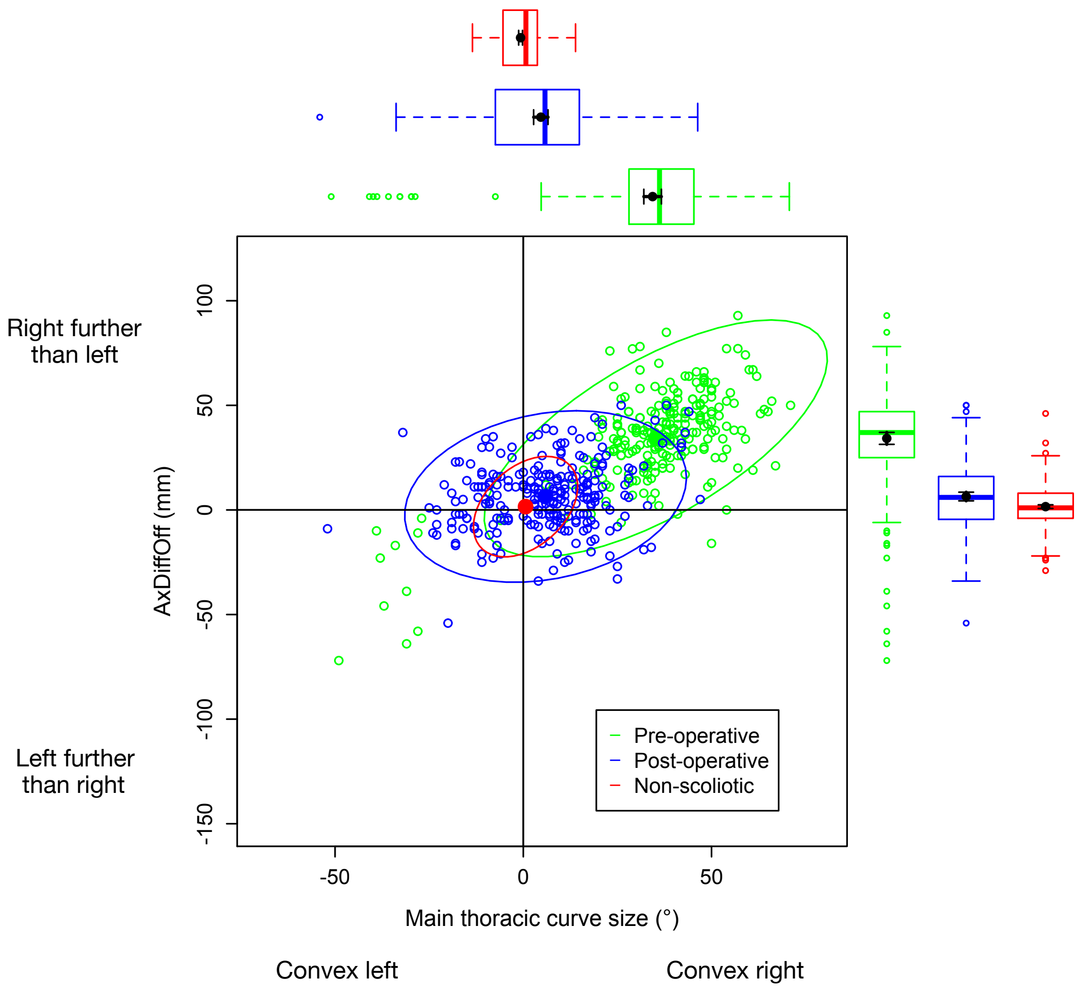


Figure 96: AxDiffOff (mm) versus curve (°) for the main thoracic curve in the main thoracic curve group for the non-scoliotic, pre-operative and post-operative cohorts. The mean values are the solid circles and the 95% confidence ellipses are in the matching colours. AxDiffOff is the difference in horizontal distance between the axillary points and the midline (see Figures 27 and 28 and Table 4).

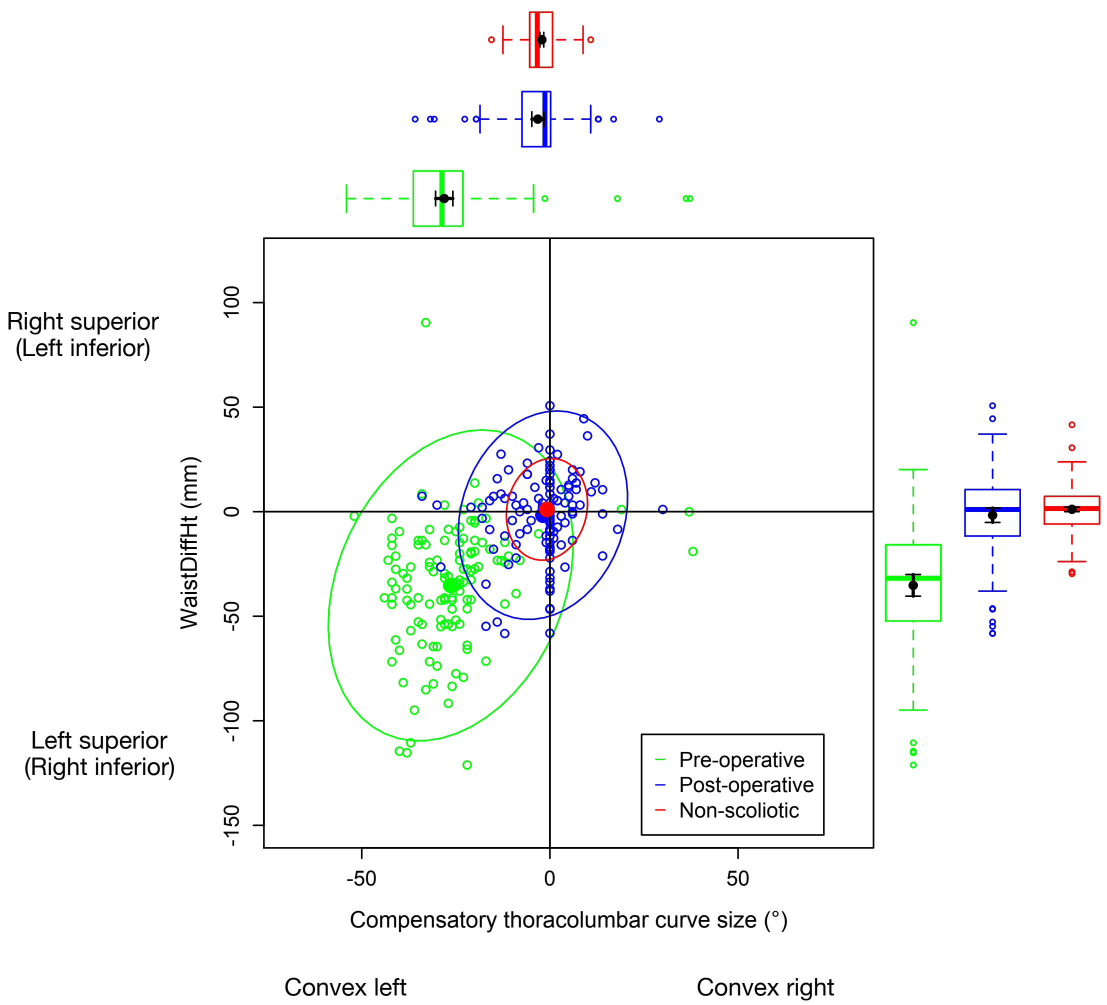


Figure 97: WaistDiffHt (mm) versus curve (°) for the compensatory thoracolumbar curve in the main thoracic curve group for the non-scoliotic, pre-operative and post-operative cohorts. The mean values are the solid circles and the 95% confidence ellipses are in the matching colours. WaistDiffHt is the difference in vertical height between the waist points (see Figures 27 and 28 and Table 4).

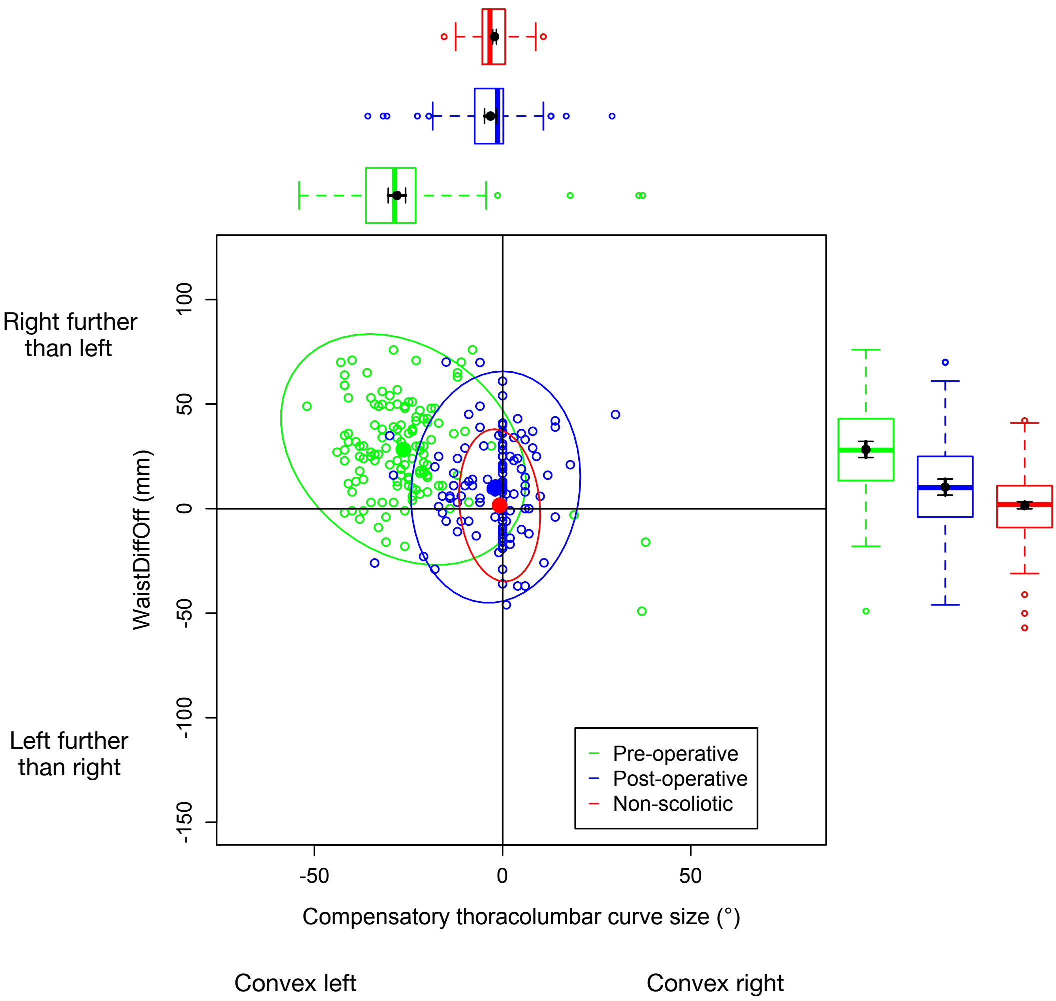


Figure 98: WaistDiffOff (mm) versus curve (°) for the compensatory thoracolumbar curve in the main thoracic curve group for the non-scoliotic, pre-operative and post-operative cohorts. The mean values are the solid circles and the 95% confidence ellipses are in the matching colours. WaistDiffOff is the difference in horizontal distance between the waist points and the midline (see Figures 27 and 28 and Table 4).

Table 28: The mean values (and standard deviation) of the torso points in the pre-operative and non-scoliotic cohorts with the statistical significance in the main thoracic curve pattern. For the definitions of the parameters see Figures 27 and 28 and Table 4.

Parameter	Pre-operative (n = 249)	Non-scoliotic (n = 831)	Statistical significance (using t-test)
Curve size (main curve) (°)	34.9 (18.5)	0.6 (5.6)	$p < 0.001$
ShDiffHt (mm)	-6.2 (14.0)	-4.4 (8.5)	$p = 0.067$
AxDiffHt (mm)	29.7 (19.3)	-0.2 (10.3)	$p < 0.001$
AxDiffOff (mm)	34.0 (22.8)	1.6 (9.8)	$p < 0.001$
WaistDiffHt (mm)	-34.8 (30.2)	1.1 (9.9)	$p < 0.001$
WaistDiffOff (mm)	27.8 (22.0)	1.6 (14.7)	$p < 0.001$

Table 29: The mean values (and standard deviation) of the torso points in the post-operative and non-scoliotic cohorts with the statistical significance in the main thoracic curve pattern. For the definitions of the parameters see Figures 27 and 28 and Table 4.

Parameter	Post-operative (n = 249)	Non-scoliotic (n = 831)	Statistical significance (using t-test)
Curve size (main curve) (°)	6.0 (15.1)	0.6 (5.6)	$p < 0.001$
ShDiffHt (mm)	-14.1 (13.6)	-4.4 (8.5)	$p < 0.001$
AxDiffHt (mm)	4.6 (14.2)	-0.2 (10.3)	$p < 0.001$
AxDiffOff (mm)	6.5 (16.7)	1.6 (9.8)	$p < 0.001$
WaistDiffHt (mm)	1.8 (19.9)	1.1 (9.9)	$p = 0.109$
WaistDiffOff (mm)	10.9 (22.6)	1.6 (14.7)	$p < 0.001$

Table 30: The mean values (and standard deviation) of the torso points in the pre-operative and post-operative cohorts with the statistical significance in the main thoracic curve pattern. For the definitions of the parameters see Figures 27 and 28 and Table 4.

Parameter	Pre-operative (n = 249)	Post-operative (n = 249)	Statistical significance (using t-test)
Curve size (main curve) (°)	34.9 (18.5)	6.0 (15.1)	$p < 0.001$
ShDiffHt (mm)	-6.2 (14.0)	-14.1 (13.6)	$p < 0.001$
AxDiffHt (mm)	29.7 (19.3)	4.6 (14.2)	$p < 0.001$
AxDiffOff (mm)	34.0 (22.8)	6.5 (16.7)	$p < 0.001$
WaistDiffHt (mm)	-34.8 (30.2)	1.8 (19.9)	$p < 0.001$
WaistDiffOff (mm)	27.8 (22.0)	10.9 (22.6)	$p < 0.001$

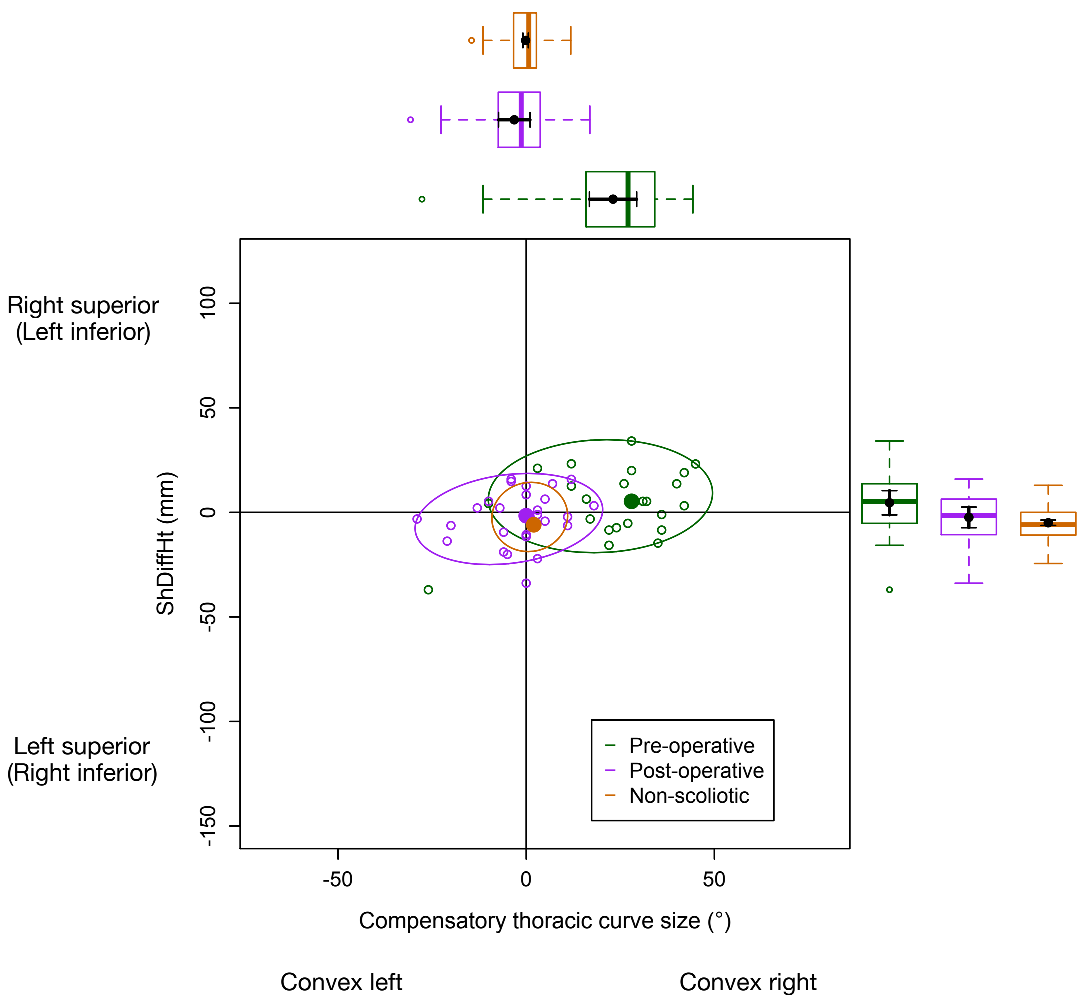


Figure 99: ShDiffHt (mm) versus curve (°) for the compensatory thoracic curve in the main thoracolumbar curve group for the non-scoliotic, pre-operative and post-operative cohorts. The median values are the solid circles and the 95% centiles are in the matching colours. ShDiffHt is the difference in vertical height between the shoulder points (see Figures 27 and 28 and Table 4).

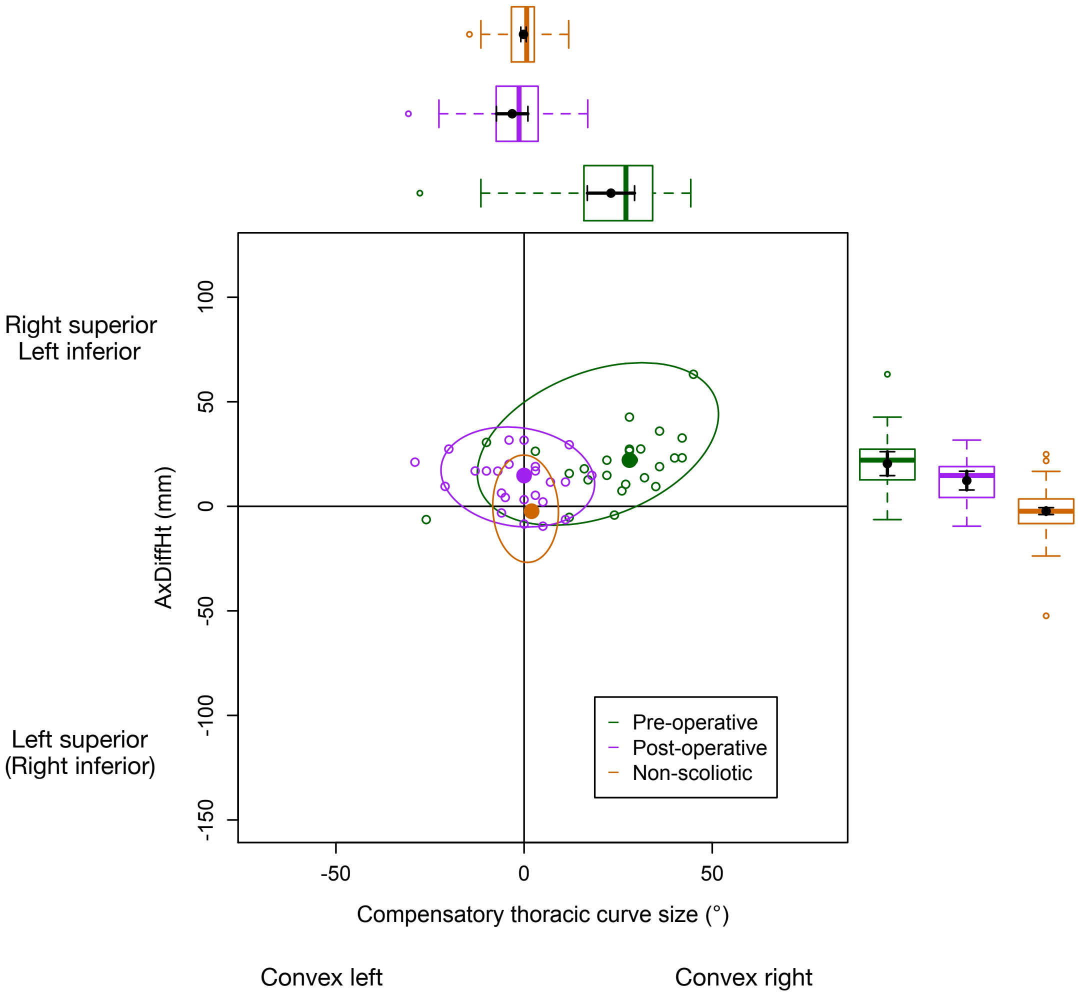


Figure 100: AxDiffHt (mm) versus curve (°) for the compensatory thoracic curve in the main thoracolumbar curve group for the non-scoliotic, pre-operative and post-operative cohorts. The median values are the solid circles and the 95% centiles are in the matching colours. AxDiffHt is the difference in vertical height between the axillary points (see Figures 27 and 28 and Table 4).

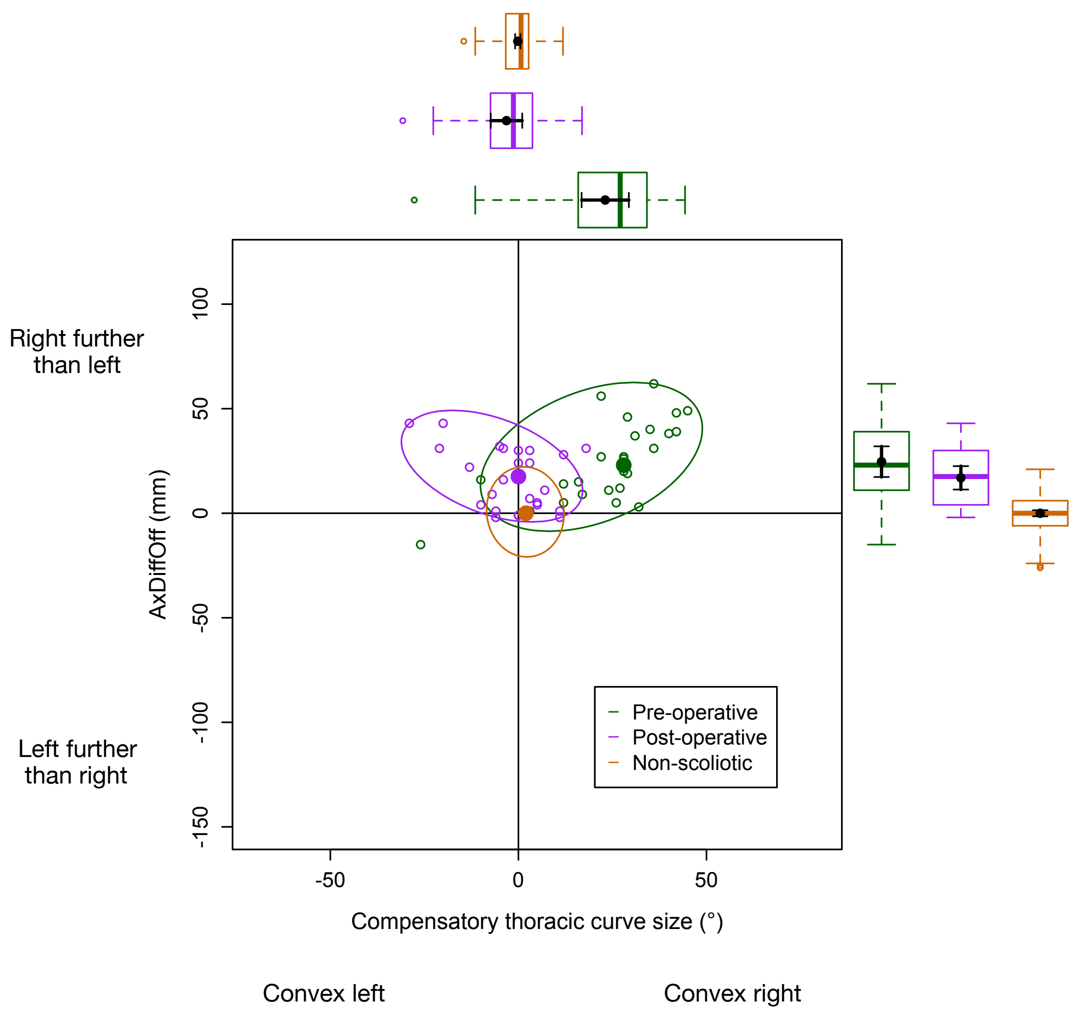


Figure 101: AxDiffOff (mm) versus curve (°) for the compensatory thoracic curve in the main thoracolumbar curve group for the non-scoliotic, pre-operative and post-operative cohorts. The median values are the solid circles and the 95% centiles are in the matching colours. AxDiffOff is the difference in horizontal distance between the axillary points and the midline (see Figures 27 and 28 and Table 4).

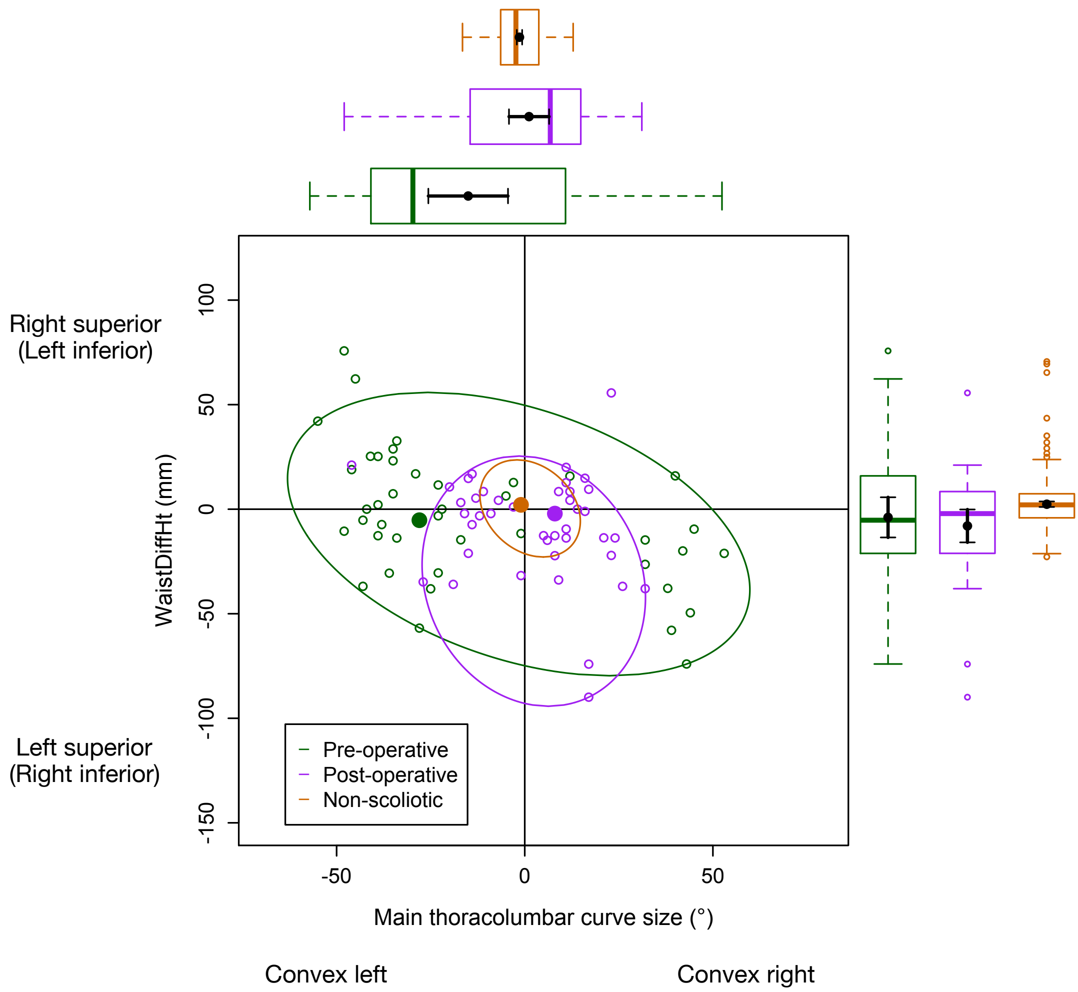


Figure 102: WaistDiffHt (mm) versus curve (°) for the main thoracolumbar curve in the main thoracolumbar curve group for the non-scoliotic, pre-operative and post-operative cohorts. The median values are the solid circles and the 95% centiles are in the matching colours. WaistDiffHt is the difference in vertical height between the waist points (see Figures 27 and 28 and Table 4).

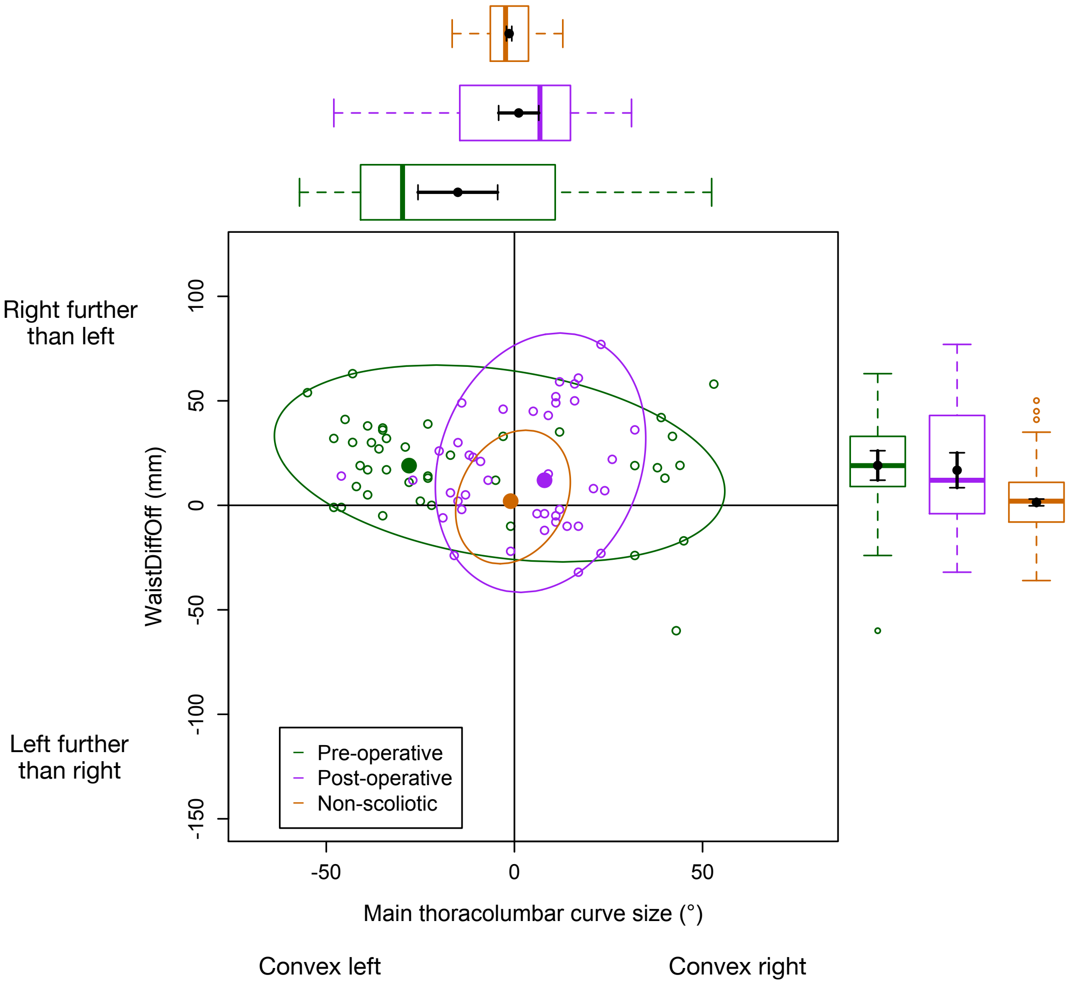


Figure 103: WaistDiffOff (mm) versus curve (°) for the main thoracolumbar curve in the main thoracolumbar curve group for the non-scoliotic, pre-operative and post-operative cohorts. The median values are the solid circles and the 95% centiles are in the matching colours. WaistDiffOff is the difference in horizontal distance between the waist points and the midline (see Figures 27 and 28 and Table 4).

Table 31: The median values (and Q1 and Q3 of the interquartile range) of the torso points in the pre-operative and of the non-scoliotic cohorts with the statistical significance in the main thoracolumbar curve pattern. For the definitions of the parameters see Figures 27 and 28 and Table 4.

Parameter	Pre-operative (n = 40)	Non-scoliotic (n = 831)	Statistical significance (using Wilcoxon sum rank test)
Curve size (main curve) (°)	-28.5 (-39.0 to 16.8)	-1.0 (-5.0 to 5.0)	$p < 0.001$
ShDiffHt (mm)	5.3 (-2.1 to 13.7)	-4.3 (-9.9 to 1.1)	$p = 0.004$
AxDiffHt (mm)	22.2 (13.7 to 27.3)	-2.0 (-7.3 to 5.3)	$p < 0.001$
AxDiffOff (mm)	24.0 (11.0 to 37.0)	1.0 (-5.0 to 7.5)	$p < 0.001$
WaistDiffHt (mm)	-4.2 (-22.5 to 16.2)	2.1 (-4.1 to 7.4)	$p = 0.158$
WaistDiffOff (mm)	19.1 (7.3 to 33.5)	2.0 (-8.0 to 11.0)	$p < 0.001$

Table 32: The median values (and Q1 and Q3 of the interquartile range) of the torso points in post-operative and of the non-scoliotic cohorts with the statistical significance in the main thoracolumbar curve pattern. For the definitions of the parameters see Figures 27 and 28 and Table 4.

Parameter	Post-operative (n = 40)	Non-scoliotic (n = 831)	Statistical significance (using Wilcoxon sum rank test)
Curve size (main curve) (°)	8.0 (-13.3 to 14.5)	-1.0 (-5.0 to 5.0)	$p = 0.095$
ShDiffHt (mm)	-1.1 (-10.6 to 6.3)	-4.3 (-9.9 to 1.1)	$p = 0.305$
AxDiffHt (mm)	14.7 (3.18 to 16.9)	-2.0 (-7.3 to 5.3)	$p < 0.001$
AxDiffOff (mm)	16.0 (4.0 to 30.0)	1.0 (-5.0 to 7.5)	$p < 0.001$
WaistDiffHt (mm)	-2.1 (-21.4 to 8.4)	2.1 (-4.1 to 7.4)	$p = 0.039$
WaistDiffOff (mm)	13.5 (-4.25 to 43.5)	2.0 (-8.0 to 11.0)	$p < 0.001$

Table 33: The median values (and Q1 and Q3 of the interquartile range) of the torso points in the pre-operative and post-operative cohorts with statistical significance in the main thoracolumbar curve pattern. For the definitions of the parameters see Figures 27 and 28 and Table 4.

Parameter	Pre-operative (n = 40)	Post-operative (n = 40)	Statistical significance (using Wilcoxon sum rank test)
Curve size (main curve) (°)	-28.5 (-39.0 to 16.8)	8.0 (-13.3 to 14.5)	p = 0.006
ShDiffHt (mm)	5.3 (-2.1 to 13.7)	-1.1 (-10.6 to 6.3)	p = 0.033
AxDiffHt (mm)	22.2 (13.7 to 27.3)	14.7 (3.18 to 16.9)	p = 0.016
AxDiffOff (mm)	24.0 (11.0 to 37.0)	16.0 (4.0 to 30.0)	p = 0.095
WaistDiffHt (mm)	-4.2 (-22.5 to 16.2)	-2.1 (-21.4 to 8.4)	p = 0.722
WaistDiffOff (mm)	19.1 (7.3 to 33.5)	13.5 (-4.25 to 43.5)	p = 0.45

7.3.4 Summary - The analysis of 2D torso points.

The data shows the variability of the torso points with an increasing scoliotic curve size. The variability in the pre-operative scoliotic cohort is an amplification of the non-scoliotic data. Surgery changes the pre-operative data in to a distribution far more like that seen for the non-scoliotic data but with a greater variability.

7.3.5 Results - Measures of kyphosis and lordosis in pre and post-operative scoliosis cohort.

The values of kyphosis and lordosis are shown for both the main thoracic (Figures 104, 105 and Tables 34, 35 and 36) and main thoracolumbar groups (Figure 106, 107 and Tables 37, 38 and 39) using the same methodology of data ellipses.

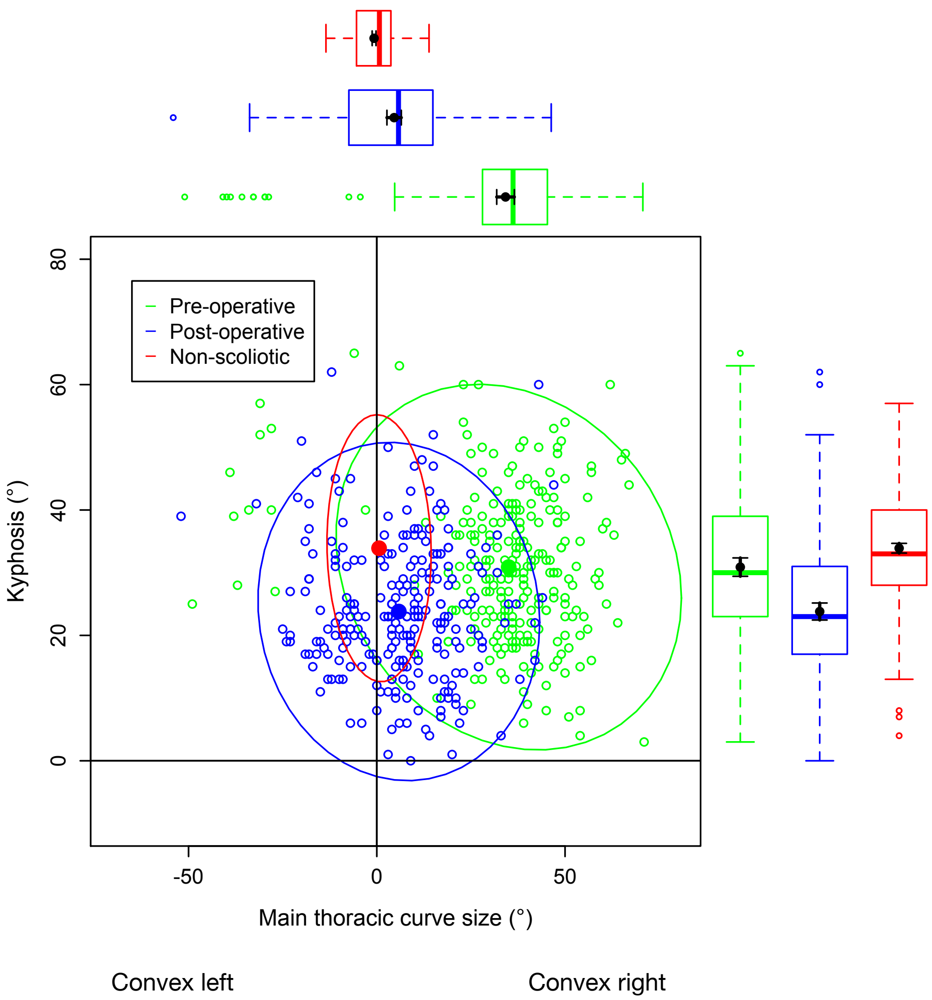


Figure 104: The kyphosis for the main thoracic curve in the main thoracic curve pattern. The mean values are the solid circles and the 95% confidence ellipses are in the matching colours.

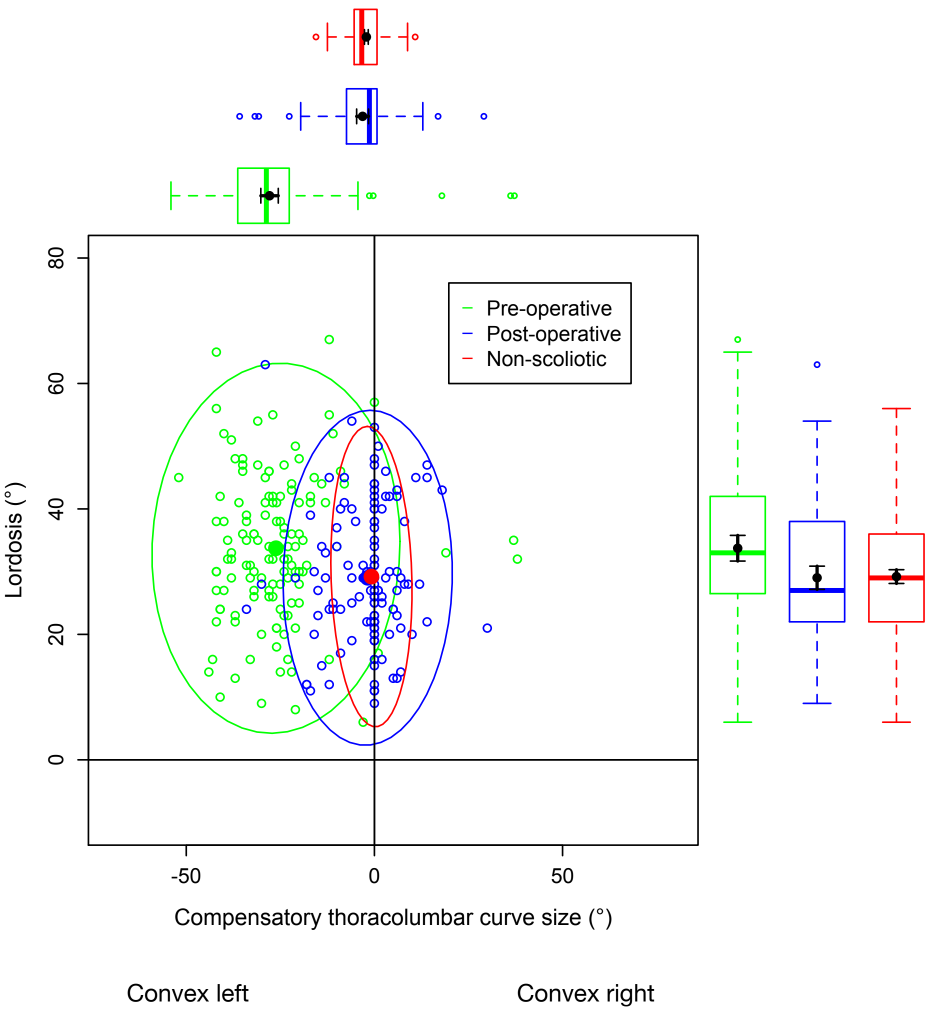


Figure 105: The lordosis for the compensatory thoracolumbar curve in the main thoracic curve pattern. The mean values are the solid circles and the 95% confidence ellipses are in the matching colours.

Table 34: The mean values (and standard deviation) of the kyphosis and lordosis in the pre-operative and of the non-scoliotic cohorts with the statistical significance in the main thoracic curve pattern.

Parameter	Pre-operative (n = 249)	Non-scoliotic (n = 831)	Statistical significance (using t-test)
Kyphosis (°)	30.9 (11.8)	33.9 (8.7)	$p < 0.001$
Lordosis (°)	34.1 (12.5)	29.1 (9.4)	$p < 0.001$

Table 35: The mean values (and standard deviation) of the kyphosis and lordosis in the post-operative and of the non-scoliotic cohorts with the statistical significance in the main thoracic curve pattern.

Parameter	Post-operative (n = 249)	Non-scoliotic (n = 831)	Statistical significance (using t-test)
Kyphosis (°)	23.8 (10.9)	33.9 (8.7)	$p < 0.001$
Lordosis (°)	29.6 (10.7)	29.1 (9.4)	$p = 0.551$

Table 36: The mean values (and standard deviation) of the kyphosis and lordosis in the pre-operative and of the post-operative cohorts with the statistical significance in the main thoracic curve pattern.

Parameter	Pre-operative (n = 249)	Post-operative (n = 249)	Statistical significance (using t-test)
Kyphosis (°)	30.9 (11.8)	23.8 (10.9)	$p < 0.001$
Lordosis (°)	34.1 (12.5)	29.6 (10.7)	$p < 0.001$

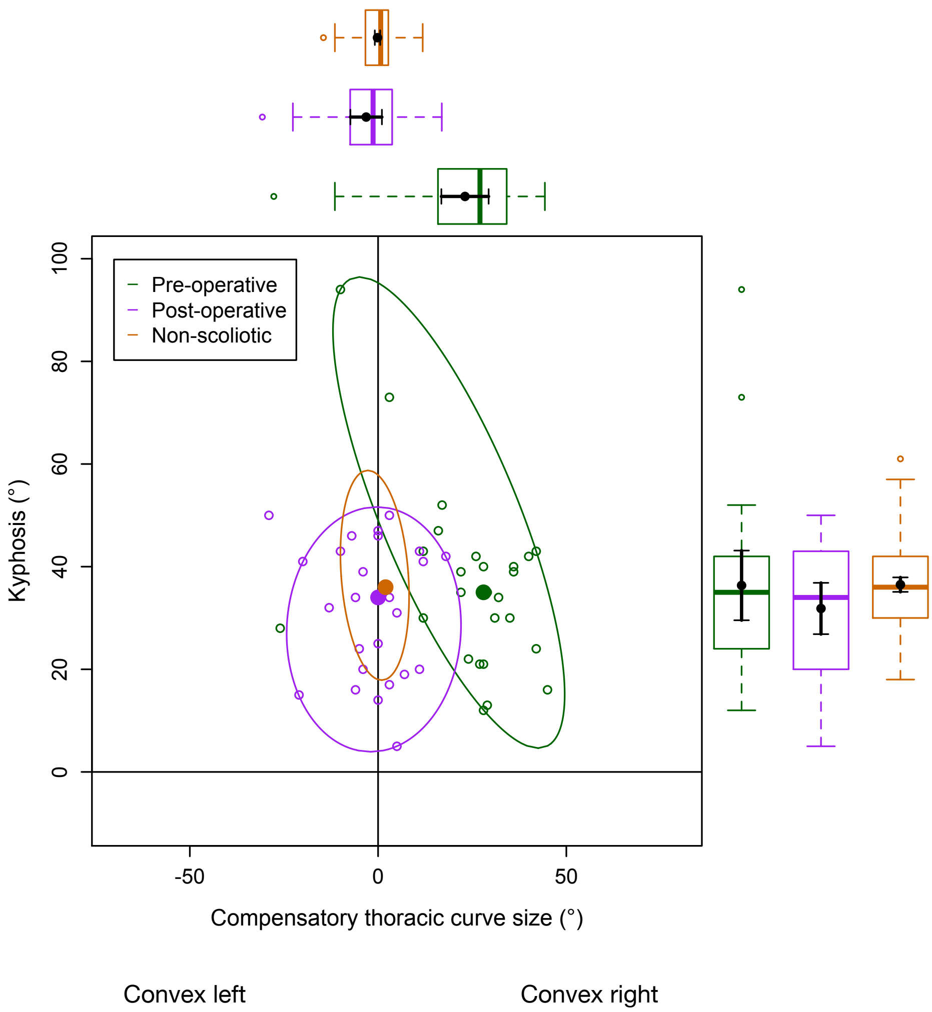


Figure 106: The kyphosis for the compensatory thoracic curve in the main thoracolumbar curve pattern. The median values are the solid circles and the 95% centiles are in the matching colours.

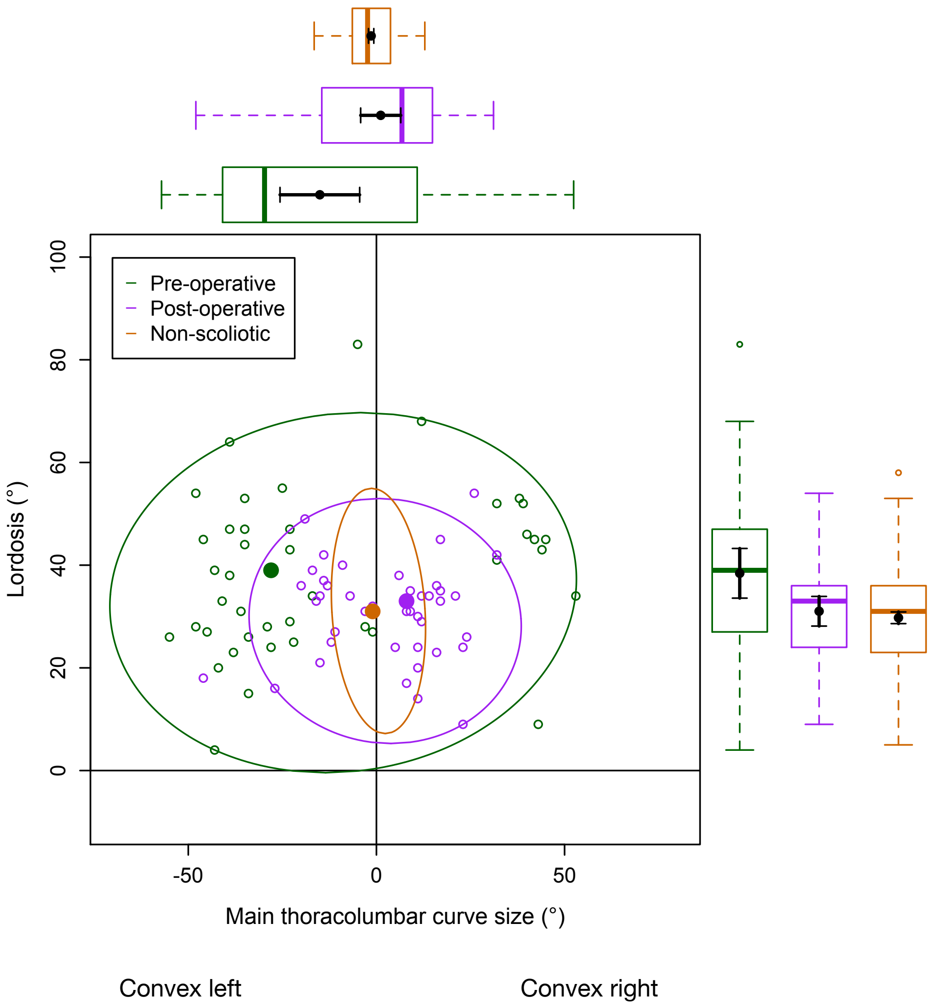


Figure 107: The lordosis in the main thoracolumbar curve in the main thoracolumbar curve pattern. The median values are the solid circles and the 95% centiles are in the matching colours.

Table 37: The median values (and Q1 and Q3 of the interquartile range) of the kyphosis and lordosis in the pre-operative and of the non-scoliotic cohorts with the statistical significance in the main thoracolumbar curve pattern.

Parameter	Pre-operative (n = 40)	Non-scoliotic (n = 831)	Statistical significance (using Wilcoxon sum rank test)
Kyphosis (°)	34.5 (25.0 to 42.0)	33.0 (28.0 to 40.0)	$p < 0.001$
Lordosis (°)	38.5 (27.0 to 47.0)	29.0 (23.0 to 36.0)	$p < 0.001$

Table 38: The median values (and Q1 and Q3 of the interquartile range) of the kyphosis and lordosis in the post-operative and of the non-scoliotic cohorts with the statistical significance in the main thoracolumbar curve pattern.

Parameter	Post-operative (n = 40)	Non-scoliotic (n = 831)	Statistical significance (using Wilcoxon sum rank test)
Kyphosis (°)	30.0 (20.8 to 41.3)	33.0 (28.0 to 40.0)	$p < 0.001$
Lordosis (°)	32.5 (24.8 to 36.0)	29.0 (23.0 to 36.0)	$p = 0.776$

Table 39: The median values (and Q1 and Q3 of the interquartile range) of the kyphosis and lordosis in the pre-operative and of the post-operative cohorts with the statistical significance in the main thoracolumbar curve pattern.

Parameter	Pre-operative (n = 40)	Post-operative (n = 40)	Statistical significance (using Wilcoxon sum rank test)
Kyphosis (°)	34.5 (25.0 to 42.0)	30.0 (20.8 to 41.3)	$p < 0.001$
Lordosis (°)	38.5 (27.0 to 47.0)	32.5 (24.8 to 36.0)	$p < 0.001$

7.3.6 Summary - Measures of kyphosis and lordosis in pre and post-operative scoliosis cohort.

Both kyphosis and lordosis are reduced in magnitude by surgery between the pre-operative and post-operative values. However, whilst there is a statistical difference to that of the non-scoliotic cohort, the difference is smaller than the measurement error of the Cobb angle [208]; the change would therefore not be classed as clinically significant.

7.3.7 Results - Most prominent points over the back.

The same methodology of data ellipses is used for the most prominent points over the back. Figures 108, 109 and 110 and Tables 40, 41 and 42 document the relationship between the most prominent points over the back to the thoracic curve in the main thoracic curve pattern and Figures 111, 112 and 113 and Tables 43, 44 and 45 the thoracic curve in the main thoracolumbar curve pattern. Note that these are referenced to only the thoracic curve (main thoracic or compensatory thoracic) as this is the curve anatomically the closest to the most prominent points.

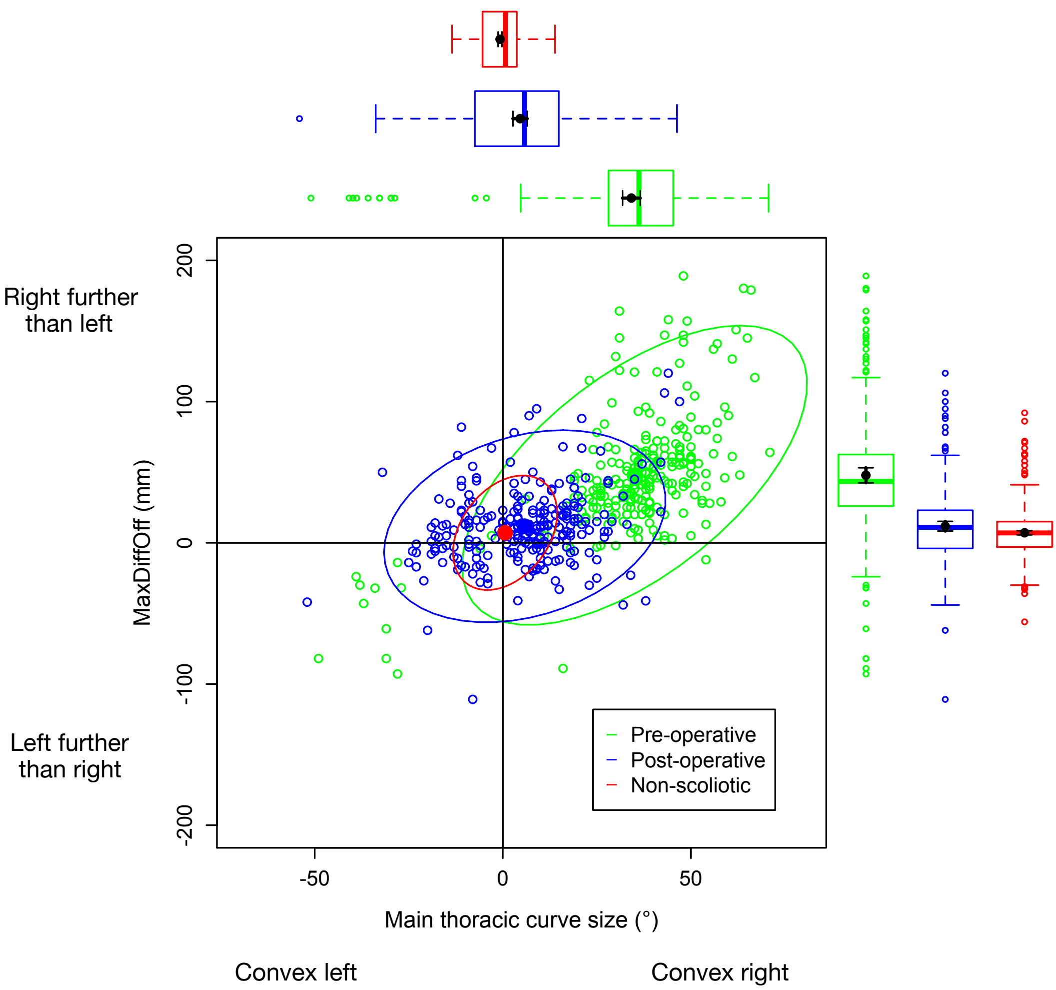


Figure 108: MaxDiffOff (mm) versus main thoracic curve in the main thoracic curve pattern (°). The mean values are the solid circles and the 95% confidence ellipses are in the matching colours. MaxDiffOff is the difference in horizontal distance from the midline of the most prominent points (x coordinates) (see Figure 29 and Table 5.)

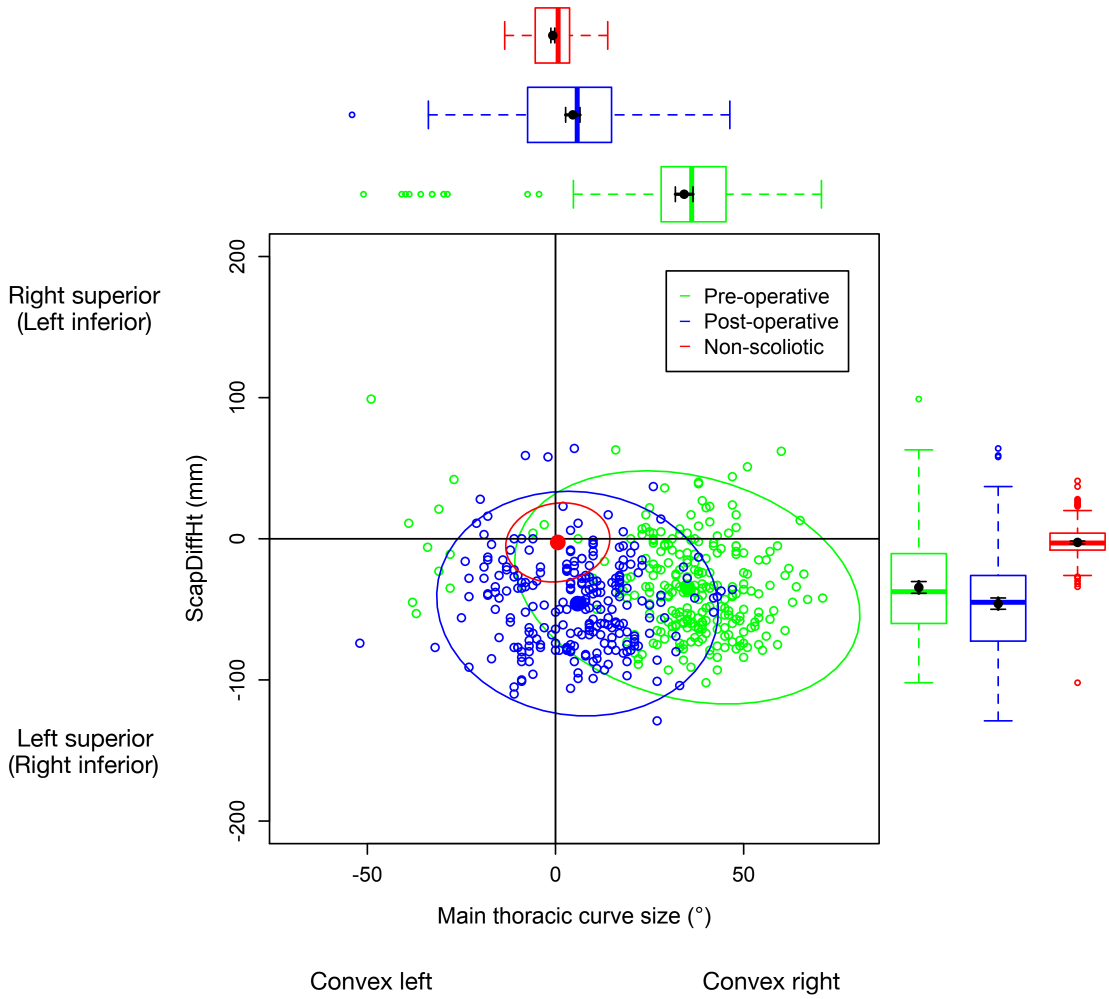


Figure 109: ScapDiffHt (mm) versus main thoracic curve in the main thoracic curve pattern (°). The mean values are the solid circles and the 95% confidence ellipses are in the matching colours. ScapDiffHt is the difference in vertical height of the most prominent points (y coordinates) (see Figure 29 and Table 5.)

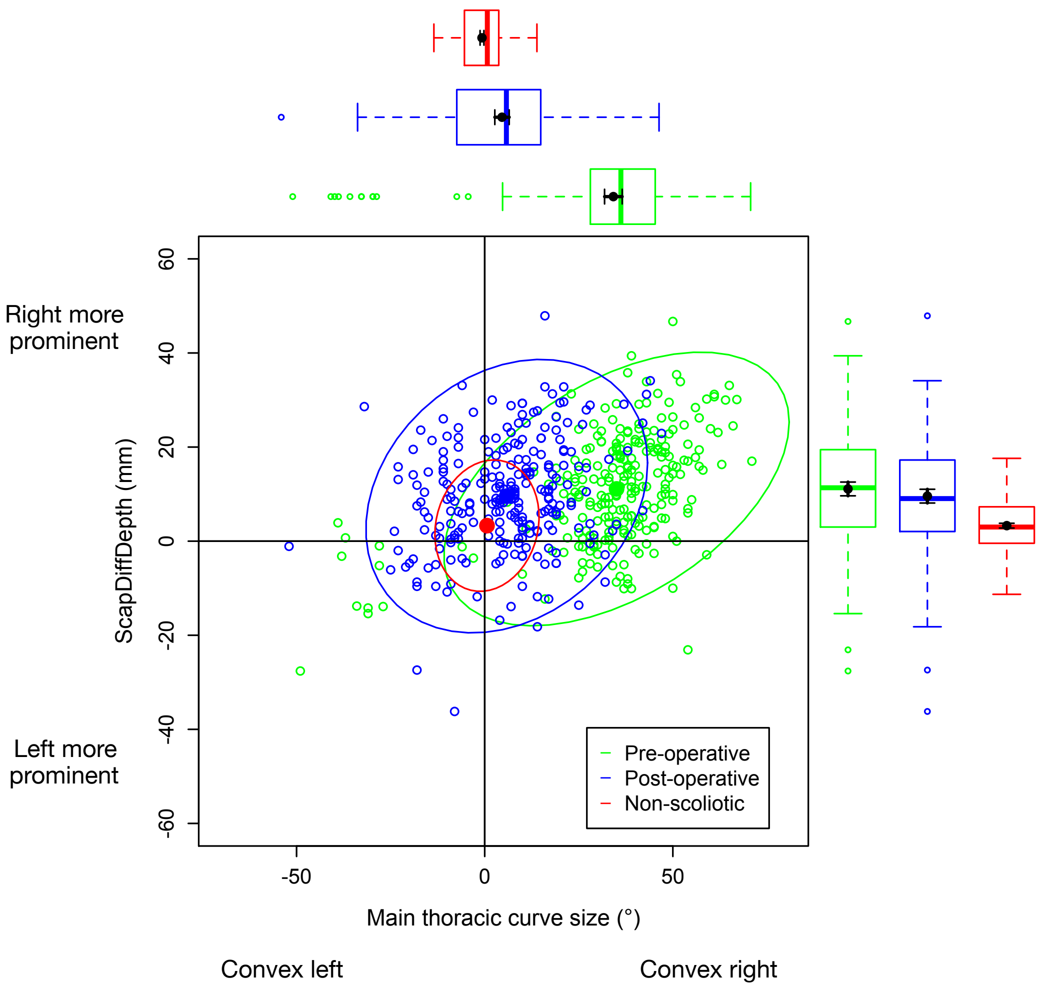


Figure 110: ScapDiffDepth (mm) versus main thoracic curve in the main thoracic curve pattern (°). The mean values are the solid circles and the 95% confidence ellipses are in the matching colours. ScapDiffDepth is the difference in prominence of the most prominent points (z coordinates) (see Figure 29 and Table 5.)

Table 40: The mean values (and standard deviation) of the most prominent points in the pre-operative and of the non-scoliotic cohorts with the statistical significance in the main thoracic curve pattern. For the definitions of the parameters see Figure 29 and Table 5.

Parameter	Pre-operative (n = 249)	Non-scoliotic (n = 831)	Statistical significance (using t-test)
MaxDiffOff (mm)	47.7 (42.7)	7.1 (16.5)	$p < 0.001$
ScapDiffHt (mm)	-35.0 (33.6)	-2.6 (11.4)	$p < 0.001$
ScapDiffDepth (mm)	11.0 (11.8)	3.3 (5.7)	$p < 0.001$

Table 41: The mean values (and standard deviation) of the most prominent points in post-operative and of the non-scoliotic cohorts with the statistical significance in the main thoracic curve pattern. For the definitions of the parameters see Figure 29 and Table 5.

Parameter	Post-operative (n = 249)	Non-scoliotic (n = 831)	Statistical significance (using t-test)
MaxDiffOff (mm)	11.4 (27.6)	7.1 (16.5)	$p < 0.001$
ScapDiffHt (mm)	-45.5 (32.8)	-2.6 (11.4)	$p < 0.001$
ScapDiffDepth (mm)	9.5 (11.8)	3.3 (5.7)	$p = 0.022$

Table 42: The mean values (and standard deviation) of the most prominent points in pre-operative and of the post-operative cohorts with the statistical significance in the main thoracic curve pattern. For the definitions of the parameters see Figure 29 and Table 5.

Parameter	Pre-operative (n = 249)	Post-operative (n = 249)	Statistical significance (using t-test)
MaxDiffOff (mm)	47.7 (42.7)	11.4 (27.6)	$p < 0.001$
ScapDiffHt (mm)	-35.0 (33.6)	-45.5 (32.8)	$p < 0.001$
ScapDiffDepth (mm)	11.0 (11.8)	9.5 (11.8)	$p = 0.153$

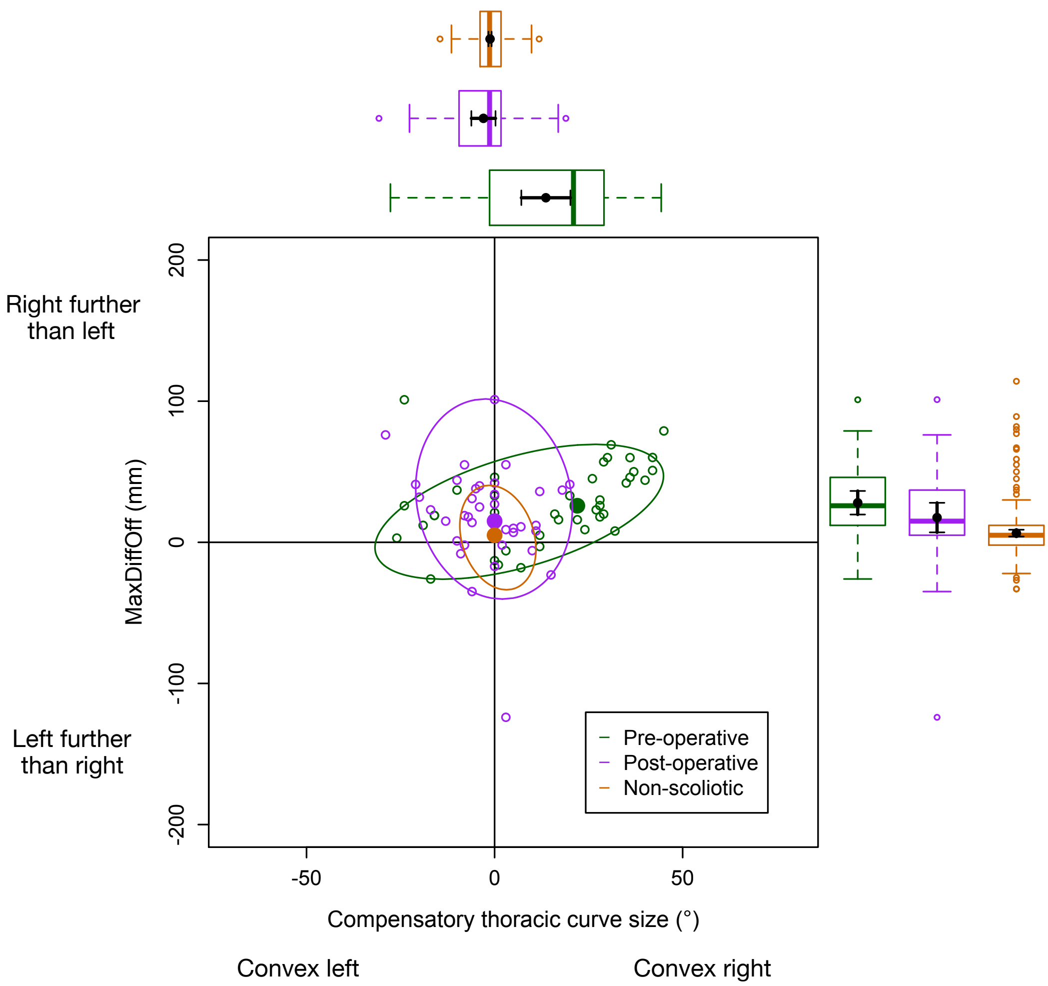


Figure 111: MaxDiffOff (mm) versus compensatory thoracic curve in the main thoracolumbar curve pattern (°). The median values are the solid circles and the 95% centiles are in the matching colours. MaxDiffOff is the difference in horizontal distance from the midline of the most prominent points (z coordinates) (see Figure 29 and Table 5.)

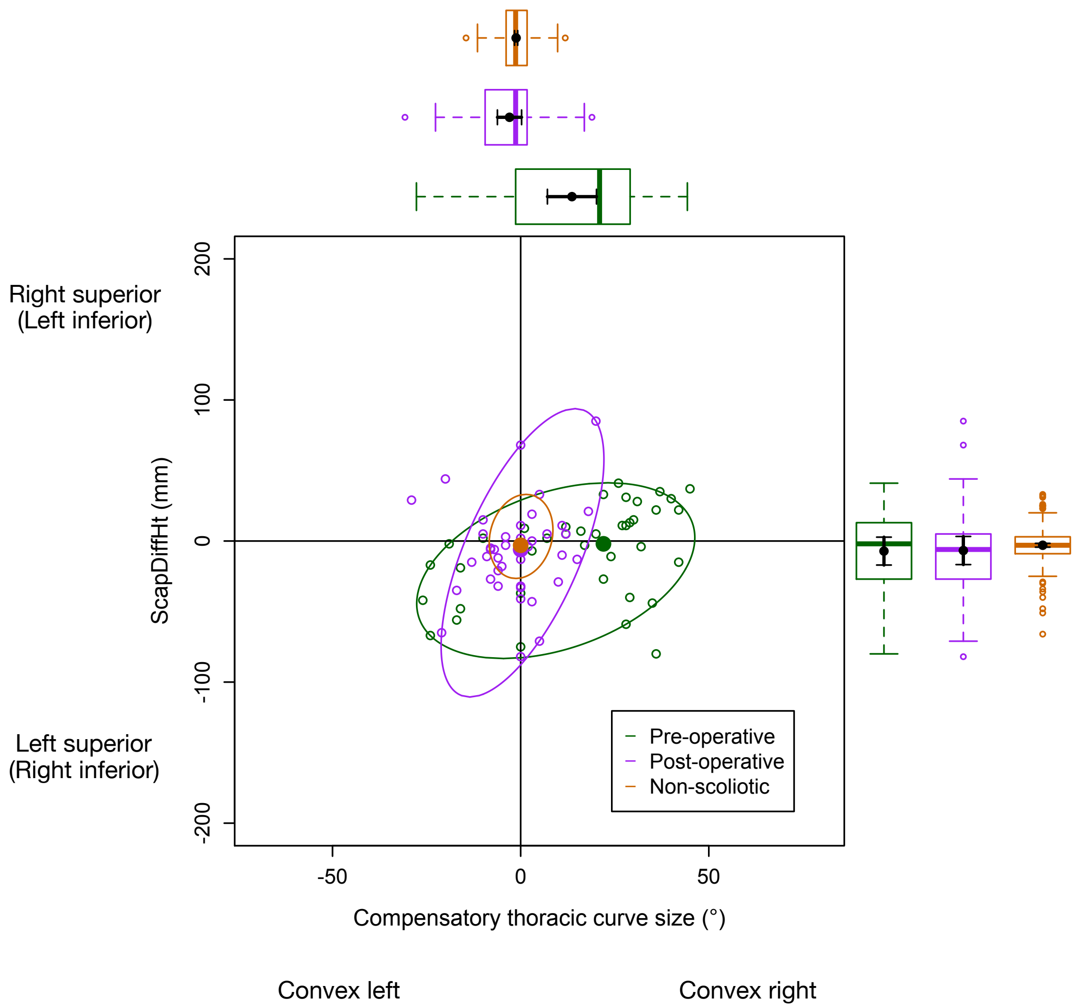


Figure 112: ScapDiffHt (mm) versus compensatory thoracic curve in the main thoracolumbar curve pattern (°). The median values are the solid circles and the 95% centiles are in the matching colours. ScapDiffHt is the difference in vertical height of the most prominent points (y coordinates) (see Figure 29 and Table 5.)

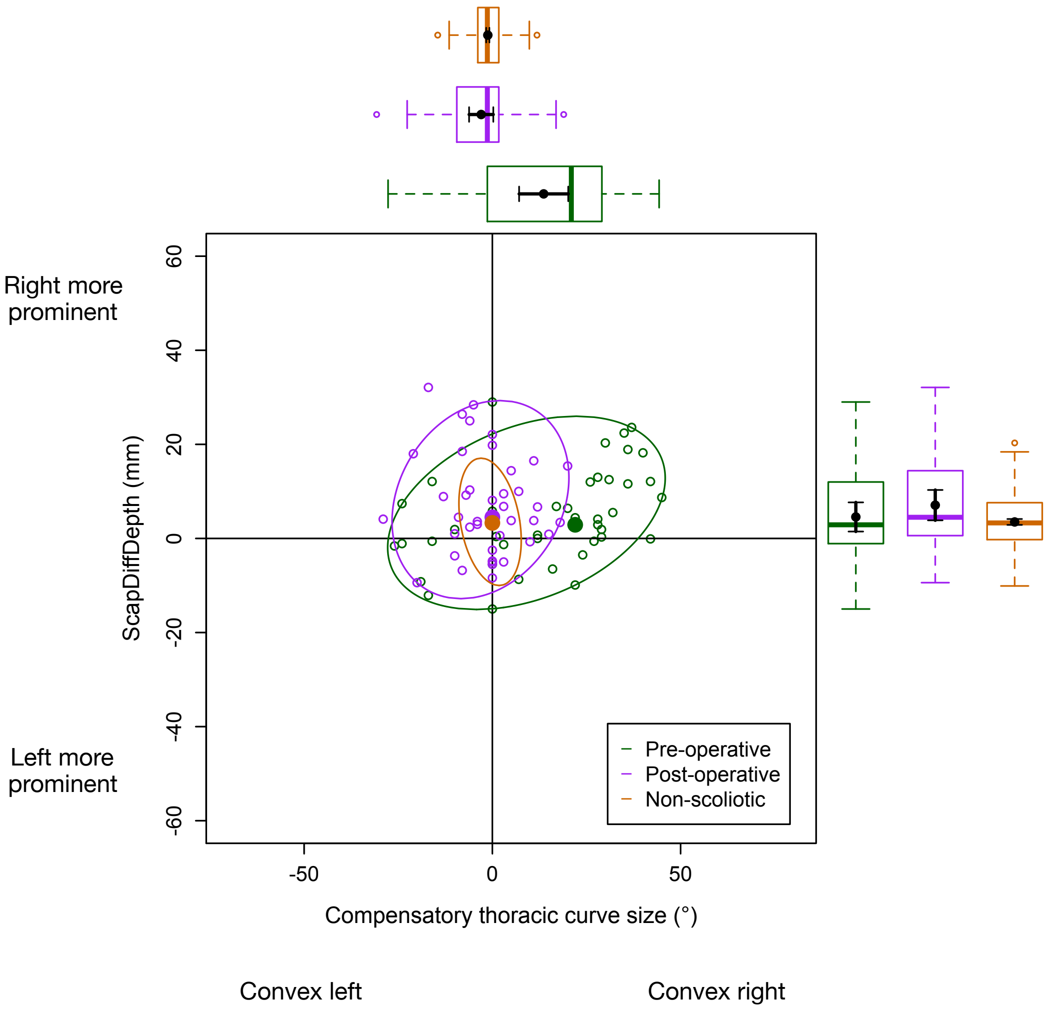


Figure 113: ScapDiffDepth (mm) versus compensatory thoracic curve in the main thoracolumbar curve pattern (°). The median values are the solid circles and the 95% centiles are in the matching colours. ScapDiffDepth is the difference in prominence of the most prominent points (z coordinates) (see Figure 29 and Table 5.)

Table 43: The median values (and Q1 and Q3 of the interquartile range) of the most prominent points in the pre-operative and of the non-scoliotic cohorts with the statistical significance in the main thoracolumbar curve pattern. For the definitions of the parameters see Figure 29 and Table 5.

Parameter	Pre-operative (n = 40)	Non-scoliotic (n = 831)	Statistical significance (using Wilcoxon sum rank test)
MaxDiffOff (mm)	51.1 (34.0 to 66.1)	5.0 (-2.0 to 12.0)	$p < 0.001$
ScapDiffHt (mm)	-40.0 (-61.0 to -4.0)	-3.0 (-9.0 to 3.0)	$p < 0.001$
ScapDiffDepth (mm)	14.3 (6.7 to 21.6)	3.3 (-0.3 to 7.6)	$p < 0.001$

Table 44: The median values (and Q1 and Q3 of the interquartile range) of the most prominent points in the post-operative and of the non-scoliotic cohorts with the statistical significance in the main thoracolumbar curve pattern. For the definitions of the parameters see Figure 29 and Table 5.

Parameter	Pre-operative (n = 40)	Non-scoliotic (n = 831)	Statistical significance (using Wilcoxon sum rank test)
MaxDiffOff (mm)	14.0 (3.0 to 31.0)	5.0 (-2.0 to 12.0)	$p < 0.001$
ScapDiffHt (mm)	-42.0 (-74.0 to -21.0)	-3.0 (-9.0 to 3.0)	$p < 0.001$
ScapDiffDepth (mm)	11.7 (4.7 to 34.1)	3.3 (-0.3 to 7.6)	$p < 0.001$

Table 45: The median values (and Q1 and Q3 of the interquartile range) of the most prominent points in pre-operative and of the post-operative cohorts with the statistical significance in the main thoracolumbar curve pattern. For the definitions of the parameters see Figure 29 and Table 5.

Parameter	Pre-operative (n = 40)	Post-operative (n = 40)	Statistical significance (using Wilcoxon sum rank test)
MaxDiffOff (mm)	51.1 (34.0 to 66.1)	14.0 (3.0 to 31.0)	$p < 0.001$
ScapDiffHt (mm)	-40.0 (-61.0 to -4.0)	-42.0 (-74.0 to -21.0)	$p = 0.001$
ScapDiffDepth (mm)	14.3 (6.7 to 21.6)	11.7 (4.7 to 34.1)	$p = 0.009$

7.3.8 Results - Volumetric asymmetry (VA).

Figures 114 to 117 show the VA for the main thoracic curve pattern. Figures 118 to 121 show the VA for the main thoracolumbar curve pattern. The absolute values of VA between the pre-operative and post-operative situation and also show the amount of change (the pre-operative minus the post-operative values) that occurs between pre-operative and post-operative in each of the VA subgroups as described in the normal child (Chapter 6 - right only, left only and right and left together). Statistical analysis of the size of the change was performed using the Wilcoxon signed rank test as seen in Tables 46 and 47.

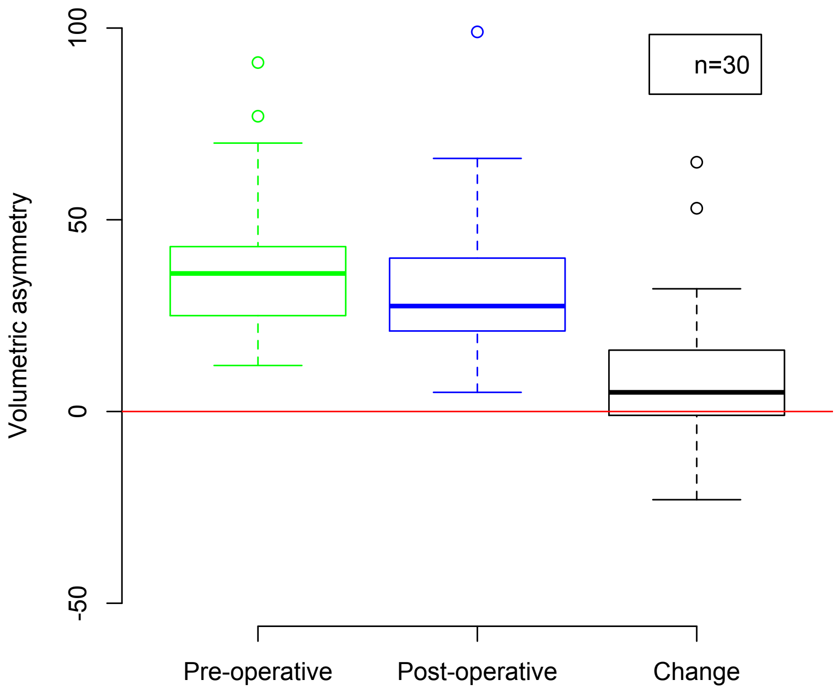


Figure 114: The VA for the pre-operative to post-operative cohorts in the VA on the right only subgroup for the main thoracic curve pattern. VA is a unit-less parameter that describes the difference in 3D volume between each side of the torso (see Figure 26).

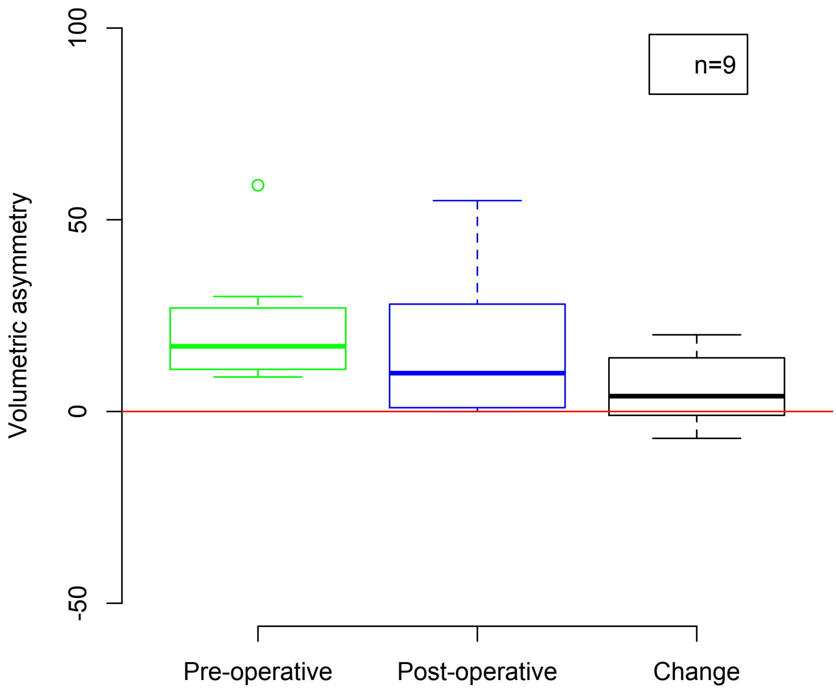


Figure 115: The VA for the pre-operative to post-operative cohorts in the VA in the left only subgroup for the main thoracic curve pattern. VA is a unit-less parameter that describes the difference in 3D volume between each side of the torso (see Figure 26).

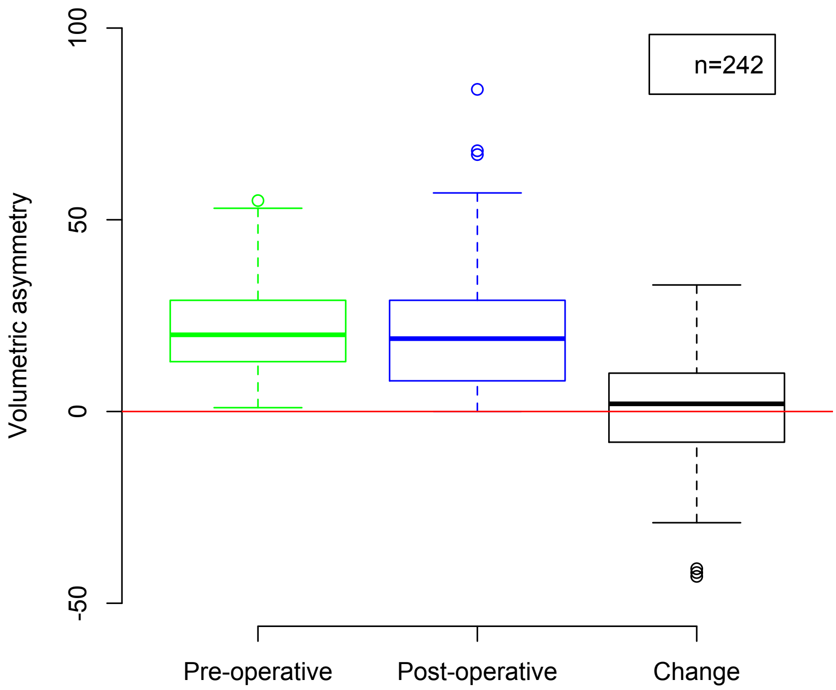


Figure 116: The VA on the right for the pre-operative to post-operative cohorts with both left and right sided VA for the main thoracic curve pattern. VA is a unit-less parameter that describes the difference in 3D volume between each side of the torso (see Figure 26).

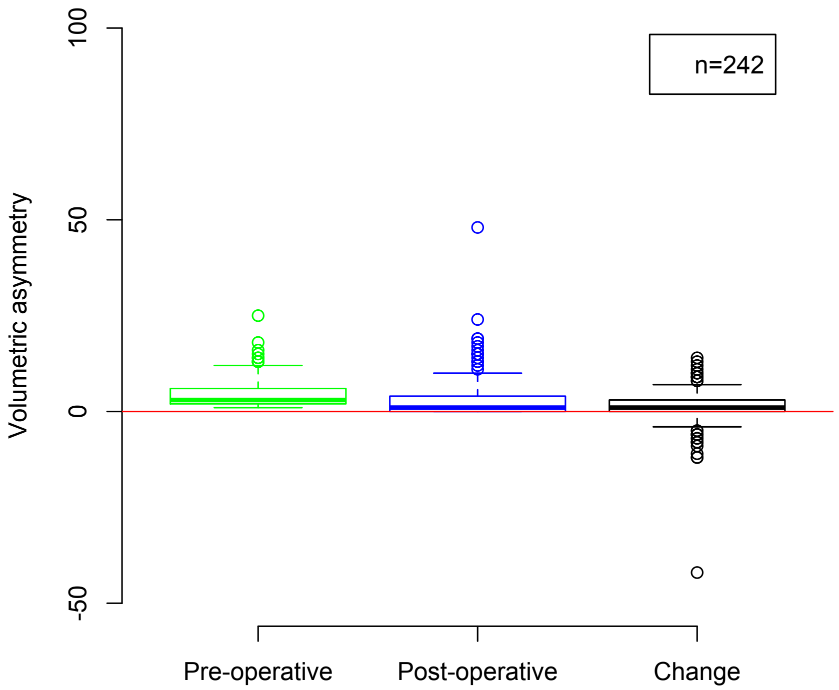


Figure 117: The VA on the left for the pre-operative to post-operative cohorts with both left and right sided VA for the main thoracic curve pattern. VA is a unit-less parameter that describes the difference in 3D volume between each side of the torso (see Figure 26).

Table 46: The statistical significance of the change between the size of pre-operative to post-operative VA in the main thoracic curve pattern. VA is a unit-less parameter that describes the difference in 3D volume between each side of the torso (see Figure 26).

Parameter	Statistical significance	n
VA pre-operative to post-operative for VA on the right only subgroup	$p = 0.017$	30
VA pre-operative to post-operative for VA on the left only subgroup	$p = 0.173$	9
VA pre-operative to post-operative for VA on both the right and left subgroup showing only the right	$p = 0.134$	242
VA pre-operative to post-operative for VA on both the right and left subgroup showing only the left	$p < 0.001$	242

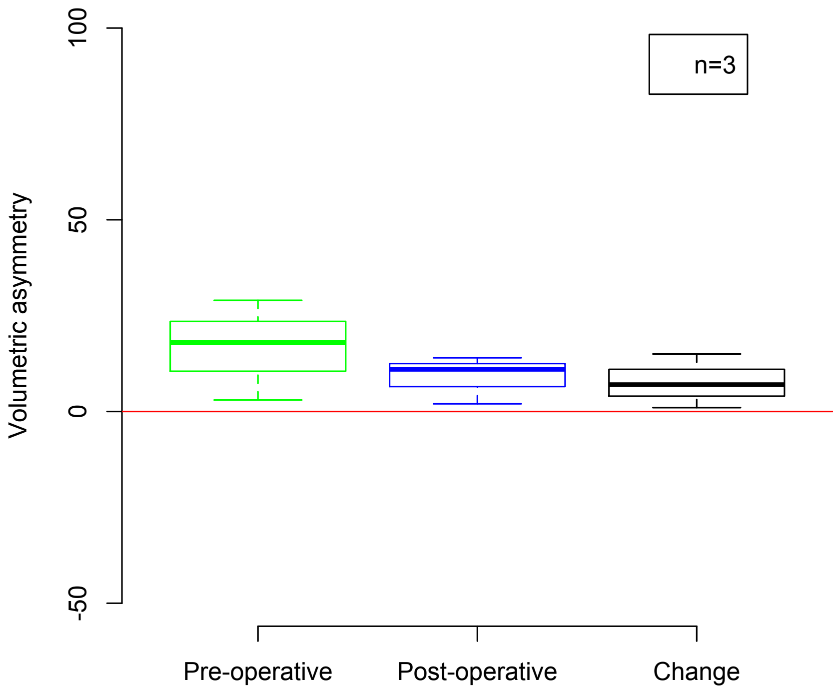


Figure 118: The VA for the pre-operative to post-operative cohorts in the VA on the right only subgroup for the main thoracolumbar curve pattern. VA is a unit-less parameter that describes the difference in 3D volume between each side of the torso (see Figure 26).

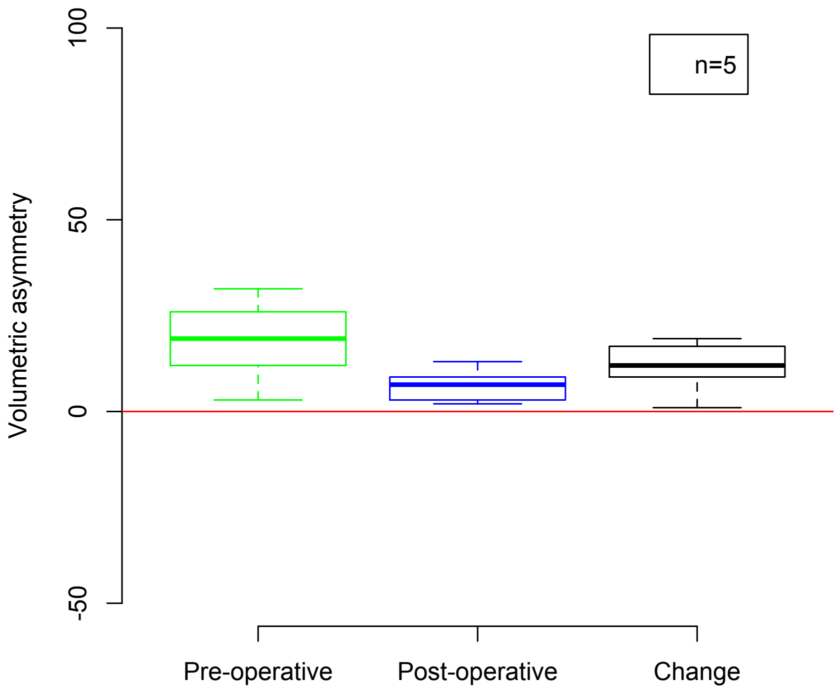


Figure 119: The VA for the pre-operative to post-operative cohorts in the VA on the left only subgroup for main thoracolumbar curve pattern. VA is a unit-less parameter that describes the difference in 3D volume between each side of the torso (see Figure 26).

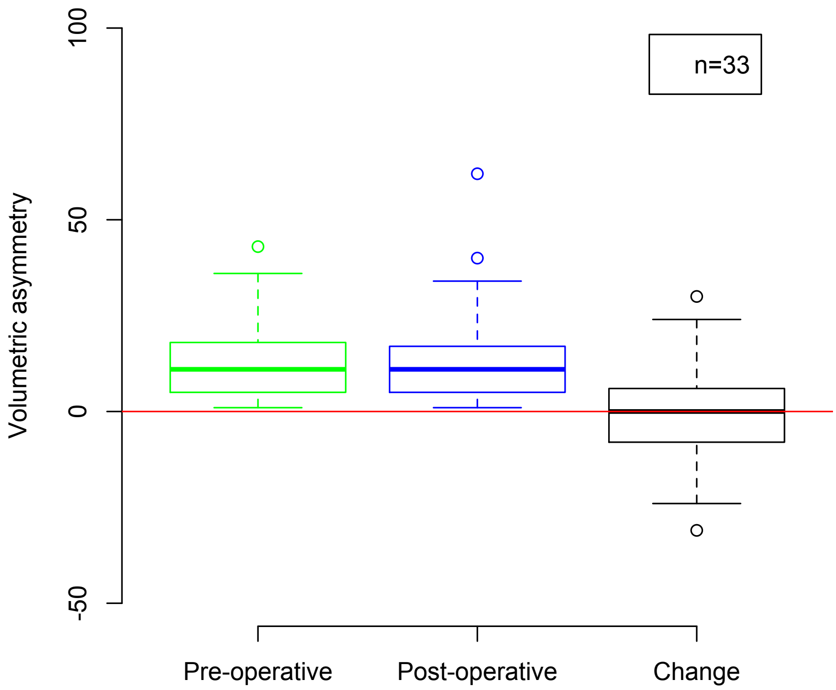


Figure 120: The VA on the right for the pre-operative to post-operative cohorts with both left and right sided VA for the main thoracolumbar curve pattern. VA is a unitless parameter that describes the difference in 3D volume between each side of the torso (see Figure 26).

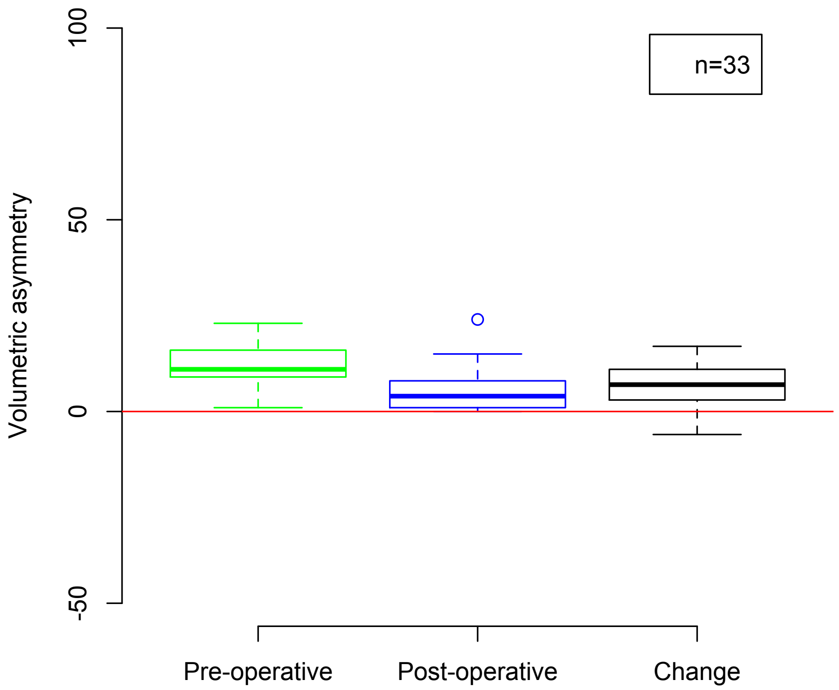


Figure 121: The VA on the left for the pre-operative to post-operative cohorts with both left and right sided VA for the main thoracolumbar curve pattern. VA is a unitless parameter that describes the difference in 3D volume between each side of the torso (see Figure 26).

Table 47: The statistical significance of the change between the size of pre-operative to post-operative VA in the main thoracolumbar curve pattern. VA is a unit-less parameter that describes the difference in 3D volume between each side of the torso (see Figure 26).

Parameter	Statistical significance	n
VA pre-operative to post-operative for VA on the right only subgroup	p = 0.250	3
VA pre-operative to post-operative for VA on the left only subgroup	p = 0.063	5
VA pre-operative to post-operative for VA on both the right and left subgroup showing only the right	p = 0.823	33
VA pre-operative to post-operative for VA on both the right and left subgroup showing only the left	p < 0.001	33

7.3.9 Summary - Most prominent points over the back and volumetric asymmetry.

Both the most prominent points and VA measure the 3D shape of the back, but in different ways. The most prominent points show that scoliosis changes the position of the points, particularly in the y coordinate (ScapDiffHt) although also in the z coordinate (ScapDiffHt). Of interest is that scoliosis surgery seems to act to increase the ScapDiffHt measure, rather than reduce it. VA seems to be more resistant to surgical intervention.

7.4 Power analysis.

The required sample size to adequately power the investigation of pre-operative to post-operative change, is assessed in section 6.8.6. Reference is made to the measure of ShDiffHt as described by Akel et al [148] and subsequently in this thesis (Figures 27 and 28 and Table 4). Within the scoliotic pre-operative group, the ShDiffHt parameter measures 6.2 $\pm$14.0 mm and the post-operative parameter measures 14.1 $\pm$ 13.6 mm. Again, using a power calculation of two independent means with a desired power of 80% and an alpha value of 0.05, 47 subjects are required to show a difference between the pre-operative and post-operative groups. Whilst, as acknowledged, the number of subjects in the main thoracolumbar group are not large enough to draw firm conclusions, the number of subjects in the main thoracic group is adequately powered for the conclusions made.

7.5 Conclusion.

7.5.1 Summary of findings.

This chapter shows the change that scoliosis causes in the shape of both the torso and the spine. In comparison to the symmetrical torso, a right sided thoracic curve leads to a greater height difference in the axillae and in the distance from the midline of the axillae. This is not seen in the height difference of the shoulders. At the waist, a convex to the left thoracolumbar curve causes the waist points to become more superior on the left and further from the midline on the right. In comparison to the variability of shape for these parameters seen in the non-scoliotic cohort, this change in shape caused by the presence of scoliosis is an amplification seen in the non-scoliotic group. With scoliosis, kyphosis and lordosis are statistically different to the non-scoliotic group but the actual change is within the measurement error of Cobb angle measures [208] and would thus not be clinically significant. The most prominent points, as a measure of 3D shape, show that a right thoracic curve causes the right most prominent point to move further from the midline than the left, more inferior than the left and more prominent than the left.

The changes in VA are similar in that scoliosis increases the asymmetry seen. Reference is made here to right thoracic and left thoracolumbar curves, the main thoracic and compensatory thoracolumbar curve pattern, as this is the most commonly seen in the study data. The results for a main thoracolumbar and compensatory thoracic curve pattern must be taken with the knowledge of the small numbers of participants in those groups.

Scoliosis surgery acts to reduce the asymmetries and this is seen across all of the torso points (AxDiffHt, AxDiffOff, WaistDiffHt, WasitDiffOff). However, ShDiffHt becomes more asymmetric with the left side more superior compared to the right. Kyphosis and lordosis are both reduced in value, as is the size of the coronal curve and these changes are all greater than the Cobb angle measurement error [208] and would be changes of import. With regards the 3D shape, the most prominent points are equalised in the x plane (MaxDiffOff) but a greater difference is seen in the y plane with ScapDiffHt, with the left being vertically higher than the right. Surgery makes little difference in the prominence of the points in the z plane. VA is different to the most prominent points in that there is little change wrought by surgery, in those sub-groups where there are enough subjects to make any statisitical analysis reasonable.

7.5.2 Relevance of findings.

Growth standards. With regards to the parameters of standing height, weight, BMI, sitting height and sitting height to standing height ratio, the data show a distribution similar to the standards. This is also seen with the number and percentage of data points above or below the median line or the 5th or 95th centiles between the paired pre-operative and post-operative data. Consequently it can be stated that scoliosis surgery, across a population, does not cause a difference in the numbers of patients who would be measured as standing outside the 5th and 9th centiles from the median value when compared to the UK-WHO [82] and Dutch standards [83]. This is of course a different question to that of "how much height have I lost because of my scoliosis?" or to the question "will I gain height following my scoliosis surgery?", both of which are often asked in clinical practice. The data presented here show an increase in both standing and sitting height but not BMI following scoliosis surgery. The evidence as to whether scoliosis causes a true height loss is mixed and can be difficult to interpret. This is because it relies on a comparison of the height of those with scoliosis to those without scoliosis. This is further confused by the point when measurements are made in the growth spurt. Goldberg et al [209] suggest that the reason young girls with scoliosis are taller than their peers is because those with scoliosis have rapid early growth. When reassessed at full growth, this discrepancy no longer exists. There is contrary evidence, however, that suggests scoliotic children, before an operation, are taller than their non-scoliotic peers [54] and that the size of the scoliotic curve is a factor, with those with larger curves being taller than those with smaller curves. This is thought to be secondary to the uncoiling effect of the thoracic kyphosis caused as part of the genesis of the scoliosis, which is greater as the size of the scoliosis increases and leads to a greater overall height [210, 211, 212]. Previous work has used mathematically derived formulae to assess height loss from scoliosis [213, 214, 215, 216, 217]. When pre-operative height was corrected with each formula, and then compared to the WHO growth standards, it was felt that the Kono [215] and Stokes [213] formulae were the most accurate [218]. There may well be a gain in height following the surgery. This has been reported and then calculated secondary to the procedure performed [219, 220, 221].

Torso points. The assessment of the 2D torso points in the pre-operative cohort when compared to the non-scoliotic cohort reveals how the torso alters shape and becomes asymmetric with a scoliosis. Chapter 6, describing the growth of the normal child, has shown the spread of shape that is seen in the non-scoliotic cohort and the scoliotic cohort is an amplification of this variability. The observation that the parameter ShDiffHt seems not to be affected (within the value of the previously defined MCID in Section 2.4) by the presence of the scoliosis curve or the surgery on that curve again suggests that the functional anatomy of the shoulder girdle allows for the underlying change in spinal shape, keeping the shoulder joint complex and thus the arms and hands the same distance from the floor. This may well help to explain why the quoted radiographic measures for intraoperative balancing of the shoulders during scoliosis surgery, in a prone position with the shoulders flexed and abducted, are not strongly correlated and thus do not explain all of the final position seen when the patient mobilises after surgery in an upright stance with the shoulders in the anatomical position [222].

Whilst the observations over the significance of the differences in the pre-operative, post-operative and non-scoliotic data are of interest, it is necessary to highlight the differences between the mean (or median) values. The largest pre-operative value is 30 mm greater than the non-scoliotic value. The largest post-operative value is 10 mm larger than the non-scoliotic value. A 30 mm difference is likely to be of clinical import and visible to onlookers whereas a 10 mm difference is less likely to be noticed by clinician or patient [148]. It can be said that with regards to the 2D shape of the torso, scoliosis surgery reduces asymmetries towards those seen in a non-scoliotic population but with some statistically significant and possibly clinically significant differences remaining.

Measures of kyphosis and lordosis. The measurement of the sagittal plane, through the size of the thoracic kyphosis and lumbar lordosis is of some import in scoliosis surgery. Recent evidence would suggest that the pathogenesis of scoliosis may be associated with the sagittal rather than the coronal plane [204, 205]. AIS is a lordosing pathology in the thoracic spine and surgical strategies are aimed at restoring kyphosis [223] and preventing the development of further deformity of the spine outside the instrumented levels [224]. In the main thoracic curve pattern the pre-operative scoliosis cohort were less kyphotic and more lordotic than the non-scoliotics and this was statistically significant. This would be expected given the anatomical location of the main curve. Surgery reduces the thoracic kyphosis further by a mean of 7°. Lordosis on the other hand is normalised to the non-scoliotic mean value by surgery but with a difference between pre-operative and post-operative values. The main thoracolumbar curve pattern has an anatomically lower main curve in the spine and the effect of that on the sagittal profile will be different in comparison to the main thoracic curve pattern. This is seen in the pre-operative cohort where the kyphosis and lordosis are significantly larger than the non-scoliotic cohort. Surgery reduces both the kyphosis and lordosis, normalising the lordosis. In both curve patterns scoliosis surgery is a flattening procedure to both thoracic kyphosis and lumbar lordosis.

Most prominent points. In the symmetrical torso, the most prominent points on the left and right scapula should be a symmetrical distance from the midline, with no difference in vertical height and an equal protrusion from the coronal plane. The parameter MaxDiffOff, measuring the difference in the horizontal distance from the midline of the most prominent points shows that, in the pre-operative cohort, the distance increases with the magnitude of the curve in both the main thoracic curve pattern and the compensatory thoracic curve in the main thoracolumbar curve pattern. This is reduced when surgery is performed. In the main thoracic curve pattern, there was a reduction with a change in the mean values of 36 mm. This is still significantly different to that seen in the non-scoliotic cohort, although in actual measurement, the difference in the mean values is only 4 mm. With the parameter of ScapDiffHt, the difference in vertical height of the most prominent points is significantly different in the pre-operative cohort compared to that in the non-scoliotic cohort and this difference increases post-operatively. As this is a negative number, it indicates that the left point is more superior than the right. Again in the pre-operative cohort, ScapDiffDepth, the prominence of the points is away from the coronal plane, a surrogate measure for the size of the rib hump, is greater on the right than the left. Whilst this is greater than the non-scoliotics, it is a small difference when compared to the other most prominent point parameters in both curve patterns. Surgery changes this value very little between the pre-operative and post-operative values. These figures documenting the position of the most prominent points represent two distinct possibilities. First that scoliosis is associated with a movement of the scapula and that the prominent point is always represented by the same anatomical structures, or second, that the prominent point is formed by a different part of the scapula that becomes prominent as the shape of the underlying thorax is altered through scoliosis and then surgery. Anatomically, the scapula is a large flat bone that overlies the posterior thoracic cage either side of the spine. The bone lies encased in sheets of muscle overlying the ribs. A change in rib shape could lead to a change in the orientation of the scapula and this will make a different part of the scapula more prominent thus changing the location of the highest points. The most likely scenario is a combination of both of these mechanisms, with the scapula being located more laterally around the rib hump in the MaxDiffOff parameter but more tilted in the ScapDiffHt and ScapDiffDepth parameters. Surgery seems to affect lateral placement to a greater degree than the tilting, hence the larger difference in the absolute mean values in MaxDiffOff versus ScapDiffHt and ScapDiffDepth.

Volumetric asymmetry. Volumetric Asymmetry is a measure of the rib hump and is described in three patterns, which are rib humps on the right side only, the left side only and both the right and left sides. The analysis shows that in all of these subtypes there is a decrease in volumetric asymmetry between the pre-operative and the post-operative cohorts; however, the change is small. This suggests that the parameter VA is not affected by surgery, raising the possibility that surgery produces little change in the shape of the rib hump, or that the parameter itself is a poor measure of the rib hump and of the changes that occur to the shape of the rib hump during surgery. This confirms the previous findings of Inami et al [134] and Jefferson et al [125].

Certainly, a parameter that converts a 3D rib hump that is variable in size and shape to a single number runs the risk of losing description of the rib hump. VA does not deal with the differences between a short sharp 'razor back' deformity and a larger, flatter rib hump, both of which could have the same value of VA. However, as is seen in the non-scoliotic cohort, VA has the ability to discriminate between different subgroups of normal growth. It may well be, that with further analysis, a different measure of rib hump could be identified. It must be also noted that this objective measure is being compared to the subjective measure of the 'eye of the surgeon' who may well have a degree of inbuilt bias. In the end, the resolution will be to compare measures of shape to a validated outcome measure assessing the patient's own view of their own shape [131].

8 Procrustes analysis of torso shape.

8.1 Introduction.

Procrustes analysis is a technique used for the analysis of the distribution of shape. It is performed using methods that analyse a series of shapes to create a mean shape. The name Procrustes comes from ancient Greek mythology [184]. Procrustes was the other name of Damastes, who invited travellers to spend the night in his house. They were then robbed after being killed by being fitted to Procrustes' bed, either through stretching in a rack if too short, or through amputation of body parts if too long. Procrustes was a victim of his own methods when he met and was defeated by Theseus, who was a King of Athens and better known through his adventures on Crete in the labyrinth with the Minotaur [184]. Procrustes analysis allows the assessment of the mean shape and the variability of that mean shape from a number of shapes of different sizes and orientation, but that all have common identifiable landmarks. This chapter describes the use of Procrustes analysis methods using the techniques described by Dryden and Mardia [185] to compare the shapes of the non-scoliotic and scoliotic cohorts, both pre-operatively and post-operatively.

8.2 Methods.

A Procrustes analysis is a statistical comparison of a group of shapes with common landmarks when the effects of scale, rotation and location are removed. First, all of the contributing shapes are moved to a common location and, for all of the analysis performed in this chapter, that was defined as the position of the vertebra Prominens (VP), which was defined at a location of zero in the x, y and z plane. The locations of all of the landmarks are then measured relative to this point on each shape. This is called the 'raw data'. In a Procrustes analysis, the first step is the translation of all of the shapes to a common centroid. The scaling, rotation and translation matrix is then calculated which provides the least squared best fit for all of the shape data. This is known as the 'rotated data' (although this is actually the data after translation, rotation and scaling). From the rotated data it is possible to extract the location of the mean position of the centroid of data for each individual landmark (known as the mean shape or mshape). The variability of the data that forms the mean shape at each landmark is calculated using principal component analysis (PCA) and this is illustrated by the PCA plot. The axes of variability shown correspond to the first three principal components. These axes are not necessarily orthogonal and are known as the eigenvectors.

There are two different methods of comparing shape described by Dryden et al [185], an ordinary Procrustes analysis (OPA) and a generalised Procrustes analysis (GPA). A GPA is the method used to compare more than two shapes and will give a mean shape for all of the contributing shapes whereas an OPA will compare two shapes only. In this chapter, GPA was used to find the mean shape and principal components for the non-scoliotic, pre-operative scoliotic and post-operative scoliotic cohorts. When comparing the mean shape from one of the cohorts against the mean shape of another cohort (non-scoliotic against pre-operative scoliotic) an OPA was used. Assessment of any statistically significant differences between data sets was by the statistical tests described by Hotelling [191], Goodall [192] and James [193] with significance pre-defined as $p < 0.05$. This description of Procrustes analysis is valid for both 2D and 3D landmark data.

For the 2D analysis, the landmarks were the x and y locations of the torso points used in previous chapters for the data ellipses (found in the Methods chapter as Figures 27 and 28 and Table 4), mamely:

1. The vertebra prominens (VP) at the superior end of the palpated spine.
2. The sacrum, a point midway between the locations of the dimples of Venus.
3. The left and right shoulder points.
4. The left and right axillae points.
5. The left and right waist points.

To allow 3D analysis, the x, y and z locations of the landmarks for 2D analysis were combined with the most prominent points over the back (Figure 29 and Table 5 in the Methods chapter).

For both the 2D and 3D data, GPA was applied as described by Dryden and Mardia [185] using the shapes package [225] to give a mean shape for both males and females in both the non-scoliotic and scoliotic cohorts (pre and post-operative). It was felt important to look at 2D shape before considering 3D shape because there was a possibility that the accuracy of the z parameter of the torso points may be affected in an x and y plane in the 3D analysis as it would be found over an area of rapid depth change at the edge of the body. The 2D shape of the non-scoliotic cohort was examined to identify the differences between the sexes, along with the change in torso shape and size with increasing age.

For 3D Procrustes analysis, the raw data, the rotated data and the principal components were plotted. To realise the differences both graphically and statistically, the mean shape function was used. The mean shapes of both males and females in the non-scoliotic cohort were superimposed to demonstrate any difference in size and then manipulated using OPA to eliminate the effects of size, allowing a pure assessment of asymmetry. Combining the male and female groups in the non-scoliotic cohort was important for the comparison of the non-scoliotic cohort to the scoliotic cohort where, as seen in Chapter 7 (where the pre-operative to post-operative change is described), the number of males was too small for meaningful analysis.

As in Chapter 6 (The normal child) and Chapter 7 (Pre-operative to post-operative change), the data from both the non-scoliotic and scoliotic cohorts were divided into those with a main thoracic curve pattern and those with a main thoracolumbar curve pattern [55]. The analysis of the scoliotic cohort was performed with the most common curve types and did not mix curves with convexities on both the right and left sides. Thus the thoracic curves are only curves convex to the right and the thoracolumbar curves are only curves convex to the left.

8.3 Results of Procrustes analysis in 2D.

Figures 122 to 125 show representative plots of the raw, rotated and PCA data for the non-scoliotic cohort with a main thoracic curve pattern. Note that all figures in this chapter are presented as if looking from the back of the subject making the left side of the figure the left side of the subject. There was a significant difference between the males and females in the non-scoliotic cohort by all statistical tests for both curve types ($p < 0.001$). The majority of the variability seen in the position of the landmarks was around the position of the shoulder, axillae and waist points and described the change in overall torso shape from a taller and thinner torso to a shorter and rounder torso (Figures 124 and 125). This, however, was also symmetrical.

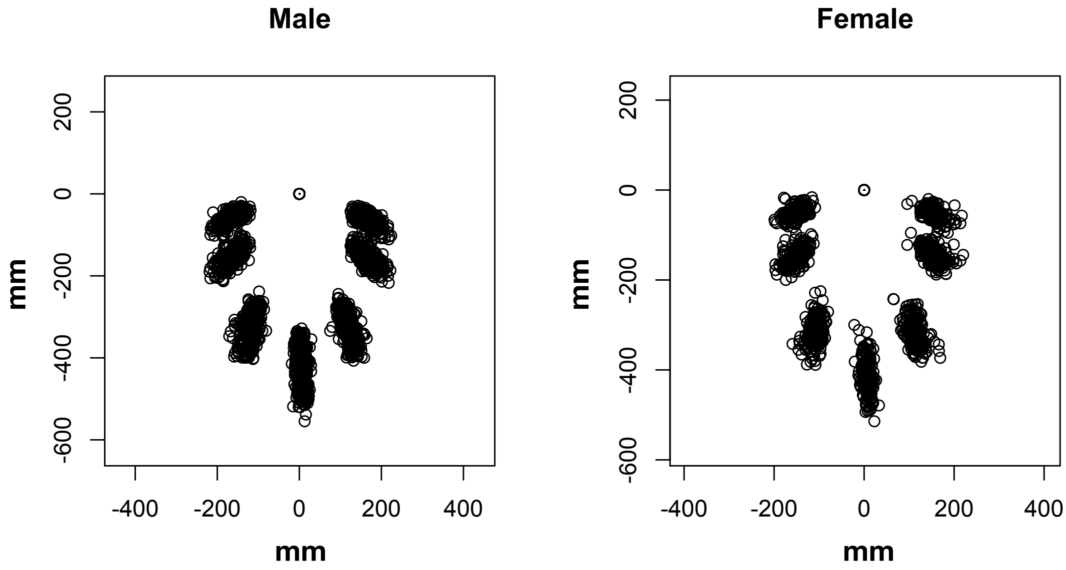


Figure 122: The raw data position (mm) for males and females for the non-scoliotic cohort with a main thoracic curve pattern. Each clump of points represents one individual landmark defined in Figures 27 and 28 and Table 4

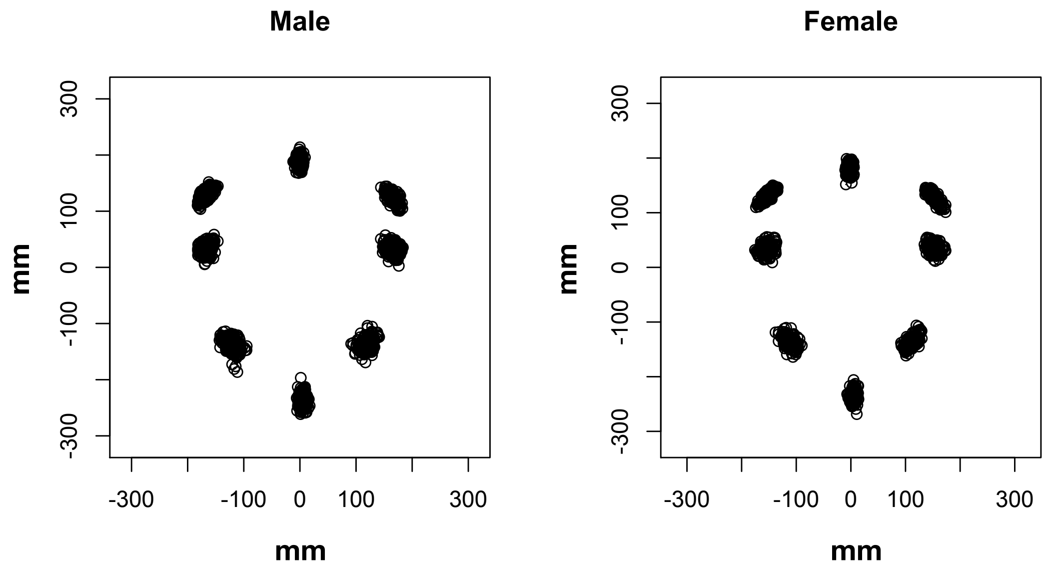


Figure 123: The rotated data position (mm) for males and females for the non-scoliotic cohort with a main thoracic curve pattern. Each clump of points represents one individual landmark defined in Figures 27 and 28 and Table 4

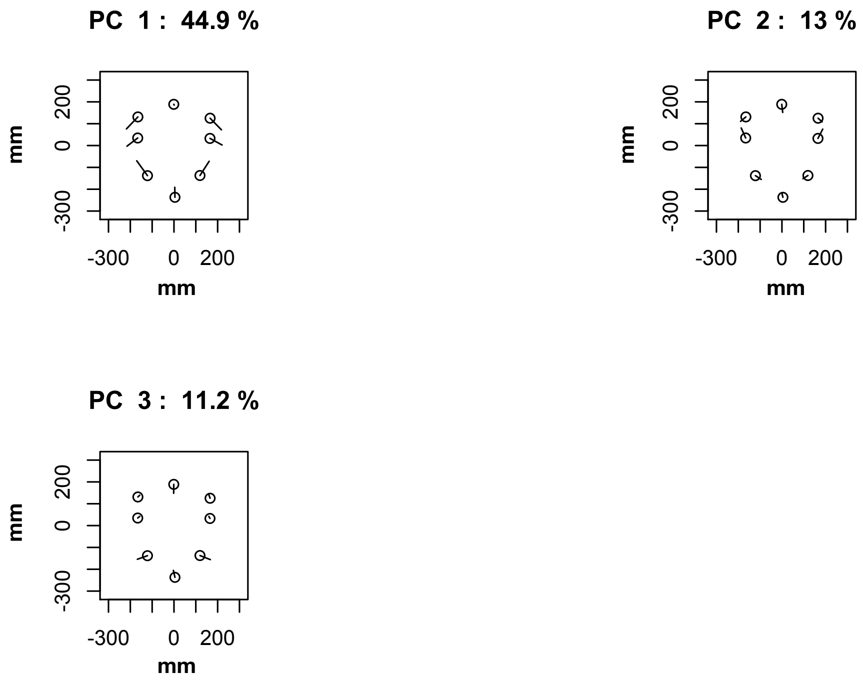


Figure 124: The PCA plots (mm) for males in the non-scoliotic cohort with a main thoracic curve pattern with the percentage contribution of each PC to the total variability in shape. Each point represents one individual landmark defined in Figures 27 and 28 and Table 4

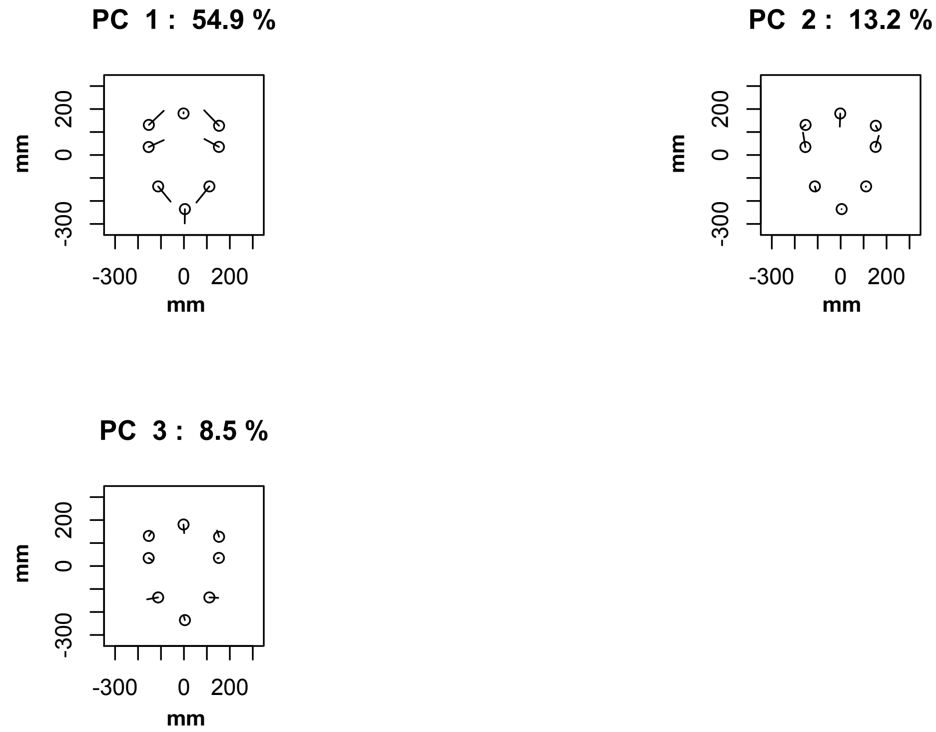


Figure 125: The PCA plots (mm) for females in the non-scoliotic cohort with a main thoracic curve pattern with the percentage contribution of each PC to the total variability in shape. Each point represents one individual landmark defined in Figures 27 and 28 and Table 4

By plotting the mean shape data for each age, the growth of the torso in 2D in both males and females is shown in Figure 126. This demonstrates growth away from the centre by all points of the torso with age for both males and females. This does cease with no difference in distance from the centre between the positions of 16 and 17 years for both males and females.

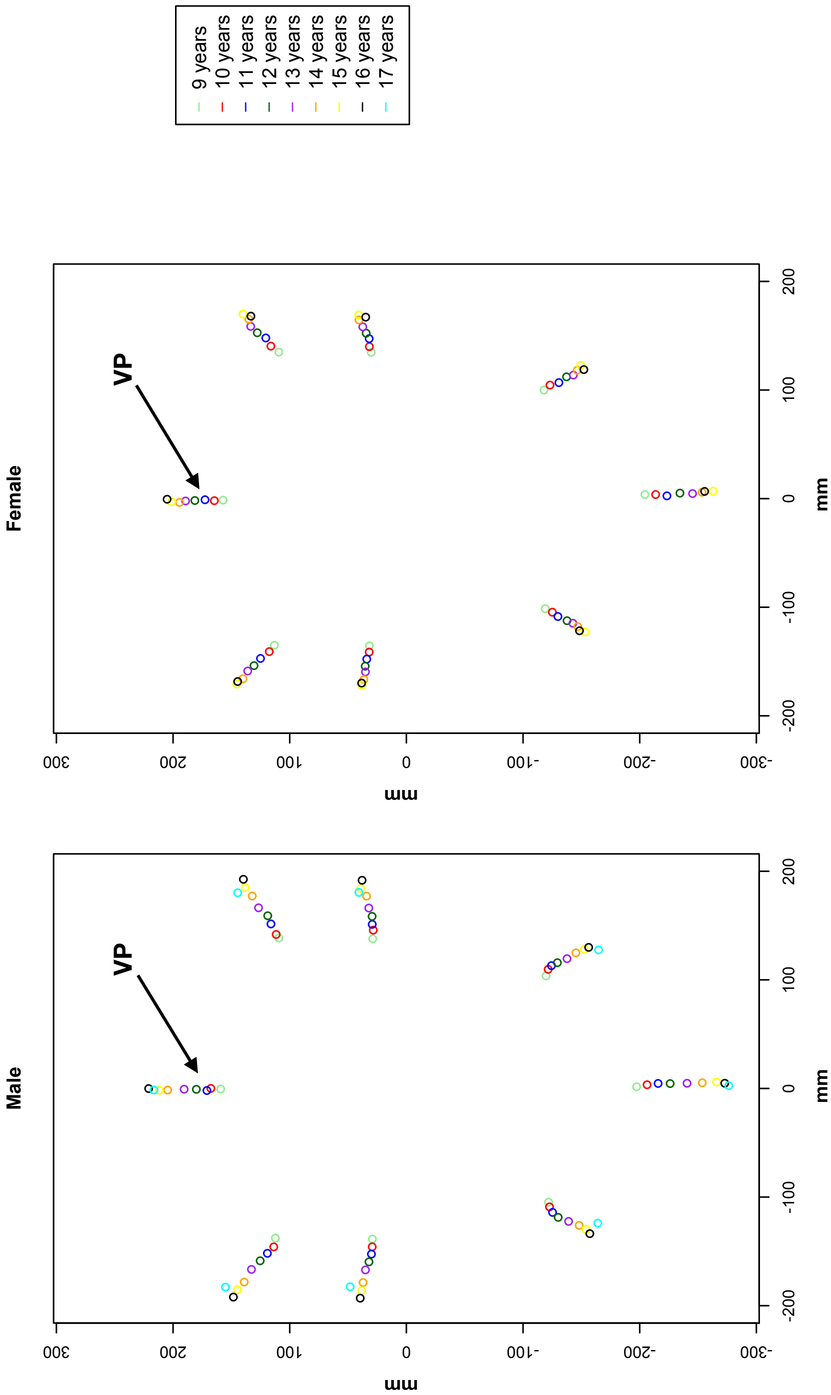


Figure 126: The growth of the torso (mm) for the non-scoliotic group subdivided for males and females. Each line of points represents one individual landmark defined in Figures 27 and 28 and Table 4. VP - vertebra prominens.

8.4 Results of Procrustes analysis in 3D.

Using the same analysis as in 2D Procrustes analysis, 3D torso data were analysed to give raw, rotated and PCA data for the non-scoliotic, the pre-operative and the post-operative cohorts. As a typical example, Figure 127 shows the combined male and female non-scoliotic raw and rotated data. The male and female data from the non-scoliotic cohort were combined to allow analysis with the scoliotic data, given the small numbers of males with scoliosis for analysis. This follows the method established in Chapter 7.

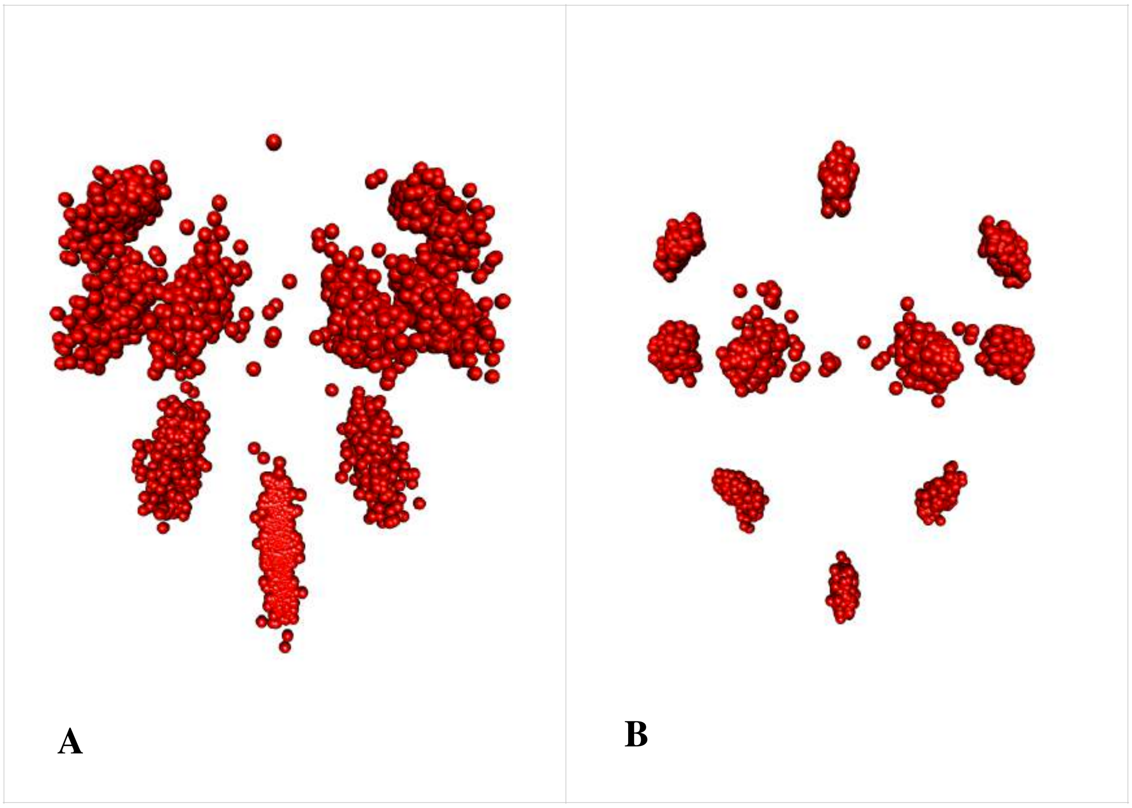


Figure 127: The raw (A) and rotated (B) data for the main thoracic curve type for the non-scoliotic cohort. Each clump of points represents one individual landmark defined in Figures 27 and 28 and Table 4 along with the most prominent points defined in Figure 29 and Table 5.

To demonstrate the variability of the mean shape of the non-scoliotic, pre-operative and post-operative cohorts, Figures 128 and 129 show the PCA plots for the main thoracic and main thoracolumbar groups. The PCs are coloured with black representing the first PC, red the second PC and green the third PC.

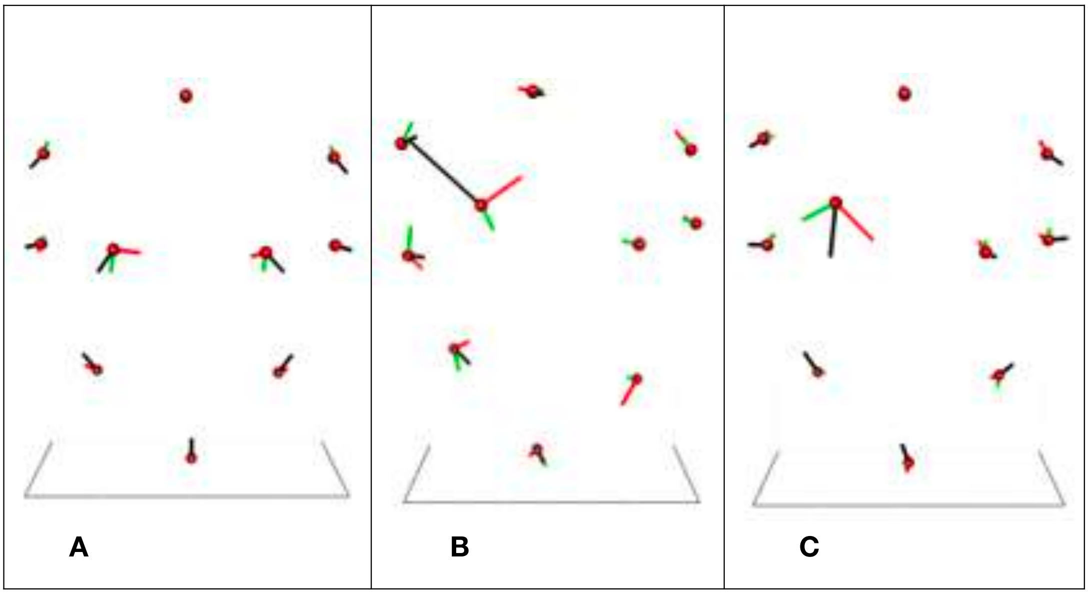


Figure 128: The PCA plots for the non-scoliotic (A), convex to the right pre-operative (B) and convex to the right post-operative (C) for the right convex main thoracic group (n=248). Each point represents one individual landmark defined in Figures 27 and 28 and Table 4 along with the most prominent points defined in Figure 29 and Table 5. The individual PCs are marked as black for PC1, red for PC2 and green for PC3.

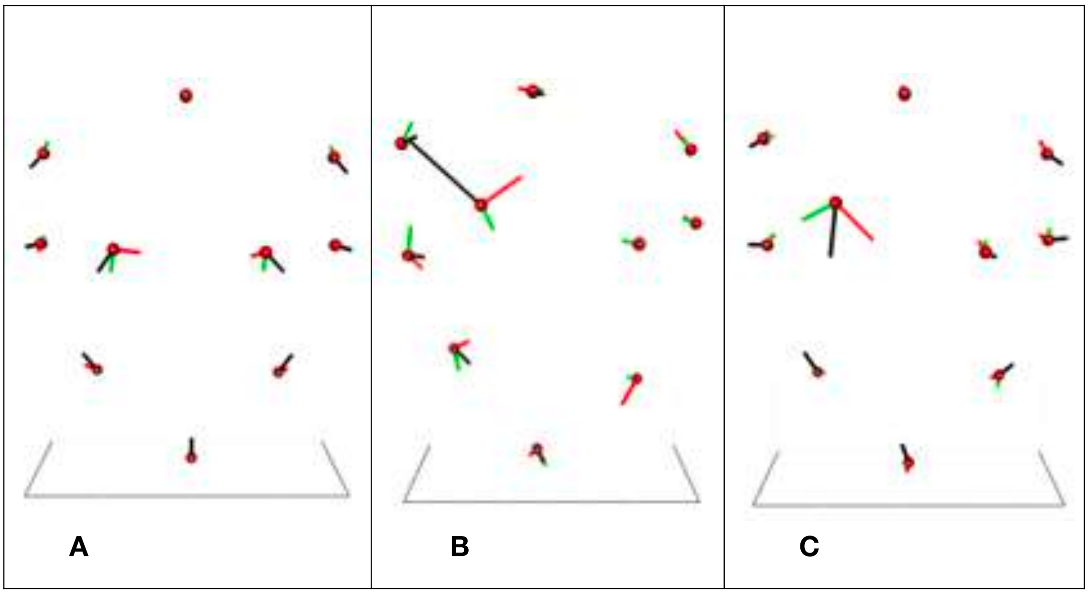


Figure 129: The PCA plots for the non-scoliotic (A), convex to the left pre-operative (B) and convex to the left post-operative (C) for the left convex main thoracolumbar group (n=41). Each point represents one individual landmark defined in Figures 27 and 28 and Table 4 along with the most prominent points defined in Figure 29 and Table 5. The individual PCs are marked as black for PC1, red for PC2 and green for PC3.

Figures 130 and 131 are plots that demonstrate the mean shapes of the non-scoliotic, pre-operative and post-operative cohorts superimposed on one another for the main thoracic and main thoracolumbar groups. The non-scoliotic is red, pre-operative is green and post-operative is blue (as for the data ellipses in Chapter 7 describing pre-operative to post-operative change). There are four views representing the 3D nature of the shape with a PA or front to back view (A), a forward bend view (B), a left rotated view (C) and a right rotated view (D). A plot combining the 3D mean shapes and PCA together for all three groups is too messy for easy interpretation, but if the plots of Figures 130 and 131 are superimposed on Figures 128 and 129 in the mind's eye, a true appreciation of the data can be appreciated.

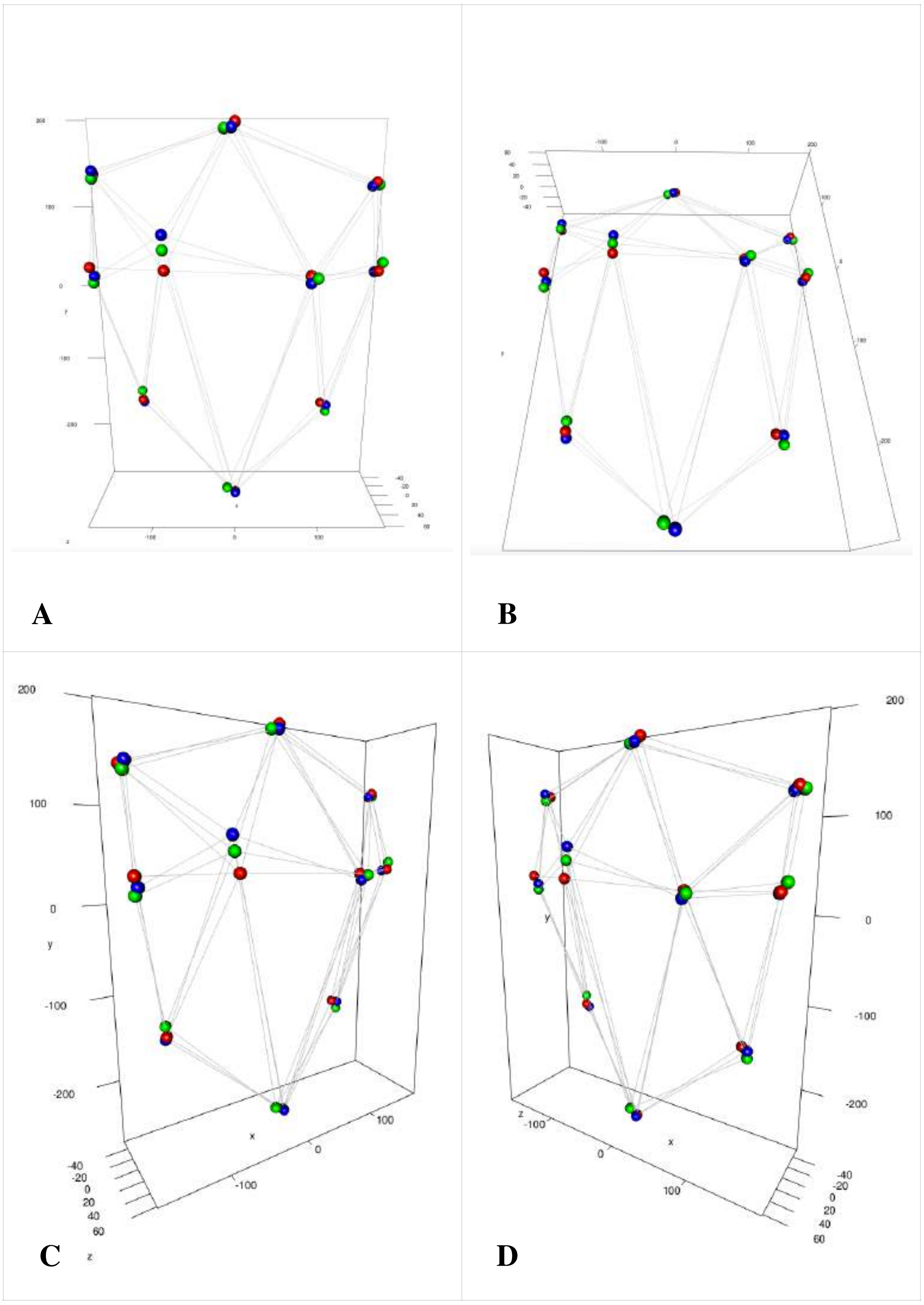


Figure 130: A composite view of the 3D mean shape of the non-scoliotic (red), pre-operative (green) and post-operative (blue) cohorts in the right convex main thoracic pattern. All units in millimetres. Each point represents one individual landmark defined in Figures 27 and 28 and Table 4 along with the most prominent points defined in Figure 29 and Table 5.

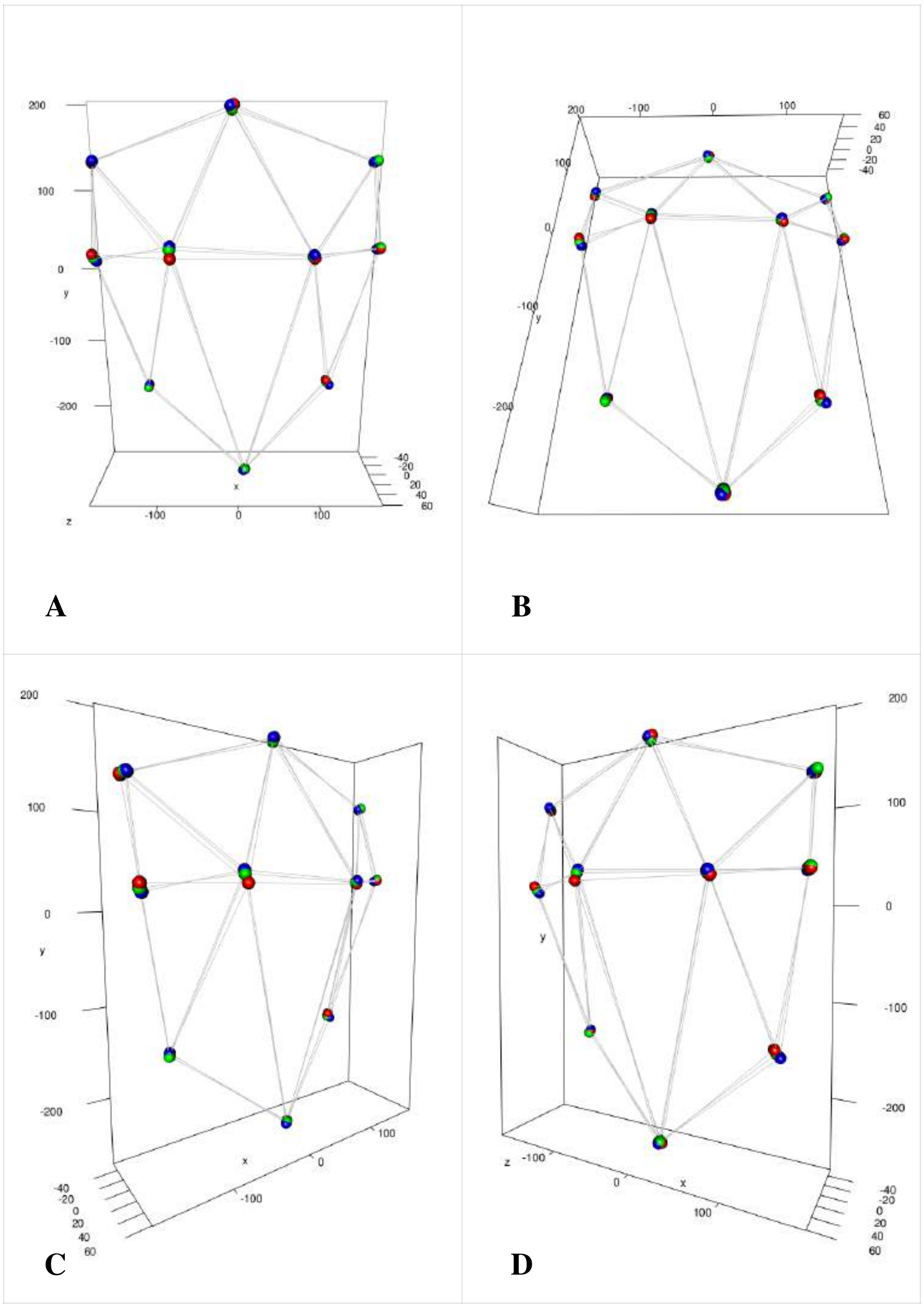


Figure 131: A composite view of the 3D mean shape of the non-scoliotic (red), pre-operative (green) and post-operative (blue) cohorts in the left convex main thoracolumbar pattern. All units in millimetres. Each point represents one individual landmark defined in Figures 27 and 28 and Table 4 along with the most prominent points defined in Figure 29 and Table 5.

In the composite plot of the main thoracic curves, the most obvious difference between the scoliotic and the non-scoliotic cohorts is that of the most prominent point on the left side of the image. This is also the point with the greatest variability in the PCA plots. Of note is, when looked at post-operatively, the most prominent point on the left side of the image is more superior than the pre-operative position. The asymmetry between the difference in the y axis of the left and right point has increased following surgery. Again the PCA plot for the post-operative cohort notes the greatest amount of variability around this point as pre-operatively, although in different directions compared to the pre-operative data. The effect of a thoracolumbar curve pattern on 3D torso shape is less marked, although again, the most prominent point on the left is the landmark with the most difference both pre-operatively and post-operatively from the non-scoliotic cohort and has the greatest variability in the PCA plots.

There was a significant difference ($p < 0.001$) between the non-scoliotic cohort and the pre-operative cohort, the non-scoliotic cohort and the post-operative cohort and the pre-operative and post-operative cohort when measured using all three statistical methods.

8.5 Conclusion.

8.5.1 Summary of findings.

In a non-scoliotic cohort, Procrustes analysis shows that the torso, as measured by landmarks around the edge of the torso along with the superior and inferior ends of the spine and the highest points over the torso, shows little asymmetry. There is a degree of variability around the position of the mean points for the landmarks, but this is also similar when comparing the left and right sides of the torso. This is true for both the main thoracic and main thoracolumbar curve patterns. Scoliosis alters the shape of the torso, particularly in the position of the most prominent point on the left scapula area. It is of interest that scoliosis surgery makes the difference in the position of the most prominent points between the left and right sides larger. This is true for both the main thoracic and main thoracolumbar curve patterns, although less so for the main thoracolumbar pattern. This is not that surprising given the anatomical proximity of a thoracic curve versus the thoracolumbar curve to the shoulder girdle. The positions of the most prominent points is invariably over the scapula. The points do not however represent a fixed anatomical point on the scapula but are what the observer will see as part of the rib hump. The position of the left most prominent point, being more superior than the right, is possibly related to the effects of the scoliosis on the underlying thoracic cage. In AIS, the rotation of the vertebral body leads to alterations in the geometry of the ribs on both the convex and concave sides of the curve [3].

8.5.2 Relevance of findings.

In the concavity of the thoracic curve, the geometry of the ribs becomes less angled to the coronal plane, appearing more horizontal to the observer. Anatomically, the scapula is a large flat bone found superficial to the thoracic cage and is only fixed to the axial skeleton by the acromioclavicular joint (ACJ). A change in the underlying geometry of the ribs could lead to a different part of the scapula becoming more prominent through tilting of the scapula around the ACJ, and the most prominent point being apparent in a different location to a non-scoliotic cohort or to the other side of the torso. Why surgery increases this difference is not clear, but the conclusion drawn from this difference is that the current procedures used to straighten the spine do not affect the ribs in such a way as to reduce the differences in geometrical position of the most prominent points between the left and right sides. Given that the rib hump is one of the key features that cause distress for those with scoliosis [31, 32, 94], further thought as to how scoliosis surgery could improve this is warranted. It is of note that the most prominent point is just one point on the rib hump and that the rib hump is a 3D structure in its own right. The position of the most prominent point is a proxy measure for the rib hump and does not reflect the totality of the rib hump which can vary in both size and shape. Current scoliosis surgery is not as effective at altering torso shape as it is in changing spinal shape.

9 Overall conclusion and discussion.

9.1 Introduction.

Adolescent idiopathic scoliosis (AIS) is a condition of concern for a group of affected young people across the globe [54, 91, 105, 116, 206, 207]. AIS causes a spectrum of deformity of the spine and torso. Management of AIS is aimed at first, the prevention of progression, but second, to minimise the visual asymmetry of the torso [93, 94, 95]. The latter is important as the asymmetry that a person perceives in themselves is a source of distress and can lead to disorders of mental health [1, 2, 30] in a small group of individuals.

This thesis has used the ISIS2 surface topography system to serially measure a group of children through their adolescent growth spurt to identify normative data for growth of the body and also the torso. Using this new knowledge, the same parameters are examined in a group with scoliosis, both pre-operatively and post-operatively. This entails an analysis of the effects of scoliosis on the growth and symmetry of the torso, and also how scoliosis surgery changes that asymmetry in comparison to the already established normative standards.

9.2 The findings of this thesis.

9.2.1 Literature review.

1. Previous studies in to the torso shape of a 'normal' population are a muddle of different populations, measurement techniques and methods of analysis [101, 102, 106, 127, 144, 134, 145, 146, 147, 148, 149, 150, 151, 152, 153, 154, 155, 156, 157]. There are flaws in the analysis that leads to mixed conclusions that are contradictory, for example the findings of Willner and Johnson [27] compared to

those of Giglio and Volpon [166] due to the analysis of cross-sectional data in a longitudinal fashion and the lack of subdivision to allow for analysis by sex and age.

2. Adolescents are bothered by their shape, and the asymmetry seen in scoliosis is associated with morbidity [1, 2]. Adolescents are particularly concerned about the appearance of their waist asymmetry, shoulder height difference and scapula prominence [30].

3. Previous methods of quantifying the 3D shape of the back using POTSI [134], Suzuki Hump Sum [135] and the DAPI score [106] have proven to be little used within the literature. These scores try to reduce all asymmetries down to a single number or percentage, rather than providing numerical values for the amount of asymmetry.

4. There are many different techniques for the measurement of surface topography. With the more modern techniques, there is little to choose between them. However, there are issues in being able to take the output of one system and relating it to the output of another system. Surface topography techniques are preferred to other methods of measuring surface shape, particularly as the images are captured in the upright position.

9.2.2 Proof of normality of the non-scoliotic cohort using published growth standards.

A cohort of children from a UK school have been shown to have parameters of growth that are similar to that described in the UK-WHO [82] growth standards for standing height, weight and BMI and the Dutch standards for sitting height and sitting height to standing height ratio [83].

9.2.3 The normal child.

Normative values for the growth of the cohort were measured and recorded, demonstrating the range of variability in the 'non-scoliotic' cohort for all parameters of torso growth. This has shown that:

1. Parameters of growth differ between males and females and with age. As might be expected, height (both sitting and standing), weight and BMI all increase with age as the child grows. The female growth spurt tails off before the male and hence, in the older years of adolescence, males are larger in all measures other than BMI. Males are broader with a greater width between the shoulders than females and to a lesser extent the waist.

2. The spine has a small amount of coronal bend in most of the cohort, although only enough to qualify as a scoliosis in a small number [8].

3. There is a pattern to the curves seen with a main thoracic and compensatory thoracolumbar pattern along with a main thoracolumbar and compensatory thoracic pattern, similar to that described in the Lenke classification [55] for scoliosis.

4. The mean amount of asymmetry in the torso points representing the positions of the shoulders, axillae and waist, relating the left to the right sides of the body, is close to symmetry. With increasing age for both males and females there is an increasing asymmetry seen in the shoulder points, but not seen elsewhere.

5. A range of asymmetries is seen in the shoulders, axillae and waist torso points when plotted against the coronal shape of the spine in data ellipses. Although the mean values are all within 5 mm of perfect symmetry, the 95% confidence intervals describing the range of the data shows that some parameters vary by $\pm$ 40 mm. For the shoulders, the direction of the curve does not seem to affect the

difference in height. This is not so for the axillae and waist where an increasing curve leads to an increasing asymmetry.

6. There is a noticeable difference in the measures of kyphosis and lordosis between males and females with increasing age over the adolescent years. Female kyphosis decreases and then increases whereas male kyphosis increases steadily until maturity. Female and male lordosis increase in step with each other but females are always more lordotic.

7. The assessment of 3D shape through analysis of the locations of the most prominent points on each side of the back shows that, other than the right side being more prominent than the left at all ages for both males and females, there is no real difference between the sexes or with age for the position of the point in either the x or y coordinate. This is not replicated when measuring volumetric asymmetry, where three patterns emerge with a right sided rib hump that increases with age whereas any left sided rib hump does not., This is true for both unilateral rib humps and for individual components of combined rib and lumbar humps, and this is independent of sex. Both parameters give a measure of the size of the rib hump. Neither method adequately captures all possible information about the rib or lumbar hump and thus both are reported.

9.2.4 Pre-operative to post-operative change.

1. The effect of AIS on the parameters of height (standing and sitting), weight and BMI is seen particularly in a loss of sitting height when compared to the UK-WHO growth standards [82], and surgery does change the parameters of height, weight and BMI in the direction of the expected spread of data across the standard, albeit imperfectly.

2. The effect of AIS on the torso asymmetry is an amplification of that seen in the non-scoliotic group. Again, the shape of the spine subdivided into a main thoracic and compensatory thoracolumbar pattern along with a main thoracolumbar and compensatory thoracic pattern, as described by the Lenke classification [55]. As expected from the Lenke paper [55], a greater number of main thoracic curves are seen in comparison to the number of main thoracolumbar curves. Increasing spinal deformity increases the asymmetry in axillae and waist but not the shoulders in the direction of the curve in all parameters apart from the distance of the waist point from the midline. Surgery acts to reduce all of the asymmetries around the torso. Whilst there is a similar mean value between the post–operative scoliotic cohort and the non-scoliotic cohort (within a maximum of 10 mm), the variability of that mean is far larger for the post-operative cohort than the non-scoliotic cohort. The small number of main thoracolumbar curves makes sensible analysis more difficult.

3. Both kyphosis and lordosis are not affected by the presence of AIS when compared to the mean values of the non-scoliotic cohort when compared to a clinically important value (defined as an amount greater than the measurement error for the Cobb angle [208]). Surgery in both the main thoracic and main thoracolumbar groups has a flattening effect with a loss of both kyphosis and lordosis.

4. AIS affects the 3D shape of the torso. The convex to the right thoracic curve makes the right sided most prominent point further from the midline, lower than the left and more prominent. Surgery reduces the distance from the midline, and the difference in prominence, but makes the left point even more superior than the right. Volumetric asymmetry follows the same pattern established in the non-scoliotic group, with all measures of the right being larger than the left. Surgery

seems to have little effect on the amount of volumetric asymmetry.

9.2.5 Procrustes analysis of torso shape.

1. When using Procrustes analysis to remove the effects of differential size and shape from the non-scoliotic and scoliotic cohorts, a true appreciation of the variability in the pre-defined landmarks (torso points) (Figures 27 and 28 and Table 4) and most prominent points (Figure 29 and Table 5) can be seen.

2. In the non-scoliotic cohort, growth leads to an increase in the size of the torso, with expansion from the centre across all landmarks. This is seen in both males and females.

3. In 3D, the non-scoliotic cohort appears reasonably symmetrical to visual inspection. There is an amount of variability in the mean position for each of the landmarks and again this is symmetrical in magnitude and direction to visual inspection.

4. The most obvious asymmetry seen, when comparing the pre-operative, post-operative and non-scoliotic mean shapes, is the position of the left sided most prominent point, along with the variability of that position. In the non-scoliotic group, the prominent points appear symmetrically placed. In the pre-operative group, where the effect of AIS on this point can be seen, the left most prominent point is positioned superiorly compared to the postiton of the right most prominent point. The right point is, however, more prominent. The change from pre-operative to post-operative makes the right most prominent point less prominent, but the superior positioning of the left most prominent point is more marked. Surgery is making this element of the most prominent points more asymmetric.

9.3 Strengths of this thesis.

The work presented in this thesis is new and novel for several reasons. Literature on the growth and development of a cohort of children without pathology is historic [144, 145, 149, 150, 151, 152], and reflects children of a time where health and thus growth of children may well have been different [194]. This thesis has established normative values for the development of parameters of growth, both in all measures of height and weight, and also in the dimensions of the torso and the shape of the spine. Changes in these parameters are mapped over the critical time of the adolescent growth spurt and subdivided for sex.

The analysis of the normative data and how it develops over time is also key to this thesis. True longitudinal analysis requires multiple measures on sequential occasions to be assessed using all of the data and recognising that the data is linked to the participating subjects. Averaging the data leads to loss of information and the possibility of an erroneous result. Using regression that does not allow for the inherent error that occurs with multiple sequential measures from the same individuals could also lead to an incorrect result. Finally, presenting cohort data as longitudinal data is very misleading. The data presented in this thesis uses linear mixed effect models (LMEM) [198, 199], a technique that addresses all of these analysis issues. LMEM allows for error in multiple measures from the same individual, represents the data longitudinally and creates a model using all of the data set, so there is no chance that key data points are ignored as part of the analytical technique. Therefore, the data presented in this thesis represents data using the appropriate technique of analysis. This is particularly well illustrated by the findings with regards to the development of kyphosis and lordosis. There is growing literature that AIS is related to the sagittal profile of the spine [29, 54], and previously this has been linked to the development of the spinal sagittal shape, both by Dickson [226] and Willner and Johnson [27]. However, as highlighted in Chapter 2, Giglio and

Volpon [166] repeated the work of Willner and Johnson [27] using the same technique but with markedly different results. Neither used LMEM analysis. The development of kyphosis and lordosis reported in this thesis partially supports the work of Willner and Johnson [27] but identifies that the difference between the sexes is not in kyphosis but in lordosis, a factor that supports the work of the Utrecht group [28, 29, 54, 204].

The use of data ellipses to represent the bivariate relationships of torso asymmetry and spinal shape is new. Whilst it is known that AIS is a condition associated with vertebral body rotation in the axial plane [3], and that AIS is associated with asymmetry of the torso [3], the link between spinal shape and torso asymmetry in both the non-scoliotic and scoliotic cohorts remains ill defined. In the non-scoliotic group, data ellipses have shown the variability in asymmetry that is seen in adolescents without spinal deformity. This is an improvement on the work previously reported because torso asymmetry is linked, in every case, to the coronal shape of the spine. The data ellipses are a novel way of presenting the variability in asymmetry in normal adolescents and can be used as the standard for comparisons of asymmetry caused by pathology.

Data ellipses were used in this research to compare the torso shape in the non-scoliotic and scoliotic groups, both pre-operatively and post-operatively. Again this is novel and visually demonstrates the effects of AIS, and the deformation in the shape of the spine, on the shape of the torso. The effects of surgery are also demonstrated and the ability of scoliosis surgery to recreate the shape of the non-scoliotic cohort can be seen. This is particularly useful because the effects can be quantified in measures useful and easily understandable to the clinician which is an improvement on the use of previous descriptions of the shape of the torso such as POTSI [134] and the like [106, 135].

Procrustes analysis is an established method for assessing variability in a group of shapes by removing the effects of location, rotation and scale. This then removes the potential issues of drawing conclusions on shape when there is not standardisation of the internal variability between those shapes. The other advantage of Procrustes analysis is its ability to review the entire 3D shape rather than just one facet of that shape. The use of 2D and 3D Procrustes analysis in this thesis strengthens the observations identified through using data ellipses, demonstrating the effects of AIS on the torso and the effects of surgery, particularly to the left most prominent point.

9.4 Weaknesses of this thesis.

As with all scientific work, there are weaknesses within the study that need to be acknowledged. The non-scoliotic cohort consists of children from a fee paying, private UK school and this, inevitably, is different to a population of children from a UK inner city and even more so from a less economically well off country elsewhere in the world. This reflects the issue of extrapolating a sample to a larger population with the confidence that the results of the sample are representative of the results of the larger population, were the experiment to be conducted in every member of the population. It is then the responsibility of the researcher to guarantee, as best as is possible, that the sample is representative. In this case, the risk that the non-scoliotic cohort was not a representative sample was tested by the comparison of the standing and sitting heights, weights and BMI against published growth standards [82, 83]. As was shown in Chapter 5, the non-scoliotic cohort did have a very similar distribution across the centiles of the growth standards. The UK-WHO growth standards [82] were based on the UK90 growth information collected in the 1990's from children in the UK. A UK population may not be equivalent to a population from another part of the world. However, given the results of Figure 4 where it is shown that the WHO [80] and CDC [81] growth charts are very similar to the UK-WHO [82] charts, it can be said with some confidence that this issue, whilst not completely resolved, is not likely to alter the results for other populations.

As noted in Chapter 4 of this thesis, there is an inherent error within the measures taken of the study participants in both the non-scoliotic and scoliotic cohorts. That error comes from the error of the ISIS2 system, the error that occurs in the preparation of the study participants for the ISIS2 image and the error that occurs in the post image processing for the identification of the torso points. All of these errors have been assessed and found to total approximately 11 mm. Of note, this is compared to the range of difference in the height of the shoulders in children who all thought that their shoulders were level and without asymmetry reported by Akel et al [148] which was 7.5 $\pm$ 5.8 mm. This has been defined within this thesis as the Mean Clinically Important Difference (MCID), over which a change is likely to be clinically important to the person concerned. The 11 mm of mean total error for the acquisition of the data for this thesis is less than the MCID from the paper of Akel et al[148].

The measure of the shape of the spine using the ISIS2 system, Lateral Asymmetry (LA) is used throughout the chapters. LA is a Cobb angle [7] proxy but the relationship is not linear. It is not possible to say that an LA of 25° is equivalent to a Cobb angle of 25° [227]. The decision to use LA as the method of measurement for coronal spinal shape is due to the inability, for ethical reasons, [37] to use whole spine radiographs, taken in upright stance to measure the Cobb angles in the non-scoliotic cohort and to compare to the angles measured in the radiographs taken as part of standard care in the scoliotic cohort. Whilst low radiation methods to image the spine including EOS imaging and Dual-energy X-ray absorptiometry (DXA) exist and have been reported for imaging scoliosis [228, 229, 230, 231], the use of these technologies are limited by availability (EOS) and not being in upright stance (DXA). Thus to make comparisons between the non-scoliotic and scoliotic cohorts possible, LA was used as the measure of coronal spinal shape for both cohorts.

It is also worth noting that the landmarks used as the torso points and as the most prominent points may not be reflective, or do not adequately represent the deformity in the torso. Other landmarks, or a different approach to the issue of documenting shape may have given a 'truer' result. However, the anatomical points used throughout this thesis reflect those previously used in the literature [106, 134, 148] and are indicative of the body issues that are reported as of concern to adolescents with scoliosis [30, 73].

9.5 Developments for the future.

The future of the care of those with scoliosis is unknown. New technologies are being reported [232] using growth guidance that will alter the methods by which AIS is managed. There is a greater awareness of the 3D torso deformity seen in AIS and this is being assessed and reported on [233, 234]. Where the results of this thesis fit in that changing world requires discussion of several issues.

First is the issue of what the normative data can reveal that might further the understanding of the aetiology of AIS. This lies within further analysis of the development of the sagittal profile and the differences with increasing age and between the sexes. It seems unlikely that kyphosis or lordosis, as standalone parameters, will explain all about how an AIS develops. The answer will probably be in the entire sagittal profile and how that interacts with other features of the spine. The results may also help to explain why AIS is more common in females compared to males [206, 207]. This is also true in identifying the relationship between vertebral body rotation and the development of the rib hump. This is important as modern techniques for AIS often have some derotation of the vertebral column as part of the corrective manoeuvre, aimed at reducing the rib hump [235]. This technique is of use surgically if there is a good relationship between vertebral body rotation and the development of the rib hump. If this relationship does not exist then it behoves surgeons to rethink the surgical techniques used in the treatment of AIS so that the best possible results are obtained. Again, the answer will be achieved by analysing the relationship between intervertebral rotation and the rib hump, combining axial imaging techniques such as MRI and information on the shape and size of the rib hump from surface topography.

Second is around the shape of the anterior torso, both in the non-scoliotic and scoliotic groups. Currently techniques for the quantitative analysis of the shape of the anterior torso are varied and do not reflect the entire issue, similar to the use of a single scoliometer measurement to describe the shape of the entire posterior torso [236, 237, 238, 239]. Whilst there is information in the literature on the variability in breast measurements, [240] this is not seen for the entirety of the anterior torso. This is also true for the effects of the surgical correction of AIS on the anterior torso where again the literature is mixed [234, 241]. All of the investigations performed in Chapters 5, 6 and 7 of this thesis could be repeated for the anterior torso. The issue is the difficulty in obtaining the surface topography of the anterior torso because, unlike the posterior torso and back, the contour of the anterior torso is affected by the presence of the breasts where it is difficult to image both the upper and lower surfaces of the breasts in a single image. The solution to this issue is to investigate methods of extending the ISIS2 technology, possibly through stitching two images together which allow a surface of the entire anterior torso to be formed that can then be analysed for relevant clinical parameters. This would then allow the assessment and management, potentially through techniques not currently available or even developed, to get the cosmetically most acceptable result as one of the products of a scoliosis operation.

Third is the use of surface topography as a method of monitoring AIS in clinical practice and using devices such as ISIS2 to reduce the requirements on radiography, most importantly from a cumulative radiation point of view [37], but also to allow efficient use of resources. Previously, ISIS1 was a commercial enterprise with Oxford

Metrics; however it is no longer available. Formetrics [79] is a commercially available system and is in use in centres in the UK. ISIS2 is in use in Leeds and London and also in Sweden and has previously been in use in Oxford and Bristol. It remains to be seen whether the information available from current surface topography systems is of enough use to physicians to make it part of widespread routine clinical practice.

Finally, the quantitative measure of asymmetry reported in this thesis is very useful to the researcher and the physician. What is not fully clear, although some literature has been published on the subject [131], is the relationship between the patients' own view of their deformity and the quantitative measure of that deformity. The difficulty here is the measurement of what is the patients' own view, such that an analysis can be performed. Current scoliosis specific tools for the measurement of the patients' view [93, 94, 98] are reasonably blunt and any use of these questionnaires would not be specific enough for the comparison with an individual parameter of asymmetry. Whilst surface topography will give quantitative data on shape, developments are required in measures for the patient themselves before that analysis can be performed.

9.6 Final thoughts.

In conclusion, AIS is a deformity of the spine and torso. Whilst scoliosis surgery straightens the spine and effects a change of shape of the torso, surgery does not recreate the shape of a non-scoliotic cohort of children. This is particularly seen in the 3D elements of torso shape. Future efforts need to be focused on understanding the development of the 3D asymmetry seen in AIS and how this can be addressed surgically or even prevented. The assessment of spine and torso shape needs to be developed to capture information on the child's own view of their shape alongside quantitative surface topography measures.

This thesis has described the spine and torso shape longitudinally in a cohort of non-scoliotic children to establish normative values. These were then compared to children with AIS and the effects of scoliosis and subsequent surgery on 3D shape established. This work will act as a benchmark for the treatment of AIS in the future.

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Appendices.

- Gardner A, Berryman F, Pynsent P. A description of three-dimensional shape of the posterior torso comparing those with and without scoliosis. Symmetry (2019). 11; 211.

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- Protocol for 'Measuring back shape with ISIS2 in non-scoliotic children'.

- Parent and child information sheets for the protocol ‘Measuring back shape with ISIS2 in non-scoliotic children’.
- Protocol for ‘The analysis of back shape in scoliosis using ISIS2 surface topography’.

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Article

A Description of Three-Dimensional Shape of the Posterior Torso Comparing Those with and without Scoliosis

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Abstract: Scoliosis results in a 3D asymmetry of the spine and torso. It is not clear what the variability in 3D shape is in a non-scoliotic population, how much that is altered by scoliosis and what surgery does to that. This study is a 3D analysis of the shape of the torso in a cohort of non-scoliotic children that is then compared with a cohort of those with scoliosis both pre- and post-operatively. Procrustes analysis is used to examine the mean 3D shape. There is variability in shape in the non-scoliotic cohort. Scoliosis increases this asymmetry, particularly around the most prominent areas of the torso. Surgery alters the torso asymmetry but increases the difference in height between the right and the left with regard to the most prominent points on the torso. There is a degree of asymmetry seen in a non-scoliotic cohort of children. Scoliosis increases that asymmetry. Surgery alters the asymmetry but causes an increase in some of the 3D elements of the most prominent areas of the torso.

Keywords: scoliosis: asymmetry; rib hump; Procrustes analysis; non-scoliotic; surface topography; ISIS2

1. Introduction

Scoliosis is a three-dimensional (3D) deformity of the spine and torso. When viewed by patients themselves or others, the scoliosis is apparent because of differences in symmetry of the shoulders, the axillae and the waist, along with the unequal prominence of the thoracic cage, otherwise called the rib hump. This visible asymmetry is of concern to patients [1] and is assessed by physicians as part of the physical examination [2,3]. Previous work using data ellipses [4,5] has analysed two-dimensional (2D) torso shape and defined the mean values and variability of the position of different torso anatomical points in both a non-scoliotic and a pre-operative and postoperative scoliotic cohort. However, the overall 3D shape of the torso in the non-scoliotic cohort and the scoliotic cohort, both pre-operatively and post-operatively, has not been previously assessed. This paper uses the techniques of 3D Procrustes analysis [6] to statistically and graphically analyse the torso shape of a cohort of non-scoliotic children, comparing the results to both the pre-operative and post-operative shape of a cohort of scoliotic children.

2. Materials and Methods

This is a comparison of two distinct cohorts. The first cohort comprised school children who have had a measurement of the shape of their back on a yearly basis from 2011 to 2017. The inclusion criteria were that there was neither scoliosis nor any other deformity of the torso. Exclusion criteria were any previous surgery to the torso or back to clinical examination. This was the non-scoliotic cohort.

The inclusion criteria for the scoliotic cohort were (1) adolescent idiopathic scoliosis, (2) no bracing undergone during treatment, (3) scoliosis treated by surgery, (4) Integrated Shape Imaging System 2 (ISIS2) back surface topography scans available pre- and post-surgery. The indication for surgery was a scoliosis in a patient that wished to pursue operative intervention for the curve, where the surgeon's view was that surgery was appropriate. This scoliotic cohort contained all curve types who had been managed with operative intervention between 2008 and 2017 using posterior-based pedicle screw techniques (269 individuals; 93%) and anterior instrumentation and fusion (20 individuals; 7%). Posterior fusion was performed using current segmental pedicle screw fixation techniques following a standard posterior approach to the spine. Correction of scoliosis was obtained using the technique most suited to get the best outcome for the individual patient and included differential rod contouring, cantilever and derotation techniques. Anterior surgery in a smaller number of patients was performed through a 10th rib thoracoabdominal, transpleural and retroperitoneal approach to expose the spine, allowing excision of the intervertebral discs and endplates back to the posterior longitudinal ligament. The instrumentation was performed using a two-screw-per-level dual rod construct over the convexity of the deformity. In posterior fusion in the main thoracic group, the levels chosen depend on each individual curve morphology and flexibility; however, in the main, the proximal level of instrumentation and fusion was T3 or T4, and the distal level was L2 or L3. For the anterior fusions performed for some of the main thoracolumbar curves, the proximal level was T11, and the distal was L3. To prevent error from mixing right and left convex curves in the scoliosis cohort, only right thoracic and left thoracolumbar curves were analysed.

In both cohorts, the surface topography of the posterior torso was captured using ISIS2 [7]. All of the captured images were then reanalysed using a custom computer interface to identify pre-defined points around the edge of the torso marking the shoulders, the axillae and the waist (Figure 1 and Table 1). When combined with the most superior and inferior ends of the spine (the vertebra prominens (VP) and the sacrum) and with the most prominent points over the left and right scapular areas of the back (all automatically generated by ISIS2) (Figure 1), there were 10 landmarks for analysis. These points are also defined in Table 1. For definition, the x-axis lies in the coronal plane, running horizontally from left to right. The y-axis runs vertically from inferior to superior. The z-axis lies in the sagittal plane and is perpendicular to the x- and y-axis. The z-values thus represent the perpendicular distance of a point on the back from the coronal plane (positive towards the observer). The origin of the axes is at the vertebra prominens in all raw data sets.

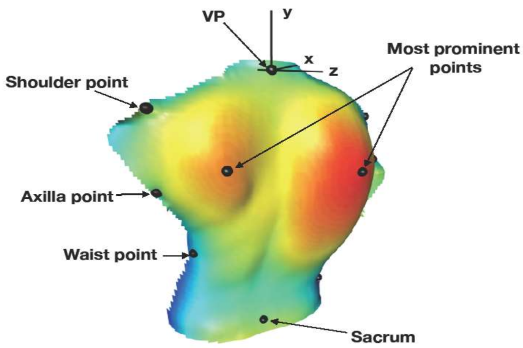

Figure 1. A diagram showing the anatomical points identified as the landmarks around the edge of the torso together with the axes.

Table 1. The definitions of the anatomical points shown in Figure 1.

Anatomical Point	Definition
Vertebra prominens (VP)	Palpable spinous process at superior end of the thoracic spine
Shoulder points	Superior aspect of shoulder girdle vertically above axillary point on the left and right sides of the body
Axilla points	Superior point of posterior axillary fold on the left and right sides of the body
Most prominent points over the torso	The most prominent point over the torso on the left and right sides of the body
Waist points	Most indrawn point of the waist crease on the left and right sides of the body
Sacrum	Distal end of the spine

As had been previously discovered from these data sets, there were two obvious patterns to the data [4,5]. Thus, prior to analysis, both the non-scoliotic and scoliotic cohorts were subdivided into a main thoracic curve pattern and a main thoracolumbar curve pattern (defined by the location of the largest curve) [8]. The size and location of the apex of the scoliosis was measured using the ISIS2 parameter lateral asymmetry (LA), which gives a measure of curve size similar to Cobb angle in the non-scoliotic cohort. This was used rather than radiographs as, due to the risks of radiation [9], it was not ethical to expose the non-scoliotic cohort to ionising radiation. The scoliotic cohort was measured using both LA and the Cobb angle [10] from pre and post-operative radiographs.

Procrustes analysis of both the non-scoliotic and scoliotic cohorts was performed using the techniques described by Dryden and Mardia [6] using the 'R shapes' package [11]. Using the predefined landmarks, Procrustes analysis was used to remove the rotation, scale and location effects from a set of shapes so that 3D comparison can be carried out. Generalised Procrustes analysis (GPA) initially translates all the shapes to be analysed to a common centroid and then works out the scaling and rotation matrix needed to achieve a least-squares-defined best fit among all the landmark shape data under analysis. The GPA results in a mean shape which gives the mean locations of the translated, scaled and rotated landmarks. A scatter plot of the scaled and rotated landmarks shows the variability in the data around the mean shape. Principal component analysis (PCA) was then performed on the scaled and rotated landmark data to demonstrate the axes of most variability for each landmark. The three axes shown are not necessarily orthogonal but rather represent the vectors of maximum variability in position (otherwise known as the eigenvectors).

Using the mean shape for both the non-scoliotic and scoliotic cohorts, an ordinary Procrustes analysis (OPA) was performed. This allows a comparison of shape for the non-scoliotic and scoliotic cohorts and the pre-operative and post-operative cohorts. OPA differs from GPA as OPA is used to compare only two data sets rather than multiple data sets which are required for comparisons of the mean shape of the non-scoliotic, the pre-operative and the post-operative scoliotic cohorts. Three methods of testing the statistical significance of the differences between the non-scoliotic, pre-operative and post-operative mean shapes were used, namely Hotelling's T test [12], Goodall's F test [13] and James's test [14]. Significance was defined as $p < 0.05$. This analysis was used to examine the differences in shape between the sexes in the non-scoliotic cohort and the differences in shape between the cohorts independent of sex. The statistical and graphical analyses were performed using R [15]. A definition of clinical significance was made following the work of Akel et al. [16].

Ethical approval for this study was given by the NRES committee West Midlands—South Birmingham (11/H1207/10) and the NRES committee East Midlands—Northampton (15/EM/0283).

3. Results

The non-scoliotic children were measured on a yearly basis, giving a total of 831 individual images for analysis comprising 479 in the main thoracic group, 307 in the main thoracolumbar group and 45 with no scoliosis. The cohort comprised 117 males and 79 females with a range of ages at the time of measurement of 9.2 to 17.3 years (Table 2).

Table 2. The demographics of the non-scoliotic cohort.

Sex	Number of Main Thoracic Curve Type	Number of Main Thoracolumbar Curve Type
Male	305	179
Female	174	128

The scoliotic gcohort comprised 39 males and 250 females with a mean age of 13.9 years (SD 1.56 years, range 9.9 to 17.9 years) at the time of the pre-operative scan. This gave a total of 289 paired pre-operative and post-operative images for review. The pre-operative major curve, measured using the Cobb angle [10], was mean 66° (SD 17°, range 32° to 133°) for the main thoracic curve and mean 60° (SD 16°, range 40° to 99°) for the main thoracolumbar curve. Post-operative values were mean 28° (SD 12°, range 12° to 88°) for the main thoracic curve and mean 23° (SD 12°, range 8° to 50°) for the main thoracolumbar curve. Following the Lenke classification [8], there were 257 Lenke 1 curves, 2 Lenke 2 curves and 30 Lenke 5 curves. Body mass index was calculated for the non-scoliotic cohort, and it was mean 20.0 kg/m^2 (SD 3.2, range 13.5 to 31.6) for males and mean 19.7 kg/m^2 (SD 2.9, range 14.0 to 33.1) for females. In the pre-operative cohort, males had a mean BMI of 20.3 kg/m^2 (SD 4.2, range 15.6 to 30.3), and females had a mean BMI of 20.7 kg/m^2 (SD 4.2, range 14.1 to 41.9). There were 248 individuals in the main thoracic group and 41 in the main thoracolumbar group (Table 3). The mean time from the pre-operative scan to surgery was 413 days (SD 304, range 1 to 2057), and the mean time from surgery to the post-operative scan was 263 days (SD 270, range 34 to 1984).

Table 3. The demographics of the scoliotic cohort.

Sex	Number of Main Thoracic Curve Type	Number of Main Thoracolumbar Curve Type
Male	35 (mean age 14.5 years, SD 1.6, range 9.9 to 17.9)	4 (mean age 15.4 years, SD 0.9, range 14.3 to 16.5)
Female	213 (mean age 13.7 years, SD 1.5, range 10.2 to 17.7)	37 (mean age 14.3 years, SD 1.8, range 10.5 to 17.0)

The raw landmark data and the scaled and rotated results after GPA were plotted for the two patterns of scoliosis already described (main thoracic and main thoracolumbar) for the non-scoliotic, the pre-operative and the post-operative cohorts. An example of these plots is shown in Figure 2A (raw data) and Figure 2B (translated, scaled and rotated data) for the main thoracic group of the non-scoliotic cohort.

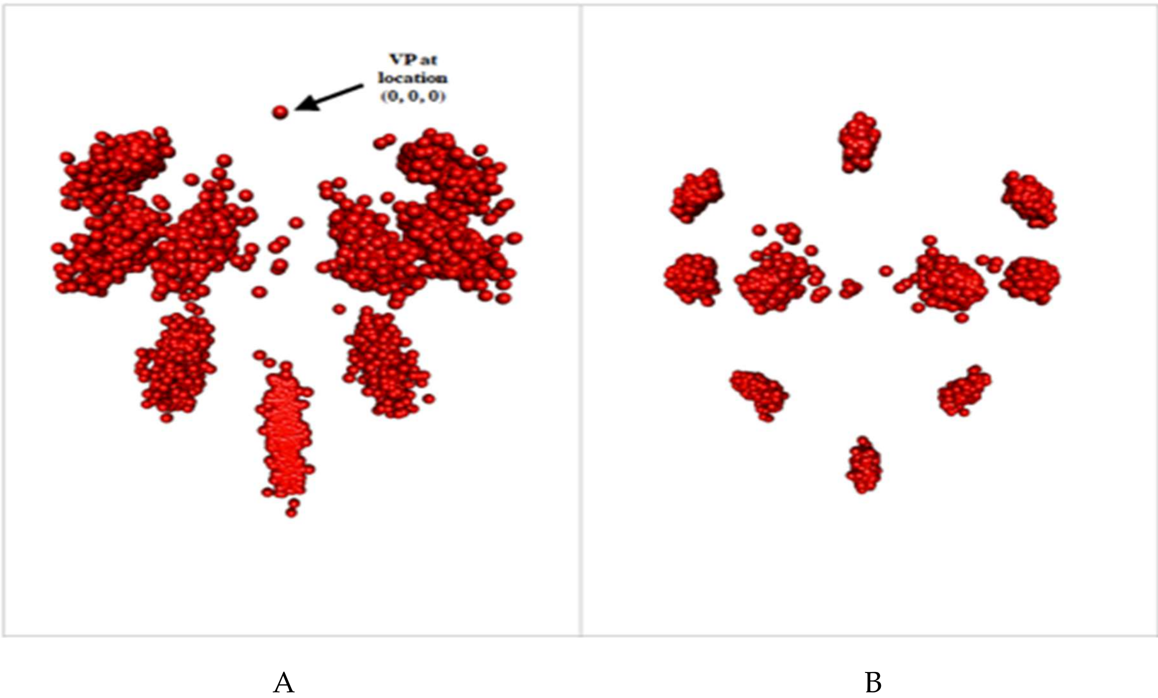

Figure 2. The raw data (**A**) and the translated, rotated and scaled data (**B**) of the non-scoliotic cohort with a main thoracic curve pattern. In Figure 2A, all images have been manipulated such that the VP point is located at coordinates 0,0,0 (marked).

The results of the OPA for the non-scoliotic cohort demonstrating the difference in mean shape between males and females is shown in Figure 3.

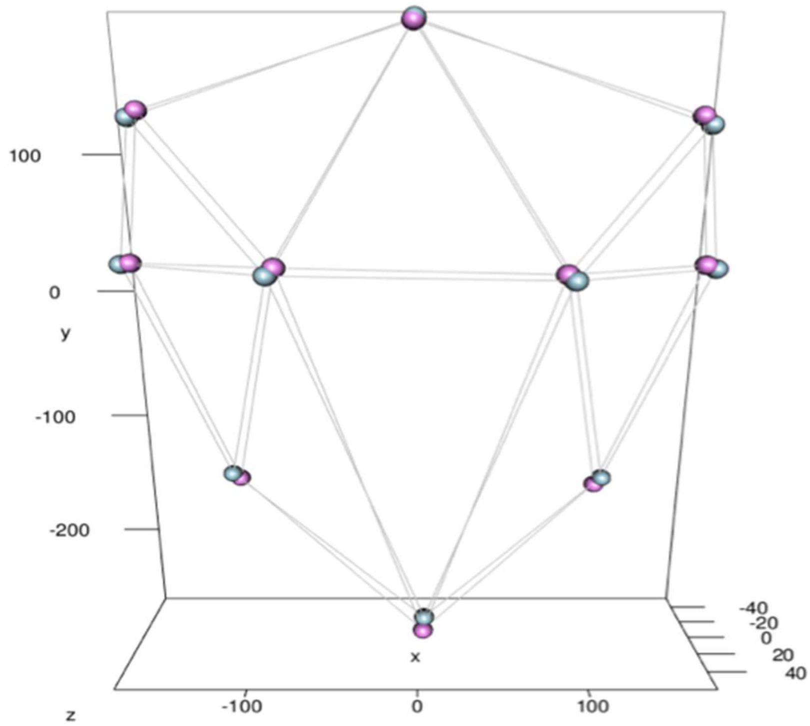

Figure 3. Male (blue) and female (pink) mean shapes following ordinary Procrustes analysis (OPA) in the main thoracic curve pattern of the non-scoliotic cohort.

Plots of the mean shapes with PCA vectors are shown for all of the cohorts for the main thoracic curves (Figure 4A–C) and the thoracolumbar curves (Figure 5A–C). The mean shapes of the non-scoliotic, pre-operative and post-operative cohorts are plotted together and viewed from different angles in Figure 6 (main thoracic) and Figure 7 (main thoracolumbar).

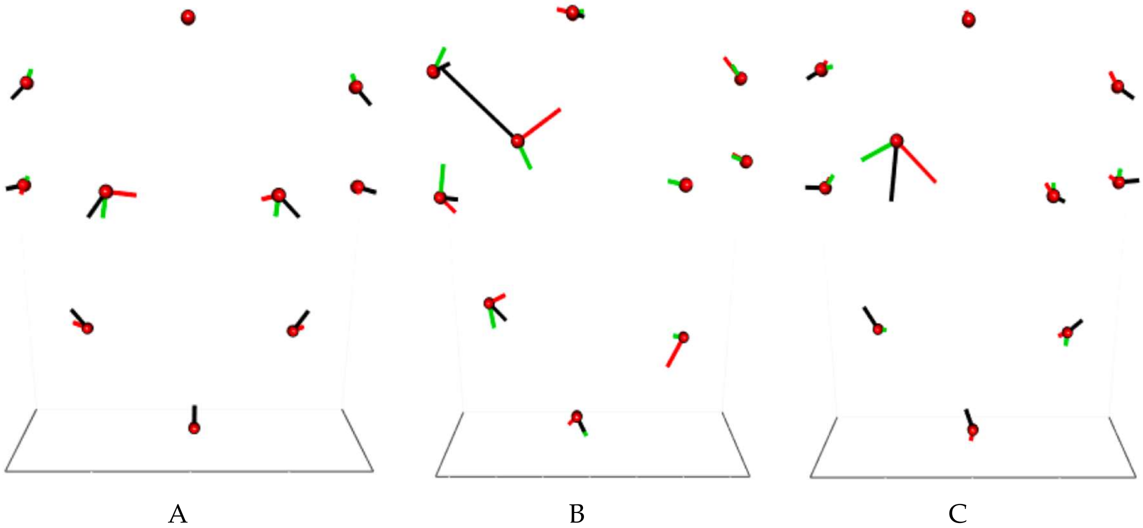

Figure 4. The principal component analysis (PCA) plots for the nonscoliotic (**A**), pre-operative (**B**) and post-operative (**C**) for the convex to the right main thoracic group. Black equals the vector of the first principal component (PC1), red equals PC2 and green equals PC3.

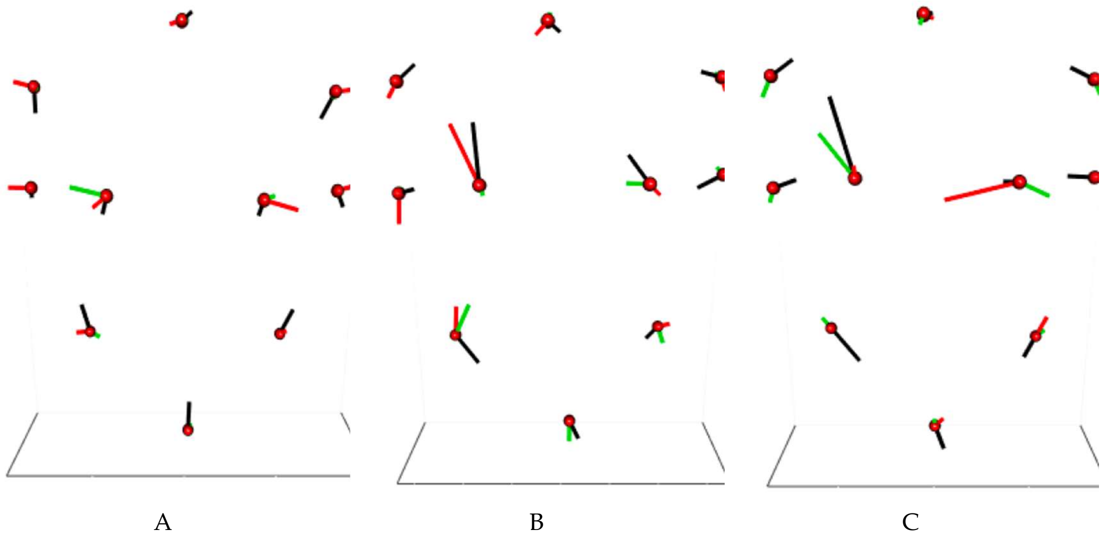

Figure 5. The PCA plots for the non-scoliotic (**A**), pre-operative (**B**) and post-operative (**C**) for the convex to the left main thoracolumbar group. The colours are as for Figure 4.

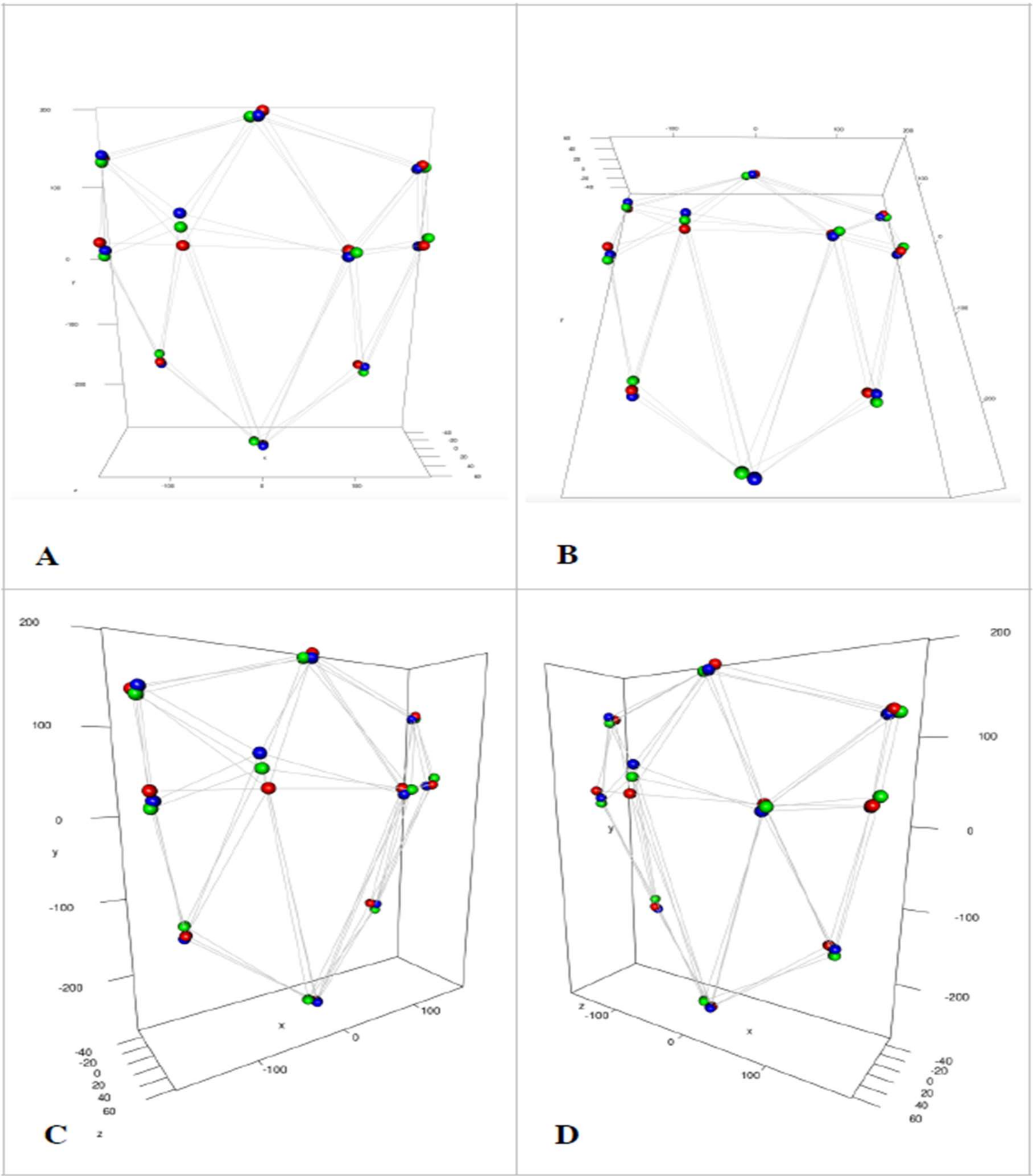

Figure 6. The mean shape plots of the non-scoliotic (red), pre-operative (green) and post-operative (blue) data for the convex to the right main thoracic curve type. (**A**) Posterior view, (**B**) Adams forward bend view, (**C**) left rotated view, (**D**) right rotated view.

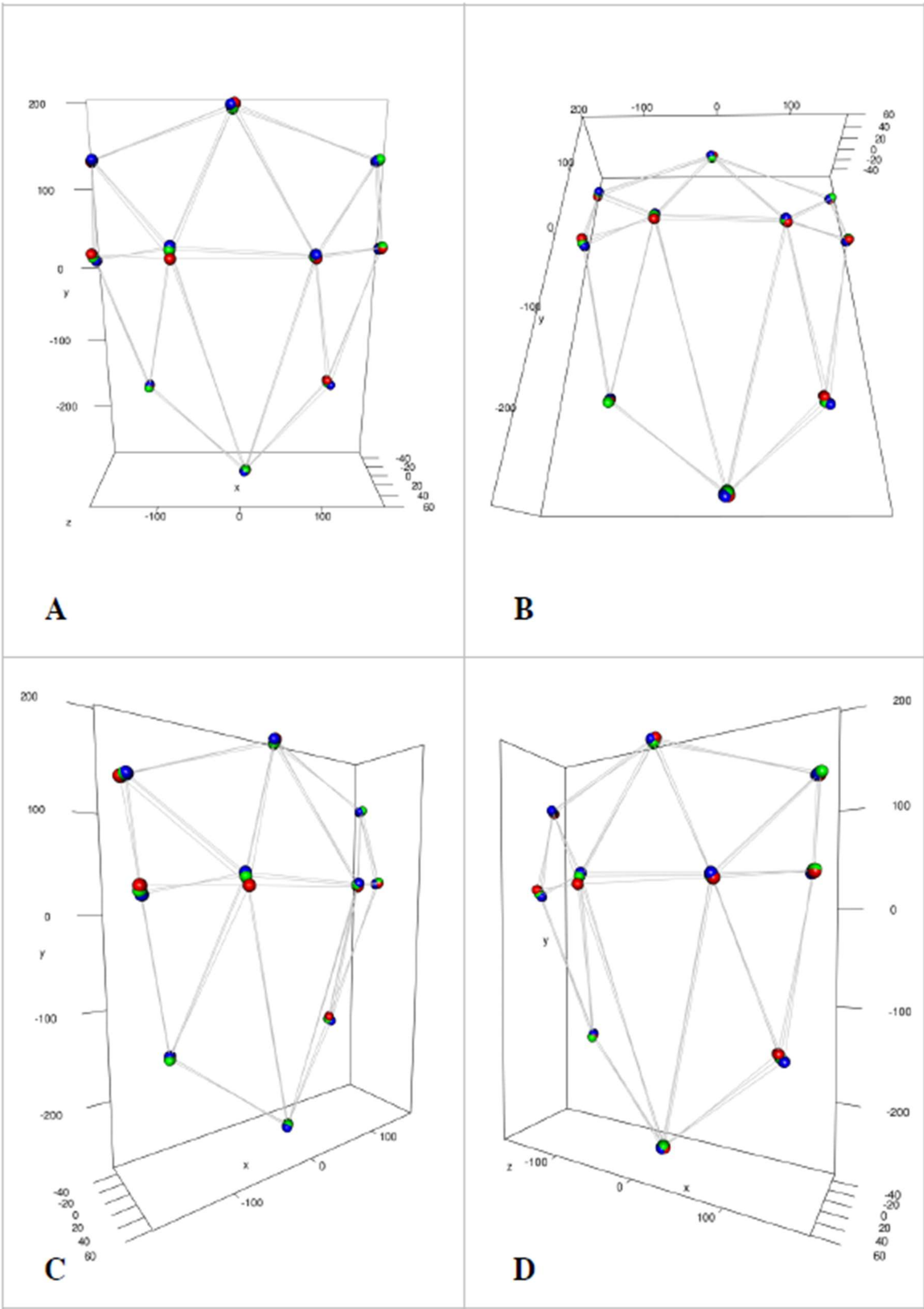

Figure 7. The mean shape plots of the non-scoliotic (red), pre-operative (green) and post-operative (blue) data for the convex to the left main thoracolumbar curve type. (**A**) Posterior view, (**B**) Adams forward bend view, (**C**) left rotated view, (**D**) right rotated view.

The mean shape of the non-scoliotic data (Figures 6 and 7—red spheres) shows that, for both the main thoracic and main thoracolumbar curve patterns, there is little asymmetry seen. This is confirmed with the PCA analysis, where the axes of variability are in similar positions and of similar length (mirrored) between the two sides (Figures 4A and 5A).

There were statistically significant differences between males and females in the non-scoliotic cohort by all three statistical tests ($p < 0.05$). However, as can be seen in Figure 3, there is little visual difference between the shapes of the males and the females, and these differences would not be within the range regarded by Akel et al. as clinically significant [16]. For the scoliotic cohort, the small numbers of males when compared to the number of females made the analysis by sex unreliable. Thus, following the methodology that has already been established in the literature by Gardner et al. [5], further analysis was performed independent of sex.

Using the plots of the mean shape, in the pre-operative main thoracic group, a difference is seen compared to the non-scoliotic cohort in the position of the most prominent points on the back (Figure 6). This is seen in both the y and z dimensions with the left point being higher (y) and less prominent (z) than the corresponding right point. Whilst the mean shape is similar for the other landmarks, the PCA plots demonstrate the variability seen (Figure 4A,B). Following surgery, the asymmetry in height increases but the difference in prominence decreases (Figures 4A,C and 6).

In the main thoracolumbar group, pre-operatively, the asymmetry seen is less than that of the main thoracic group (Figure 7). Surgery changes the asymmetry seen, particularly in the position of the most prominent points, but to a lesser amount than in the main thoracic group (Figure 7). Again, the variability is reflected in the PCA plots (Figure 5).

Using all three statistical tests (Hotelling [11], Goodall [12] and James [13]), there was a statistically significant change ($p < 0.001$) between the non-scoliotic and pre-operative data, non-scoliotic and post-operative data and the pre-operative and post-operative data in both main thoracic and main thoracolumbar curve types.

The possible errors in measurement are twofold: first, that from the interaction with the subject and second, from the subsequent identification of landmarks. The error that comes from the interaction with the subject is documented in the paper by Berryman et al. [7]. The error from the placement of landmarks were assessed by inter- and intra-rater testing, and all intraclass correlation (one way agreement) values were greater than 0.935.

4. Discussion

Scoliosis is a disorder of the shape of the spine and torso [17]. The visual appearance of the torso to the patient and family is often the trigger for a referral for a specialist opinion [17]. Patients and surgeons are concerned about the spine and the asymmetry of the torso [2,3,18]. Previous literature has identified the variability of the posterior torso shape in both a non-scoliotic cohort [4] and scoliotic cohort [5]. These papers examined the pre-operative and post-operative shape in comparison to the non-scoliotic cohort using data ellipses. These ellipses show that there is a degree of asymmetry in non-scoliotic children. Scoliosis increases the degree of asymmetry, and surgery decreases that asymmetry towards the range seen in the non-scoliotic cohort. However, these ellipses look at particular anatomical areas in isolation rather than the torso as a whole shape.

Procrustes analysis is a method of shape analysis described by Dryden and Mardia [6] that has previously been used in the spine [19] and in scoliosis [20]. The method allows assessment of the whole posterior torso shape in 3D.

For both non-scoliotic and scoliotic cohorts, the subjects were sub-divided to allow for different curve types [8]. This again followed previous methodology [5] and was to ensure that any variability of shape seen between different curve types was identified. In the non-scoliotic cohort, there was a statistically significant but clinically insignificant difference in the mean shapes between males and females [16]. Whilst the PCA analysis shows what is responsible for this statistical difference (as the spread of the data around each landmark is different in size and direction), there is minimal asymmetry between the left and right sides of the torso. Given the small numbers of males in the scoliotic cohort, and following the methodology of previous work [5], further analysis was performed without sub-dividing for sex. The results of the analysis of shape in the non-scoliotic cohort provide reference data of normality for the future.

As expected from the work of Lenke [8], in the scoliotic cohort, there were many more subjects with a main thoracic curve type compared to a main thoracolumbar curve type. In the main thoracic curve group, the most obvious asymmetry in both the mean shape and the PCA analysis is of the most prominent points over the back. This is not seen to the same degree in the main thoracolumbar group. This would be expected given the anatomical proximity of the scoliotic curve to the most prominent points in the main thoracic group as the most prominent points of the back are invariably seen over the scapulae; however, the points do not represent the underlying bony anatomy. They do, however, represent what the patient will see as part of their rib hump. In the main thoracic curve group, the right side is more inferior and more prominent than the left (Figure 6). For the points around the edge of the torso, the amount of change in position is not as much as that for the most prominent points. This is also seen in the main thoracolumbar group (Figure 7).

Surgery not only changes the shape of the spine but also the shape of the torso [21]. One of the aims of a surgical procedure is to minimise any asymmetries in the torso and spine [17]. It is of interest to see that the results of this paper show that surgery increases the difference in the y-axis but decreases the difference in the z-axis of the most prominent points between the left and right sides of the torso. It is not immediately clear why this occurs, but it can be hypothesised that it is caused by a change in the geometry of the underlying bony anatomy such that a different part of the scapula becomes more prominent. The scapula is a flat bone lying superficial to the thoracic cage and is only attached to the axial skeleton via the acromioclavicular joint. Thus, the 3D position of the scapula can be changed through surgery by changing the shape of the underlying thoracic cage, tilting the scapula and making a more superior part of the scapula prominent. The mean post-operative points around the edge of the torso, on the other hand, are in a similar position to the pre-operative and non-scoliotic cohorts.

Statistically, there was a significant change ($p < 0.001$) in 3D shape between the non-scoliotic, pre-operative and post-operative cohorts. This demonstrates the underlying variability of the data, which cannot be seen visually but would be appreciated by imagining the mean shape and PCA plots superimposed.

The effect of surgery is to change the shape of the torso but, in agreement with our previous work [5], there is still a significant amount of variability and difference in the shape of the post-operative cohort compared to the non-scoliotic cohort.

The most prominent points represent just one measure of 3D shape. The 3D coordinates of these points do not describe the total 3D shape of the rib hump. The current analysis does not measure the totality of the rib hump, which can vary in shape and size. Future work will be aimed at the 3D analysis of the rib hump as a shape rather than one 3D point.

This work identifies the 3D shape of the posterior torso in both non-scoliotic and scoliotic cohorts. Scoliosis surgery does change the shape of the torso but to a lesser degree than it changes the spine [21]. This information is of use to surgeons, allowing the planning of a procedure that will result in the least asymmetry.

The importance of this work is, first, in establishing a methodology that can be used to assess the 3D shape of the torso. Second, using this methodology, the 3D shape of the torso has been demonstrated in a non-scoliotic cohort and both a pre-operative and post-operative scoliotic cohort. This shows how different scoliotic curves alter the shape of the torso and, in particular, which part of the torso is most affected by scoliosis. This matters as one of the aims of scoliosis surgery is the equalisation of asymmetry. From this work, the conclusion can be drawn that scoliosis surgery does not always reduce, and in some areas actually increases, the asymmetry of the torso. Thus, future developments in scoliosis surgery need to be targeted to deal with this. This work will enable clinicians to understand the effects of scoliosis and scoliosis surgery on the torso such that future developments in surgery are aimed at achieving greater symmetry and less torso deformity.

Author Contributions: Conceptualization, all authors; methodology, F.B.; software, F.B. and P.P.; validation, all authors; formal analysis, A.G.; investigation, A.G.; resources, A.G.; data curation, A.G. and F.B.; writing—original draft preparation, A.G.; writing—review and editing, all authors; supervision, P.P.; project administration, A.G.; funding acquisition, A.G.

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Conflicts of Interest: The authors declare no conflict of interest. The funders had no role in the design of the study; in the collection, analyses, or interpretation of data; in the writing of the manuscript; or in the decision to publish the results.

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The Development of Kyphosis and Lordosis in the Growing Spine

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Study Design. A longitudinal cohort study.

Objective. The aim of this study was to do the analysis of the development of kyphosis and lordosis in the growing spine.

Summary of Background Data. Previous studies have measured kyphosis and lordosis in different ways with differing techniques. None of the previous literature has a truly longitudinal design and there is disagreement as to whether there exists a difference between the development of kyphosis and lordosis between males and females.

Methods. Repeated measures using Integrated Shape Imaging System Integrated Shape Imaging System 2 surface topography over 5 years of a group of children aged 5 to 16 years without spinal deformity. Longitudinal analysis was performed using linear mixed effects modeling.

Results. There were 638 measures in 194 children. Both kyphosis and lordosis increased with age in both males and females ($P < 0.001$ for kyphosis and $P = 0.002$ for lordosis). There was no statistical difference in the development of kyphosis between males and females ($P = 0.149$). However, there was a significant difference in lordosis between males and females ($P < 0.001$) with female lordosis larger than that seen in males. Kyphosis and lordosis increased in a nonlinear fashion with age.

Conclusion. Kyphosis and lordosis increase as children age. Between males and females there is no difference in the increase in the size of kyphosis, but there is difference in the size of lordosis with females having greater lordosis versus males at the same age.

Key words: female, growth, ISIS2, kyphosis, linear mixed effect modeling, LOESS regression, longitudinal, lordosis, male, scoliosis, surface topography.

Level of Evidence: 2

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Scoliosis and Spinal Disorders

RESEARCH Open Access

Do the SRS-22 self-image and mental health domain scores reflect the degree of asymmetry of the back in adolescent idiopathic scoliosis?

James Cheshire[1*], Adrian Gardner[2,3], Fiona Berryman[2] and Paul Pynsent[3]

Abstract

Background: Patient-reported outcomes are becoming increasingly recognised in the management of patients with adolescent idiopathic scoliosis (AIS). Integrated Shape Imaging System 2 (ISIS2) surface topography is a validated tool to assess AIS. Previous studies have failed to demonstrate strong correlations between AIS and patient-reported outcomes highlighting the need for additional objective surface parameters to define the deformities associated with AIS. The aim of this study was to examine whether the Scoliosis Research Society-22 (SRS-22) outcome questionnaire reflects the degree of measurable external asymmetry of the back in AIS and thus is a measure of patient outcome for external appearance.

Methods: A total of 102 pre-operative AIS patients were identified retrospectively. Objective parameters were measured using ISIS2 surface topography. The associations between these parameters and the self-image and mental health domains of the SRS-22 questionnaire were investigated using correlation coefficients.

Results: All correlations between the parameters of asymmetry and SRS-22 self-image score were of weak strength. Similarly, all correlations between the parameters of asymmetry and SRS-22 mental health score were of weak strength.

Conclusion: The SRS-22 mental health and self-image domains correlate poorly with external measures of deformity. This demonstrates that the assessment of mental health and self-image by the SRS-22 has little to do with external torso shape. Whilst the SRS-22 assesses the patient as a whole, it provides little information about objective measures of deformity over which a surgeon has control.

Keywords: Adolescent idiopathic scoliosis (AIS), Surface topography, Scoliosis Research Society-22 (SRS-22), Patient-reported outcomes, Health-related quality of life (HRQOL), ISIS2

Background

Adolescent idiopathic scoliosis (AIS) is a three-dimensional deformity of the spine typically associated with a range of torso abnormalities including rib and scapula prominences, asymmetry of the shoulders, chest wall deformity and waist asymmetry [1].

Correction of visible deformity is increasingly becoming recognised as an important indication for surgical intervention [2] with one of the goals of surgery being to improve both physical health and health-related quality of life (HRQOL) [3]. Both AIS patients and their parents have associated aesthetic concerns [4, 5], with reduction of visible deformity found to be the second most common reason for patients requesting surgical intervention [5].

In light of the increasing recognition and importance of patient-reported outcomes, attempts have been made to develop objective measures to address patient's HRQOL. One questionnaire by the Scoliosis Research Society (SRS), the SRS-22 [6], has been validated in pre-operative AIS and adult scoliosis patients and has been shown to have excellent internal consistency and reliability [7–9].

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It is established that patients with AIS suffer from reduced HRQOL, often experiencing more pain, impaired function, lower self-esteem and increased rates of depression than their contemporaries [10–13]. A review by Rushton and Grevitt found that, compared to unaffected peers, patients with AIS had statistically worse pain and poorer self-image [14]. Of these SRS domains, self-image was the only one found to be consistently worse clinically.

The traditional measurement for quantifying spinal deformity is the Cobb angle [15], which is a measurement of the size of the curve in the spine in the coronal plane measured on a posterior-anterior radiograph. This measurement assesses spinal deformity in a two-dimensional uni-planar manner. Due to the three-dimensional nature of the deformity in AIS, the use of the Cobb angle has drawbacks and fails to take into account patients' perceptions of their deformity. Furthermore, several studies have demonstrated that radiological parameters do not correlate well with patients' subjective perception of body image [1, 16–18]. For this reason, it is increasingly recognised that in addition to radiological measurements, supplementary outcome measures are required to better quantify the deformity [16].

Over the years, new modes of assessing deformity have been developed. Surface topography is one such method allowing a non-invasive, three-dimensional assessment of the surface of the back or torso to be performed, and it has been well validated for assessing spinal deformity in scoliosis [19–23]. Several studies have demonstrated moderate correlation between surface topography and the SRS-22 scores specifically in the self-image and mental health domains [8, 23, 24]. Despite demonstrating these correlations, Brewer et al. [24] concluded that the patients' view of deformity may be related to other factors that were not fully assessed by their current methodology, highlighting a need to determine additional objective parameters that would better correlate with the patients' perceptions of their condition.

When attempting to define these additional parameters, reference was made to previous work demonstrating that the shoulder balance, scapula prominence and waistline asymmetry are the most important factors that contribute to overall trunk deformity in AIS patients [25–27].

The purpose of the study was to analyse how well the SRS-22 domains of mental health and self-image reflect the objective parameters of asymmetry measured using the Integrated Shape Imaging System 2 (ISIS2) surface topography system. The overriding aim was to assess whether the SRS-22 questionnaire reflects the measured trunk deformity in areas known to be of concern in AIS and that the surgeon has the opportunity to influence during surgery.

Methods

This study retrospectively identified 102 pre-operative patients with previously untreated AIS. Patients between 10 and 18 years of age were included. Patients undergoing conservative management with bracing were excluded from the study. The patient cohort was a consecutive series of patients presenting to the spinal clinic at our institution that met the inclusion/exclusion criteria. Each patient had undergone clinical assessment and surface topography using ISIS2 within 6 weeks of completing the SRS-22 questionnaire (mean difference 1 day, SD 6 days, range 0–41 days). Available spinal radiographs were only considered to be appropriate for assessment if taken within 6 weeks of the ISIS2 scan. All patients had undergone a whole spine MRI confirming a diagnosis of idiopathic scoliosis, as is standard practice at our institution. Prior ethical approval was gained (15/EM/0283) through the national ethical approval process.

A perfectly symmetrical back is one without difference between the right and left side of the body. Noting the importance of shoulder balance, scapula prominence and waistline asymmetry [24–26], the following parameters were chosen for use in our study.

The parameters 'AxDiffOff' for the axilla and 'WaistDiffOff' for the waist describe the difference (right minus left) in the distances from the midline for points marking the proximal end of the posterior axillary fold and the most medial part of the flank for the waist as shown in Fig. 1. A positive number indicates that the right side had a larger offset than the left. The parameters 'ShDiffHt', AxDiffHt and WaistDiffHt describe the difference (right minus left) in the relative heights of the shoulders, axillae and waist in a similar fashion. A positive number indicates that the right side was higher than the left. The parameters AxDiffOff, WaistDiffOff, ShDiffHt, AxDiffHt and WaistDiffHt were all measured from a two-dimensional photograph. The point used for the shoulder in ShDiffHt is marked from a vertical line from the axillary point as that line crosses over the edge of the shoulder girdle.

The three-dimensional aspect of ISIS2 is defined using volumetric asymmetry. The methodology for this parameter is as follows. Markers are placed on the bony landmarks of the spine and lumbar dimples so that the three-dimensional surface of the back can be related to body axes. A zero plane is defined through the sacrum and the vertebra prominens, parallel to the line running between the markers on the lumbar dimples. A curve is fitted through the markers on the spinous processes on the measured surface and is then used as the axis of symmetry. The difference between the areas of the back surface above the zero plane on each side of the symmetry line is then calculated for each transverse (horizontal) section and allocated to the higher side. The left

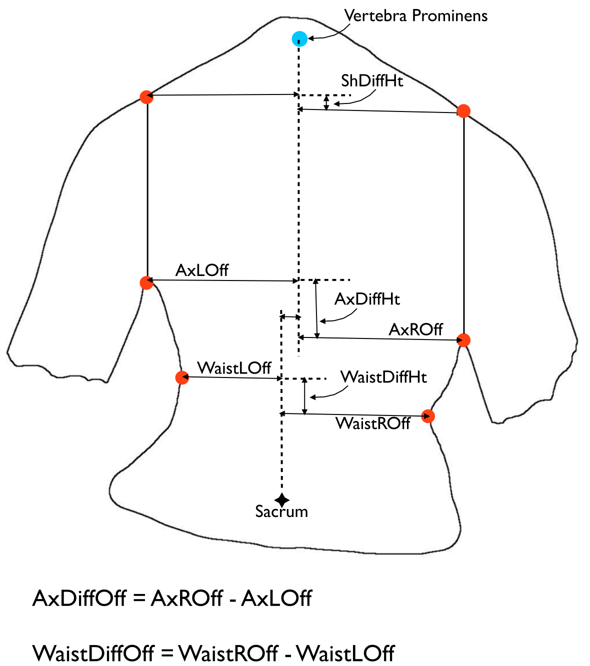

Fig. 1 A schematic illustrating asymmetry parameters as measured from a two-dimensional photograph. Reproduced with permission from Gardner et al. [45]

and right volumetric asymmetry parameters are then calculated by summing the area differences on each side and normalising for back length, as shown in Fig. 2. The parameters 'VolL' and 'VolR' give objective values for the size (volume) of any rib or lumbar humps seen on the back. A new parameter 'VolSum' is defined as the sum of VolL and VolR. 'VolDiff' is defined as the difference of VolR minus VolL. These parameters give a measure of the total amount of asymmetry (right and left together) and the difference in the asymmetry between the two sides. An additional parameter 'ZScapDiff' is defined as the difference in magnitude between the maximum point (maximum height away from the zero plane) in the left and right scapular areas. These parameters give a measure of the three-dimensional asymmetry of the back.

Modifications were coded adding to the standard ISIS2 user interface to allow the user to locate the positions of the waist creases, axillae and shoulders by identifying these points with the mouse. The remaining parameters based on the standard ISIS2 parameters were calculated automatically as normal [21]. The analysis was carried out by a single researcher (AG) on the new two-dimensional parameters based on the manual identification of the waist, axilla and shoulder locations. The magnitudes of the radiographic spinal curves were measured using the Cobb angle method by the treating surgeon using Picture Archiving and Communication System software (GE Systems, New York, NY, USA).

The relationships between the scores for the SRS-22 self-image and mental health domains and the surface topography parameters were investigated using either the Pearson correlation coefficient (r) or Spearman's rank correlation coefficient depending on distribution of data type. R software was used for all data analysis [28]. The strength of correlation is defined as 0–0.29 is weak, 0.3–0.69 is moderate and 0.7–1.0 is strong [29]. Statistical significance was set at $p < 0.05$.

Results

Of the 102 patients included in the study, six (5.9%) were males and 96 (94.1%) females. The mean age of the patients at time of assessment was 14.3 years (standard deviation 1.29 years, range 11.32–17.6 years). Of the 102 patients, only 54 had an appropriate accompanying radiograph. There were 39 patients with Lenke type 1 curves, 13 with Lenke type 3 curves and two with Lenke type 5 curves.

Median total SRS score was 3.30 (interquartile range 2.91–3.82); median self-image score was 2.65 (interquartile range 2.20–3.15) and median mental health score was 3.38 (interquartile range 2.80–4.00). Median Cobb angle was 66.0° (interquartile range 54.0–74.8°).

Table 1 shows the statistics for the parameters of asymmetry and the SRS-22 questionnaire. All correlations between the parameters of asymmetry and SRS-22 self-image score were of weak strength. Similarly, all correlations between parameters of asymmetry and SRS-22 mental health score were of weak strength. Scatterplots of the SRS-22 self-image and mental health domain scores against parameters of asymmetry were drawn, but none showed a strong relationship. A sample scatterplot for WaistDiffOff and SRS-22 self-image is shown in Fig. 3.

Correlation analysis was also carried out on the Lenke 1 subgroup. The results were similar to the whole group, with all measured correlations being of weak strength. Analysis was not done on the Lenke 3 and 5 subgroups because of the low numbers.

Discussion

It is well established that patients with untreated AIS tend to suffer a reduced HRQOL often experiencing increased pain, impaired day to day function, lower self-image and self-esteem and increased rates of depression than their contemporaries [10–13]. The need to consider HRQOL when deciding treatment strategy is becoming increasingly recognised among clinicians [2] with one of the main goals of surgery now to improve both physical health and HRQOL.

There has been an increasing use of disease-specific, patient-reported questionnaires such as the SRS-22, the Spinal Appearance Questionnaire (SAQ) [30] and the Trunk Appearance Perception Scale (TAPS) [31], to help

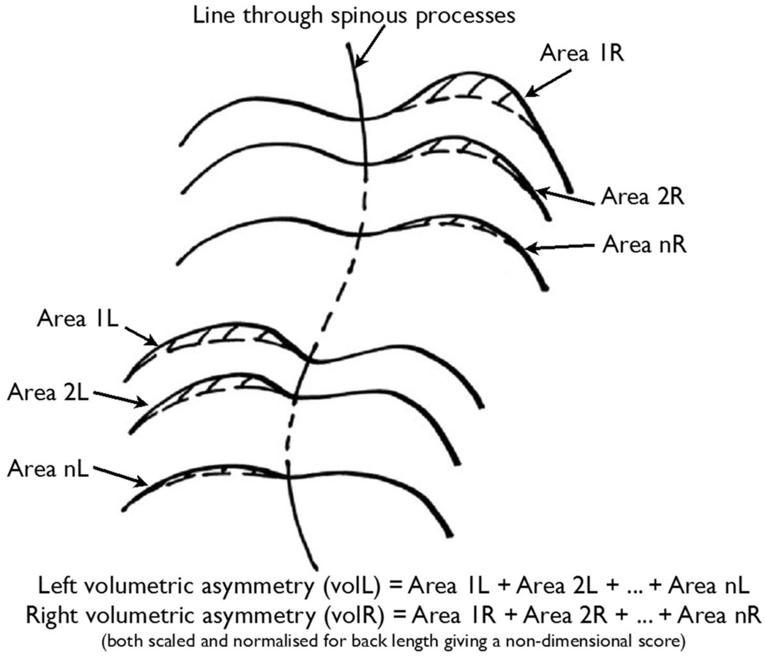

Fig. 2 An illustration of how volumetric asymmetry is calculated

clinicians assess a patient's HRQOL and decide on the most suitable management. Furthermore, questionnaires also allow clinicians to assess the impact of a specific management strategy.

Despite Cobb angle being the traditionally accepted standard for measuring the size of a scoliotic curve [15], Brewer et al. [24] demonstrated that volumetric asymmetry correlated better than the Cobb angle with the self-image and mental health domains of the SRS-22 questionnaire. This is not unexpected. Goldberg et al. in their paper of 2001 stated "it is the rib hump that the patient is unhappy with, not the value of the Cobb angle" [20]. The measurement of volumetric asymmetry enables clinicians to better address patient perceptions of their own deformity and in turn goes some way in understanding the psychological impact the resultant deformity has in AIS [11, 13].

Whilst the Brewer et al. study [24] demonstrated better correlation of the SRS-22 self-image and mental health domains with a volumetric asymmetry parameter from surface topography than with Cobb angle, the correlations were only of a moderate level. The authors concluded that volumetric asymmetry alone, as calculated by surface topography, was insufficient to completely explain a patient's own perception of self-image and mental health in AIS and that additional objective parameters were needed. This led to the development of the anatomical points for the shoulder, axilla and waist as used in this paper as it has been previously demonstrated that shoulder balance, scapula prominence and waistline asymmetry are the most important factors that contribute to overall trunk deformity in AIS patients [25–27]. Using photographic measures to evaluate waistline asymmetry in patients with idiopathic scoliosis, Matamalas et al. [32] demonstrated significant correlation between anatomic landmarks of waistline asymmetry and

Table 1 A table of correlation coefficients and *p* values from parameters of asymmetry compared with Scoliosis Research Society–22 self-image and mental health domains

	Self-image	Mental health
ShDiffHt	$r = 0.06$	$r = 0.01$
	$p = 0.58$	$p = 0.94$
AxDiffHt	$r = -0.16$	$r = -0.21$
	$p = 0.10$	$p = 0.033$
WaistDiffHt	$r = 0.24$	$r = 0.10$
	$p = 0.014$	$p = 0.31$
AxDiffOff	$r = -0.17$	$r = -0.23$
	$p = 0.084$	$p = 0.02$
WaistDiffOff	$r = -0.28$	$r = -0.22$
	$p < 0.01$	$p = 0.027$
VolDiff	$r = -0.26$	$r = -0.13$
	$p < 0.01$	$p = 0.19$
VolSum	$r = -0.22$	$r = -0.09$
	$p = 0.024$	$p = 0.30$
ZScapDiff	$r = -0.21$	$r = -0.15$
	$p = 0.035$	$p = 0.13$

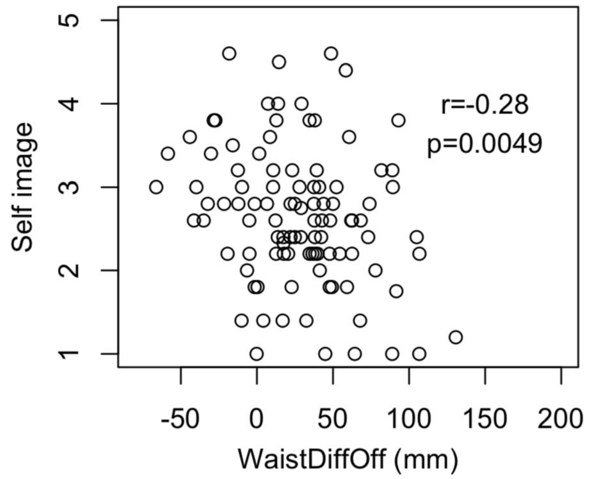

Fig. 3 A scatterplot of WaistDiffOff versus Scoliosis Research Society-22 self-image score

Cobb angle. Furthermore, a significant, yet weak, correlation between clinical measures of waistline asymmetry and the patients' perception of their deformity was demonstrated. Whilst considered a key factor in the perception of trunk deformity in scoliotic patients [25, 27], it has recently been suggested that patients' perceptions of their shoulder deformity do not correspond with clinical measures of shoulder balance. Using clinical photography, Matamalas et al. [33] demonstrated no correlation between clinical measures of shoulder balance and patients' perceptions of their deformity in non-operated scoliotic patients, calling into question the value of shoulder balance in the overall assessment of trunk deformity. Interestingly in a normal study population, Akel et al. [34] found that 28% had a shoulder imbalance greater than 10 mm. However, all of these people perceived themselves as having balanced shoulders. These findings suggest that in the absence of other aspects of trunk deformity shoulder balance goes unnoticed. In the scoliotic population it is possible that the presence of other aspects of trunk deformity may negatively impact their perception of their own shoulder balance.

This paper adds to the literature by demonstrating that the assessment of external deformity in AIS is not well performed when using the SRS-22 scores. Despite the extensive number of parameters of asymmetry used, our study was only able to identify weak correlations with the SRS-22 self-image and mental health domains. This demonstrates that the assessment of mental health and self-image by the SRS-22 seems to have little to do with measurable external torso shape. Whilst the SRS-22 assesses the patient as a whole, it provides little information about objective measures of deformity over which a surgeon has control during a scoliosis operation, one aim of which is to change torso shape.

It was interesting to note that WaistDiffHt and ShDiffHt demonstrated a positive correlation with SRS-22 self-image and mental health domains, although only WaistDiffHt with self-image was statistically significant. One would expect that as the difference in relative heights between the shoulder and waist points increases, the self-image and mental health domain scores would decrease, demonstrating a negative correlation. The significant unexpected positive correlation for WaistDiffHt could possibly be explained by the difficulty encountered whilst identifying the waist in some patients with scoliosis. The waist crease on the concave side is often clear while the waist on the convex side is not. The ability of surgeons to reliably determine waist and shoulder asymmetry in scoliotic patients has been shown to be poor [26]. It should be noted that all correlations measured here were of weak strength whether in the positive or negative directions.

The SAQ [30, 35], TAPS [31] and SRS-22 [7–9] have all been validated in AIS, with the SAQ validated for use with surface topography [23]. Despite the robustness of the SRS-22, it has been shown to have weak to moderate correlation with scoliosis magnitude measured using the Cobb angle [36]. Bago et al. demonstrated that this problem could be overcome by adding dimensions from a pictorial scale to improve correlation with scoliosis curve magnitude [37]. Both the SAQ and TAPS are pictorial questionnaires with their designs previously described [30, 31, 35]. Whilst both the SRS-22 and SAQ have been identified as having significant floor and ceiling effects limiting their ability to detect change [38, 39], the TAPS questionnaire offers an alternative and has been shown to have lower floor and ceiling effects [31].

No studies are known to have used surface topography to directly compare which questionnaires correlate better with HRQOL in AIS. Several studies have, however, used Cobb angle to do this [40, 41]. Matamalas et al. compared three questionnaires; SRS-22, SAQ and TAPS in idiopathic scoliosis [41]. The study found that all questionnaires demonstrated good internal consistency and correlation with scoliosis magnitude. SAQ and TAPS demonstrated the strongest correlation with each other ($r = -0.8$) whilst SRS-22 demonstrated medium strength correlation with SAQ ($r = -0.67$) and TAPS ($r = 0.46$). This finding suggests that pictorial scales such as the SAQ and TAPS might assess different constructs within body image. Both SAQ and TAPS correlated better with Cobb angle compared to SRS-22 self-image ($r = 0.61$, $r = 0.62$ vs. $r = -0.41$ respectively). Specifically, in younger age groups, there was a lack of correlation between the SRS-22 and Cobb angle, thus questioning the ability of textual scales to address self-image issues effectively in the young, a finding previously highlighted by Parent et al. [38]. Whilst pictorial scales clearly demonstrated a superior ability to address body image, they also correlated lower with the other HRQOL domains than textual scales. This led the authors to conclude that the concurrent use of both pictorial and textual scales would be best to address patient-reported outcome measures in AIS, a view supported in other reviews [40, 42].

There are several limitations to this study. Firstly, both its retrospective nature and method of patient sample selection have inherent shortcomings in terms of study design. Our cohort was a consecutive series of patients presenting to our institution's spinal clinic. We acknowledge that obtaining a random sample of patients would have been preferential and would have reduced any associated sampling bias. In our cohort, the ratio of females to males (16:1) is greater than the quoted sex ratio for AIS in the literature, where a ratio of 10:1 for curves greater than 30° is reported [43]. This bias towards a greater number of females may have caused a distortion of the results as females and males may react differently to the perceived aesthetic effects of their scoliosis [44]. Secondly, study patients may well have had concomitant mental health issues that were not necessarily a result of their scoliosis meaning that we may have been measuring the psychological consequences of other unrelated issues.

Future work should look to repeat the methodology described in this study but employing the concurrent use of the SAQ, TAPS and SRS-22 questionnaires to assess which questionnaire best addresses different facets of patient HRQOL in AIS. Future development of a combined pictorial and textual questionnaire to assess outcome measures in AIS should be considered.

Conclusion

Despite extensive use of surface topography parameters known to be important to patients, only weak correlations to the SRS-22 mental health and self-image domains could be demonstrated. Whilst the SRS-22 assesses the patient as a whole, it provides little information about objective measures of deformity over which a surgeon has control.

Abbreviations

AIS: Adolescent idiopathic scoliosis; HRQOL: Health-related quality of life; ISIS2: Integrated Shape Imaging System 2; SAQ: Spinal Appearance Questionnaire; SRS: Scoliosis Research Society; SRS-22: Scoliosis Research Society-22; TAPS: Trunk Appearance Perception Scale

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Availability of data and materials

The datasets used and/or analysed during the current study are available from the corresponding author on reasonable request.

Authors' contributions

AG, FB and PP made substantial contributions to conception and design of the study. JC, AG and FB were involved in the acquisition of data, its analysis and interpretation of the data. All authors were involved in drafting the manuscript and revising it critically for important intellectual content. All authors read and approved the final manuscript.

Ethics approval and consent to participate

Prior ethical approval was gained from East Midlands – Leicester South Research Ethics Committee (15/EM/0283) on 19 June 2015.

Consent for publication

Not applicable.

Competing interests

The authors declare that they have no competing interests.

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The effects of scoliosis and subsequent surgery on the shape of the torso

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Abstract

Background: Adolescent idiopathic scoliosis (AIS) causes asymmetry of the torso, and this is often the primary concern of patients. Surgery aims to minimise the visual asymmetry. It is not clear how scoliosis makes the torso asymmetric or how scoliosis surgery changes that asymmetry when compared to the distribution of asymmetries seen in a non-scoliotic group of normal controls.

Methods: Surface topography images were captured for a group with AIS both pre-operatively and post-operatively. Identifiable points were compared between the images to identify the effects of AIS on the shape of the torso by looking at the relative heights and distances from the midline of the shoulders, axillae and waist in a two-dimensional coronal view. This was then compared to a previously reported group of normal non-scoliotic children to analyse whether surgery recreated normality.

Results: There were 172 pairs of images with 164 females and 8 males, mean age at pre-operative scan of 13.7 years. The normal group was 642 images (237 females and 405 males) from 116 males and 79 females, mean age of 12.5 years.
The curve patterns seen in the scoliotic group matched the patterns of a main thoracic curve ($n = 146$) and main thoracolumbar curve ($n = 26$). The asymmetries seen in both shoulders, axillae and waist were different between the two different types of curve. Across both groups, the shoulder asymmetry was less than that of the corresponding axillae. There was a statistically significant reduction in all asymmetries following surgery in the main thoracic group ($p < 0.001$). This was not seen in the main thoracolumbar group, thought to be due to the small sample size. In the main thoracic group, there were statistically significant differences in the asymmetries between the post-operative and normal groups in the shoulders and axillae ($p < 0.001$) but not the waist.

Conclusions: This paper demonstrates quantitatively the range of asymmetries seen in the AIS torso and the degree to which surgery alters them. Surgery does not recreate normality but does cause a statistically significant change in torso shape towards that seen in a non-scoliotic group.

Keywords: Scoliosis, Surface topography, Surgery, Shoulders, Axillae, Waist, Normal, ISIS2

Background

Within the clinical presentation of adolescent idiopathic scoliosis (AIS), it is common for concern to be raised by both patients and parents around visible asymmetry of the back [1]. This relates to various features including a difference in the height of the shoulders and axillae, inequality of the waist creases and a prominence of one of the scapulae. One of the goals of surgery for AIS is the equalisation of these asymmetries, which translates into improvement in the patient's self-esteem and life satisfaction [2].

The results of scoliosis surgery are routinely reported as changes in the radiographic Cobb angle [3]. This is a measure of the spinal shape internal to the body rather than the external appearance. There is inherent difficulty in using radiographs as a way of measuring areas and shapes within the body comprised of soft tissue rather than bone. Serial radiography also comes with the price tag of a cumulative radiation dose to the body [4]. Surface topography has been developed as a non-radiation

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method of documenting the three-dimensional shape of the back. The Integrated Shape Imaging System (ISIS) [5] is now in its second version (ISIS2) [6]. The system analyses a digital photograph of the child's back which has horizontal lines projected on to it. Fourier transform profilometry is used to create a surface for analysis. The output gives both quantitative and graphical information on the shape for the back in three-dimensions. The use of ISIS2 has been reported previously [6–8].

This paper documents the variability of the relative height of the shoulders, axillae and waist, and also the distance from the midline of the axillae and waist in a group of patients with AIS both pre-operatively and post-operatively. The post-operative values are then compared to previously established normative values for non-scoliotic children [9].

Methods

Ethical and research governance approval has been obtained for both groups in this study from the NRES committee West Midlands—South Birmingham (11/H1207/10) and the NRES committee East Midlands—Northampton (15/EM/0283).

This analysis is a comparison of two groups. The first is a group of children with AIS who, as part of standard care, have surface topography (ISIS2) measured both before and after surgery as a paired set of images. The second is a group of non-scoliotic children who are part of a longitudinal data collection of surface shape measured using ISIS2 and has been reported on previously [9]. Torso parameters were identified in both groups which were then compared.

All of the scoliotic group had an MRI scan of the whole spine as part of their routine care. Children with neural axis anomalies or other abnormal findings have been excluded from this analysis. None of the study group has been treated in a brace as part of their care. For the majority of subjects, surgery was undertaken using modern posterior based pedicle screw techniques ($n = 98$). An anterior release was used in selected cases for a large stiff curve ($n = 63$). Anterior-only surgery was used selectively for main thoracolumbar curve patterns in the absence of a large compensatory thoracic curve ($n = 11$).

All images in the study were acquired using ISIS2. The degree of spinal curvature in the coronal plane (a two-dimensional measure) was measured with the Lateral Asymmetry parameter from the automated ISIS2 analysis. In this study, a positive number indicated that the scoliosis was convex to the right, and a negative number indicated convex to the left. The ISIS2 images were analysed to find the two dimensional torso points that identify the position of the axillae, shoulders and waist. The axillae points were the most superior points of the posterior axillary folds. The shoulder points were at the superior edge of the torso along a vertical line from the axillae points [10]. The waist points identified were the 'minimal waist' [11], which corresponds to the narrowest waist and is the most suitable definition of the waist in a scoliotic population.

The positions of the points were then processed to create parameters comparing the two sides of the trunk against each other, Diff Height for a difference in vertical height and Diff Off for a difference in horizontal distance from the midline. This created the parameters Shoulder Diff Height (ShDiffHt), Axillary Diff Height (AxDiffHt) and Waist Diff Height (WaistDiffHt), Axillary Diff Off (AxDiffOff) and Waist Diff Off (WaistDiffOff) (see Table 1 and Fig. 1). Again, a positive number for the measured torso parameter indicated that the right side was higher than the left (DiffHt parameters) or further from the midline than the left (DiffOff parameters).

The data on the torso points are presented as data ellipses [12], as this clearly represents the bivariate nature of the data [13]. The layouts are displayed in the same way for each plot for the main thoracic (main thoracolumbar) curves. Pre-operative data are in green (dark green), post-operative data in blue (purple) and the non-scoliotic data in red (orange). The mean point is the solid dot in each colour. The ellipse is the 95% confidence interval about the mean in the respective colour.

Table 1 A table of the torso parameter and their definitions as shown pictorially in Fig. 1 [9]

Orientation	Torso parameter	Definition
Vertical measurements	ShDiffHt	The difference in vertical height between the shoulder points
	AxDiffHt	The difference in vertical height between the axillary points
	WaistDiffHt	The difference in vertical height between the waist points
Horizontal measurements	axRoff	The horizontal distance from the midline to the right axillary point
	axLoff	The horizontal distance from the midline to the left axillary point
	waistRoff	The horizontal distance from the midline to the right waist point
	waistLoff	The horizontal distance from the midline to the left waist point
	AxDiffOff	The difference between axRoff and axLoff
	WaistDiffOff	The difference between waistRoff and waistLoff

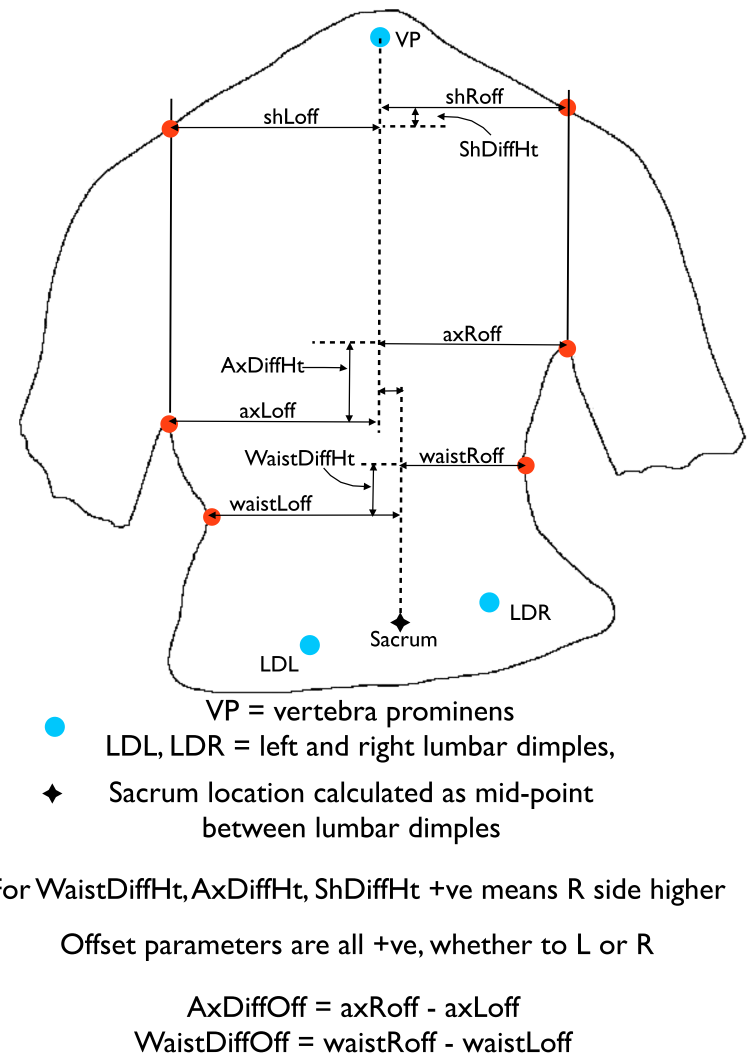

Fig. 1 A diagram demonstrating the anatomical points identified and the measurements from the midline for the shoulder, axilla and waist [9]

In the *x*-axis, a positive number is a curve convex to the right. In the *y*-axis a positive number indicates that the right side is higher, or further from the midline, than the left. The box and whisker plots show the data spread of each individual parameter with the median value as the solid bar within the box, which represents the interquartile range. The whiskers from the box represent 1.5 times the interquartile range. Within the box, the dot is the mean value with the 95% confidence interval of the mean as the bars either side.

As there is a difference in the number of pre- and post-operative cases and that of the non-scoliotic group, propensity matching was performed to confirm that this difference did not affect the results.

All analysis was carried out using R [14]. Comparisons of the data were performed with the *t* test for parametric data and the Wilcoxon rank sum test for non-parametric data. Statistical significance was defined as $p < 0.05$.

Results

The demographic information of both groups is shown in Table 2. In the non-scoliotic group, there have been serial measurements and images captured over 5 years of the same children, with subjects having between 1

Table 2 The demographic information of both groups

	Males	Females	Mean age (years)	SD age (years)	Number of images for analysis
Non-scoliotic	405	237	12.5	1.8	642 individual images
Scoliotic	8	164	13.7 (at pre-operative scan)	1.4	172 pairs of pre-operative and post-operative images

and 5 images taken depending on the length of time they have been in the study. Thus, the number of individual images available for analysis is greater than the number of participants. This group consists of 116 males and 79 females. In the scoliotic group, each subject has a pre-operative and post-operative image giving 172 sets of paired data. Neither the time between the pre-operative image and surgery nor between surgery and the post-operative image was normally distributed. Surgery was a median of 346 days after the pre-operative image (IQR 320 days, range 1 to 1211 days). The median time from surgery to the post-operative image was 200 days (IQR 246 days, range 25–1321 days).

The ethnicity in each group was predominantly Caucasian with smaller numbers of participants with either an Afro-Caribbean or Indian heritage. In the scoliotic group, 11% of the total were not Caucasian. In the non-scoliotic group, 3% of the total were not Caucasian.

In the non-scoliotic group, a small curve in the spine in the coronal plane is seen in nearly all of the participants. The major curve was judged to be proximal thoracic (PT) in 21 subjects. There was no curve seen in eight subjects.

As described previously [9], patterns of curve were used to subdivide the data into a main thoracic group with compensatory thoracolumbar curve and a main thoracolumbar curve with compensatory thoracic curve [15]. In the scoliotic group, the largest subgroup had a main thoracic curve with a smaller number with a main thoracolumbar curve. There were no main PT curves. The numbers in each subdivision are shown in Table 3.

The data in the main thoracic curve group were normally distributed. The data in the main thoracolumbar curve group were not normally distributed. Figures 2, 3, 4, 5 and 6 show the data ellipses for the main thoracic curve with compensatory thoracolumbar curve (mean and 95% confidence interval ellipse) and Figs. 7, 8, 9, 10 and 11 show the data for main thoracolumbar curve with compensatory thoracic curve (median and 95% percentile ellipse). The individual data points for the non-scoliotic group are not presented as they obscure the data points of the pre-operative and post-operative groups.

Tables 4 and 5 show the mean (median) values for the parameters in the pre-operative and post-operative groups. The significance in the change from pre-operative to post-operative is also shown. Tables 6 and 7 compare the mean (median) values of the post-operative group to that of the non-scoliotic group.

Table 3 The number in each subdivision of curve type in each group (PT- Proximal thoracic curve, NC- no curve)

	Main thoracic	Main thoracolumbar	Others
Non-scoliotic	387	227	28 (PT and NC)
Scoliotic	146	26	0

The compensatory curves had no significant difference in effect (see Tables 4 and 5) on the anatomically distant points (for example the effect of the compensatory thoracolumbar curve on the shoulder or axillae points). The waist points and associated trunk imbalance in the main thoracic curve group are due to the effects of the thoracic curve rather than the smaller thoracolumbar curve. This point is further expanded in the 'Discussion' section.

Normalising the data for size of torso did not affect the distributions shown in the analysis. The effect of this analysis using a smaller group of non-scoliotic subjects after propensity matching was not appreciably different so the entire cohort of the non-scoliotic group was kept for the analysis.

Discussion

AIS is a disorder affecting the adolescent spine and is known to come with a 'psychological burden'. There is a dislike of the asymmetry of the torso and overall body shape that presents with a spectrum of symptoms including mental health disorders [16, 17]. One of the aims of scoliosis surgery is to minimise the visible deformity, improving the symmetry of the torso as safely as possible. In a previous paper, using the same methodology as used here, Gardner et al. [9] have reported the range of normality based on two dimensional torso points in non-scoliotic children. This 'normal' group demonstrated that there is a degree of spinal curve in the coronal plane measurable in most children, with differences between the sides of the torso for the shoulder, axillae and waist points. That is, non-scoliotic children are not perfectly symmetrical in the coronal plane and tend to have some spinal curvature, although it is of low magnitude. The data from Gardner et al. [9] acts as a group of normative values to which the AIS group has been referenced.

The AIS group has a larger number of main thoracic curves with compensatory thoracolumbar curves than main thoracolumbar curves with compensatory thoracic curves. This is a similar distribution to that previously reported [15]. The main thoracic curves are mainly convex to the right and an increasing curve is associated with increasing difference between the right and left sides of the torso. The axillae are both more superior (AxDiffHt) and further from the midline (AxDiffOff) on the right in comparison to the left with an increasing scoliosis (Figs. 3 and 4). No effect of an increasing curve on ShDiffHt is seen (Fig. 2). This suggests that the shoulder girdle is compensating for an asymmetry of the underlying torso (demonstrated by the difference in position of the right and left axillae). The independence of

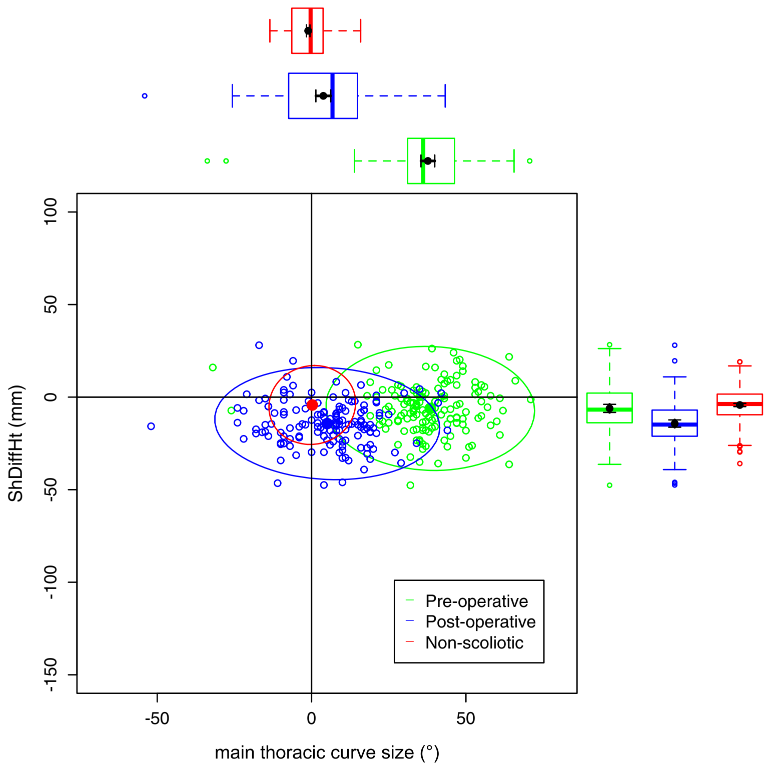

Fig. 2 Data ellipses for the main thoracic curve pattern (main thoracic curve) showing ShDiffHt

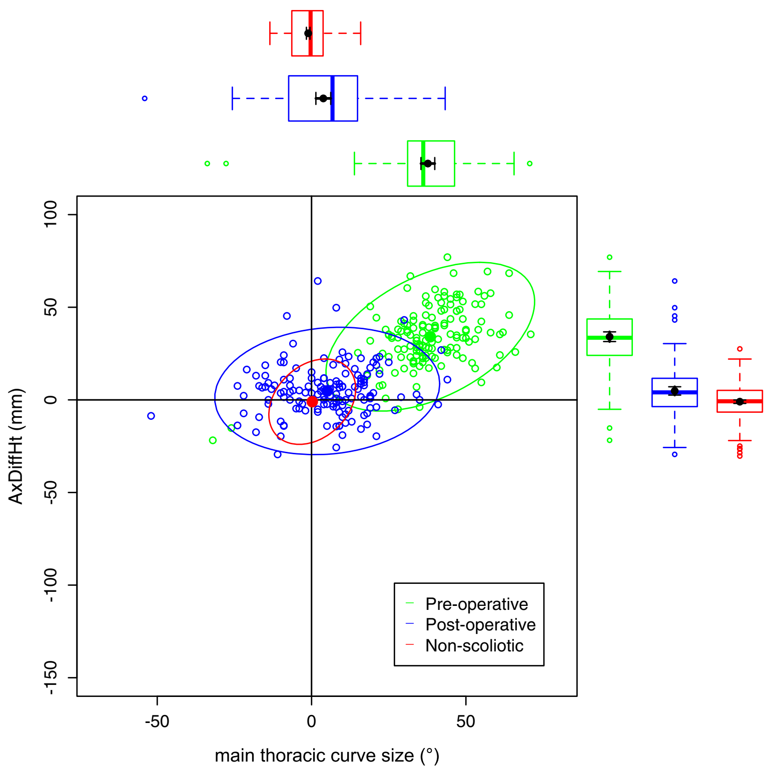

Fig. 3 Data ellipses for the main thoracic curve pattern (main thoracic curve) showing AxDiffHt

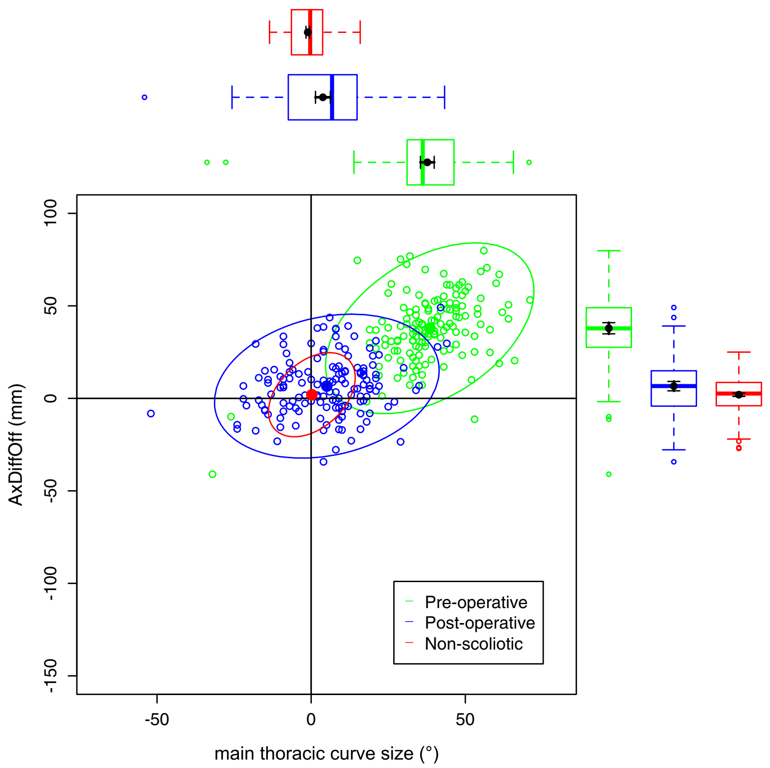

Fig. 4 Data ellipses for the main thoracic curve pattern (main thoracic curve) showing AxDiffOff

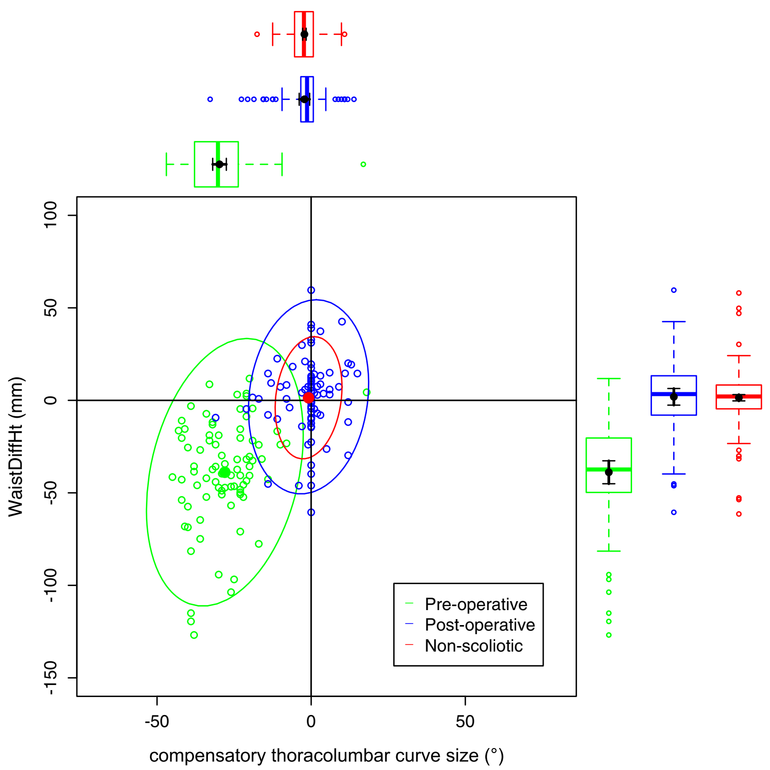

Fig. 5 Data ellipses for the main thoracic curve pattern (compensatory thoracolumbar curve) showing WaistDiffHt

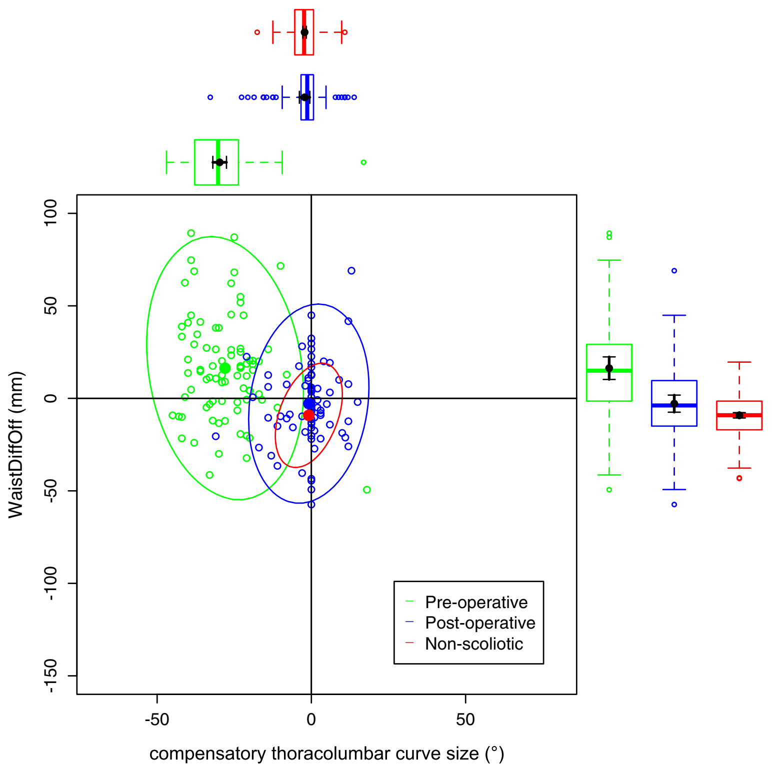

Fig. 6 Data ellipses for the main thoracic curve pattern (compensatory thoracolumbar curve) showing WaistDiffOff

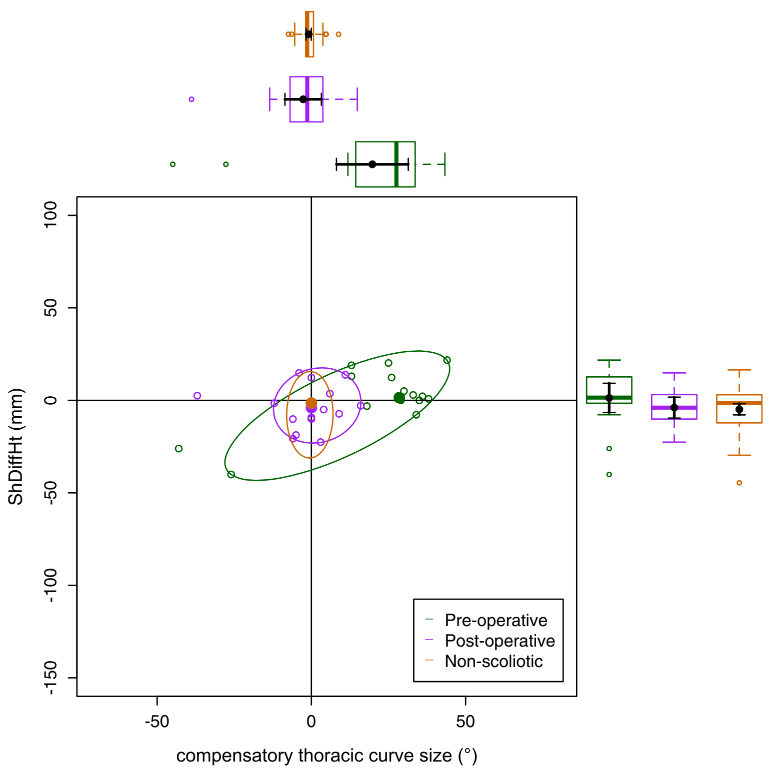

Fig. 7 Data ellipses for the main thoracolumbar curve pattern (compensatory thoracic curve) showing ShDiffHt

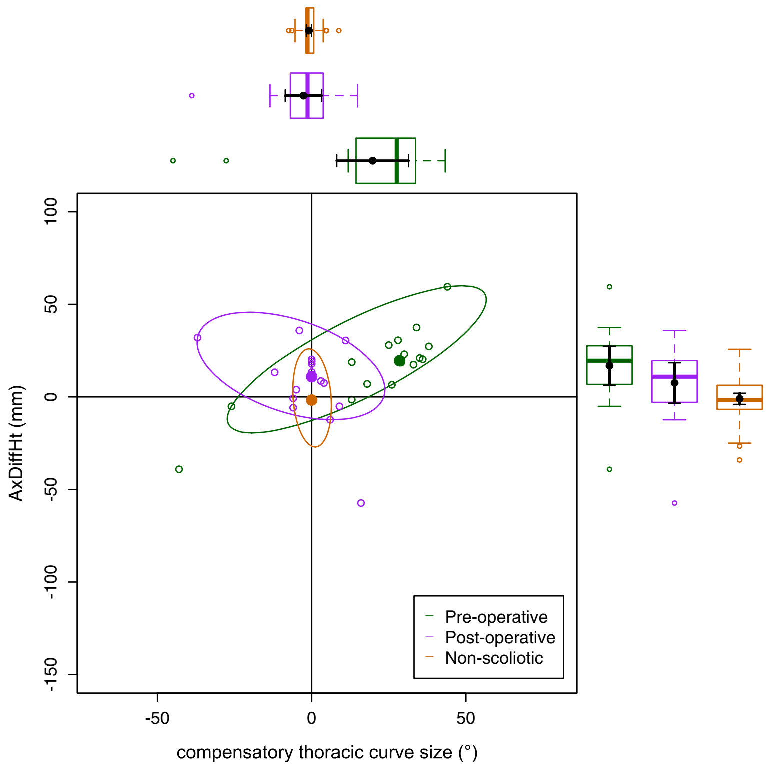

Fig. 8 Data ellipses for the main thoracolumbar curve pattern (compensatory thoracic curve) showing AxDiffHt

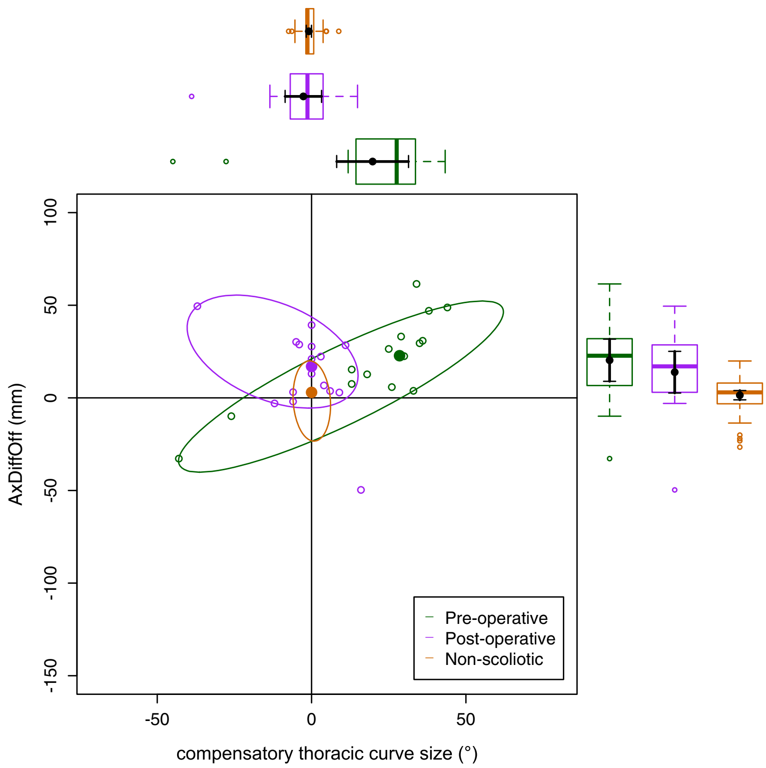

Fig. 9 Data ellipses for the main thoracolumbar curve pattern (compensatory thoracic curve) showing AxDiffOff

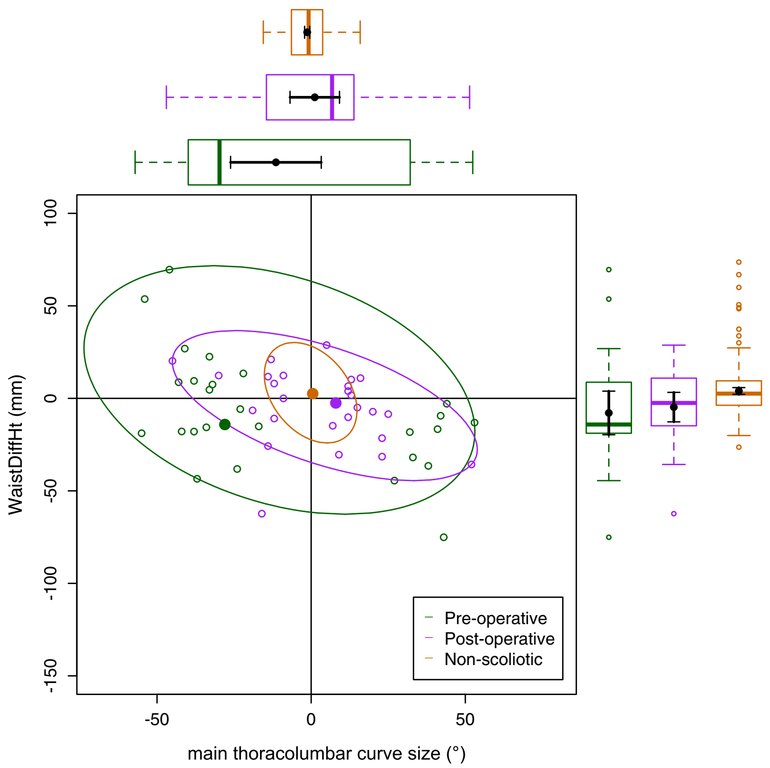

Fig. 10 Data ellipses for the main thoracolumbar curve pattern (main thoracolumbar curve) showing WaistDiffHt

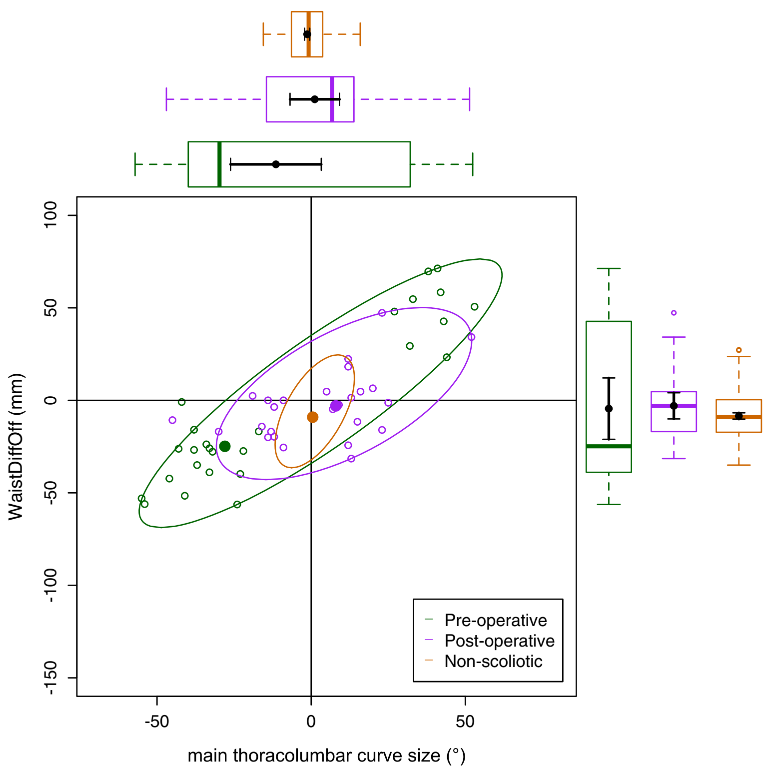

Fig. 11 Data ellipses for the main thoracolumbar curve pattern (main thoracolumbar curve) showing WaistDiffOff

Table 4 A table demonstrating the mean value (and standard deviation) of the parameters measured in the main thoracic pattern for the pre-operative and post-operative scoliotic group with the significance of the change also shown

	Pre-operative	Post-operative	Significance
Curve size (°)	38.5 (13.6)	5.1 (14.6)	< 0.001
ShDiffHt (mm)	−6.1 (13.5)	−14.3 (13.8)	< 0.001
AxDiffHt (mm)	34.1 (16.1)	4.8 (13.8)	< 0.001
WaistDiffHt (mm)	−38.8 (28.6)	1.9 (20.8)	< 0.001
AxDiffOff (mm)	37.9 (18.5)	6.6 (15.6)	< 0.001
WaistDiffOff (mm)	16.3 (28.2)	−2.9 (21.3)	< 0.001

Table 6 A table showing the statistical analysis of the post-operative group for the main thoracic curve group compared to the non-scoliotic group

	Post-operative scoliosis	Non-scoliotic	Significance of difference
ShDiffHt (mm)	−14.3 (13.8)	−4.3 (8.7)	< 0.001
AxDiffHt (mm)	4.8 (13.8)	−1.0 (9.4)	< 0.001
WaistDiffHt (mm)	1.9 (20.8)	1.4 (13.2)	0.838
AxDiffOff (mm)	6.6 (15.6)	2.0 (9.2)	< 0.001
WaistDiffOff (mm)	−2.9 (21.3)	−9.2 (11.1)	0.013

Non-scoliotic data form Gardner et al. [9]

movement of the shoulder girdle relative to the torso may well explain why there is moderate to poor correlation of the intraoperative radiographic features of shoulder position to the post-operative shoulder position [18].

The waist is also increasingly asymmetric with an increasing compensatory thoracolumbar curve. As already stated, with an increasing curve, the axillary points become higher and further from the midline on the same side as the convexity of the curve. However, with the waist points, both DiffHt and DiffOff increase in magnitude but in differing directions to each other (Figs. 5 and 6). The reasons for this are unclear but may represent the difference between the relationship of the waist to the spine and the spine to the shoulder girdle. In thoracolumbar curves, the pelvis is the fixed base on which the spine deforms. In thoracic curves the shoulder girdle moves around the already deformed spine.

The effects of the compensatory curve (a thoracic curve on the waist points or a thoracolumbar curve on the axillae and shoulder points) are less clear, although the main thoracolumbar curve has only a small effect on the shoulder and axilla (Figs. 7, 8 and 9). The effect of the main thoracic curve on the waist is more marked and reflects trunk asymmetry caused by a large thoracic curve (Figs. 5 and 6). The effects of the compensatory curve inferior to this thoracic curve are hidden in the effects of the thoracic curve. This is partly due to the mismatch of curve sizes between the main and compensatory curves, with the main curve exerting a relatively larger effect on the shape of the torso. In the main thoracolumbar curve group, a number had a small compensatory thoracic curve. In this circumstance, the overall curve pattern is known to present primarily with waist asymmetry [19]. This could explain the relationship of a thoracolumbar curve on the shoulder and axillae points suggesting that the small thoracic curve exerts a minimal effect.

Table 5 A table demonstrating the median value (and values of quartile 1 and 3) of the parameters measured in the main thoracolumbar curve pattern for the pre-operative and post-operative scoliotic group with the significance of the change also shown

	Pre-operative	Post-operative	Significance
Curve size (°)	−28.0 (−38.0 to 32.8)	8.0 (−12.8 to 14.5)	0.148
ShDiffHt (mm)	1.5 (−0.8 to 12.5)	−3.9 (−10.9 to 2.8)	0.117
AxDiffHt (mm)	19.6 (6.9 to 27.4)	10.9 (−1.9 to 19.4)	0.044
WaistDiffHt (mm)	−14.1 (−18.7 to 8.4)	−2.5 (−13.9 to 10.7)	0.473
AxDiffOff (mm)	22.8 (7.1 to 31.4)	17.0 (3.1 to 28.5)	0.348
WaistDiffOff (mm)	−24.9 (−37.9 to 39.4)	−3.0 (−16.7 to 4.1)	0.727

The number of patients in the main thoracolumbar group is much smaller compared to the number in the main thoracic group. This is the likely reason for the skewed distribution of the data (Figs. 7, 8, 9, 10 and 11) and supports the decision to use non-parametric statistics to analyse this subgroup. With a greater sample size, it would be reasonable to expect a lessening of the effect of the outliers on the average value and a more uniform distribution allowing the use of the mean and 95% predictive confidence ellipse.

Surgical intervention leads to a statistically significant reduction in the size of the scoliosis in both coronal curve patterns (Tables 4 and 5). In the main thoracic group, this is accompanied by a reduction in the amount

Table 7 A table showing the statistical analysis of the post-operative group for the main thoracolumbar curve group compared to the non-scoliotic group

	post-operative scoliosis	non-scoliotic	significance of difference
ShDiffHt (mm)	−3.9 (−10.9 to 2.8)	−3.9 (8.5)	0.844
AxDiffHt (mm)	10.9 (−1.9 to 19.4)	−2.2 (9.8)	0.004
WaistDiffHt (mm)	−2.5 (13.9 to 10.7)	2.6 (14.0)	0.136
AxDiffOff (mm)	17.0 (3.1 to 28.5)	1.0 (8.4)	< 0.001
WaistDiffOff (mm)	−3.0 (−16.7 to 4.1)	−9.1 (13.1)	0.215

Non-scoliotic data form Gardner et al. [9]

of asymmetry in the torso at the axillae and waist in both DiffOff and DiffHt (Figs. 3, 4, 5 and 6), and this is statistically significant for all parameters. Interestingly, there is a statistically significant increase in the difference between the left and right sides in ShDiffHt (Fig. 2) with the mean value suggesting that the left is more superior than the right following surgery, a worsening of shoulder height asymmetry, for reasons unknown. The difficulties in achieving balanced shoulders in the post-operative patient remain a challenge [20]. It has been shown that the effect of unbalanced shoulders can reduce over time through other compensatory mechanisms [21]. Reviewing the torso as a whole, surgery is successful in reducing the size of the curve and equalising the shape of the posterior torso.

In the main thoracic group, the ellipses show that surgery improves the torso asymmetry towards that seen in the non-scoliotic group (Figs. 2, 3, 4, 5 and 6). There is still a statistically significant difference in the means for shoulder and axillae points between the post-operative group and the non-scoliotic group (Table 6). However, there is no significant difference in waist position between the post-operative and non-scoliotic groups. The change that occurs following scoliosis surgery is towards the range of asymmetries seen in the non-scoliotic group, although surgery does not completely recreate normality. It is worth noting that in all of the parameters, although the average values are similar, the spread of the data is more dispersed in the post-operative group compared to the non-scoliotic group. Whilst scoliosis surgery changes body shape towards a non-scoliotic population, there is still a difference seen. The answer to the question 'does scoliosis surgery recreate normality?' has to be no, but surgery provides a statistically significant change towards a normal shape.

The methodology for the torso points used here is scalar and linear rather than angular as used by Matamalas et al. [22, 23]. The criticism of a non-angular measurement is that it is vulnerable to bias related to differing size between subjects that is not seen in an angular measurement. When all of data presented here was normalised using back length for ShDiffHt, AxDiffHt and WaistDiffHt, axillary width for AxDiffOff or waist width for WaistDiffOff, there were no differences seen in the analysis results and normalisation did not add to the conclusions drawn. Angular measures can be difficult to convert to useful, measurable information in a clinical practice. Linear measures are easy to understand and reproduce and thus are preferred here.

It is noted that the results quoted here represent the position of the torso at the point in time that the post-operative image was taken. With continued growth and then subsequent changes through the ageing process, it is possible that over time, the position described here would change. It would be a valid study to revisit this scoliotic group at 5 years post-surgery to document how the torso has changed over the intervening period.

Conclusion

This work demonstrates the metrics of trunk asymmetry in a scoliotic group and the effects of scoliosis surgery in reducing these asymmetries. Current surgical techniques do not make the spine straight in the coronal plane, nor do they equalise all asymmetries in the trunk. Surgery can make a statistically significant difference to body shape and when compared to a non-scoliotic group does reduce the size of the torso asymmetries towards the shape of the non-scoliotic torso. Future directions for this work will compare this change in body shape with patient-derived measures of their own deformity, such as the Spinal Appearance Questionnaire [24], to examine what the patients feel about their outcomes from surgery, which previously have been noted to be different from what the surgeon feels has been the outcome [25].

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Availability of data and materials
The datasets generated and/or analysed during the current study are not publicly available due to ongoing data collection but are available from the corresponding author on reasonable request.

Authors' contributions
AG carried out the analysis and wrote the paper. FB collected the data and guided the analysis of the data. PP conceived the idea and provided statistical and technical support. All three authors have given final approval for the work to be published and agree to be accountable for all aspects of the work.

Ethics approval and consent to participate
Ethics approval was gained for this study (11/H1207/10 and 15/EM/0283). The images taken of the AIS group were taken as part of routine care, and ethical approval did not require individual consent as long as the data was anonymised.

Consent for publication
Not applicable.

Competing interests
The authors declare that they have no competing interests.

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What is the variability in shoulder, axillae and waist position in a group of adolescents?

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Abstract

The clinical assessment of scoliosis is based on the recognition of asymmetry. It is not clear what the degree of asymmetry is in a population without scoliosis, which could make the differentiation between abnormal and normal uncertain. This study defines the range of normality in certain parameters of torso shape that are also associated with the clinical assessment of scoliosis. This was done by analysing the surface topography of a group of 195 children serially measured over a 5-year period. The analysis considered both the spinal curvature and the relative position of shoulders, axillae and waist on each side. The bivariate relationships were examined using 95% confidence interval data ellipses. Our results showed that a degree of spinal curvature was seen, either as a main thoracic or main thoracolumbar curve. The distribution of the data about a mean point is illustrated by 95% confidence interval (CI) data ellipses with shoulder, axilla and waist data plotted against spinal curvature. The mean values were close to zero (exact symmetry) for all of the measured parameters, with the ellipses showing little differences in the distributions. We conclude that mild asymmetry of the measured torso parameters is normal. These results define what is normal and beyond what point asymmetry becomes abnormal. This information is of use for those managing and counselling patients with scoliosis both before and after surgery.

Key words: axillae; normal; scoliosis; shoulders; surface topography; waist.

Introduction

The assessment of asymmetry between the right and left sides of the torso is part of the clinical management of adolescent idiopathic scoliosis (AIS) (Misterska et al. 2011). Although the diagnosis of AIS is confirmed by looking at spinal shape in the coronal plane using a radiograph, it is the impression of the patient or their family and friends of ‘something being not quite right with the shape of the back’ that leads them to seek medical attention. One of the goals of scoliosis surgery is to minimise torso asymmetry and this is a criterion by which the patient will judge the success of their surgery (Zhang et al. 2011). Features that have been judged in the past as being of most concern to patients and families with AIS include shoulder height difference, scapular prominence and a difference in waist contour (Donaldson et al. 2007; Zaina et al. 2009; Misterska et al. 2011) between the right and left sides.

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The external appearance of the ideal human form is bilaterally symmetrical when viewed in the coronal plane. However, a degree of asymmetry is seen in non-scoliotic children and adolescents. Previous literature has extensively examined the development of differences in the relative prominence of the right and left sides of the back in a growing population (Willner, 1984; Nissinen et al. 1989, 1993, 2000; Grivas et al. 2006) and the height of the shoulders has been examined by Akel et al. (2008). Other features of interest, such as the axillae and waist, have been less well documented (Vercauteren et al. 1982).

In any measured biological parameter, it is of prime importance that the variability of what is classed as ‘normal’ is known, especially if surgery is planned to correct that parameter from what is judged as ‘abnormal’ back to being ‘normal’.

This paper documents the variability of both spinal and torso shape in a non-scoliotic group of children. This allows the development of standards for normality which may be used in the assessment and management of AIS.

Methods

Serial images using the Integrated Shape Imaging System 2 (ISIS2) were taken on a yearly basis in a group of school children without visible spinal deformity to clinical examination over a 5 year period. ISIS2 uses a Fourier transform profilometry method to analyse a digital photograph of the child's back on to which horizontal lines have been projected from above at a slight downward angle. This allows the capture of graphical and quantitative data about the three-dimensional shape of the back. The accuracy and utility of ISIS2 has been reported on in the past (Berryman et al. 2008a,b,c; Brewer et al. 2013).

The stored images were then reanalysed by the first author to identify the two-dimensional torso points marking the shoulders, axillae and waist in the coronal plane (Fig. 1, Table 1). This was done using a custom-designed interface that allows the anatomical point to be manually located. These points are then referenced back to the original ISIS2 image that is calibrated for measurements of distance. The torso parameters stored are described in detail in the next paragraph.

For this study the definition of the axilla point was the most superior point of the posterior axillary skinfold (Akel et al. 2008). The definition of the waist point was the 'minimal waist' (Mason & Katzmarzyk, 2016), the narrowest waist between the thoracic cage and iliac crests. Both the horizontal and vertical measurements of the difference between the right and left sides were then calculated. Two horizontal measures were made, AxDiffOff and WaistDiffOff (Fig. 1). AxDiffOff was defined as the difference in horizontal distance between a vertical line from the vertebra prominens and the axilla points on either side of the torso. WaistDiffOff was defined as the difference in horizontal distance between a vertical line from the sacrum and the waist points on either side of the torso. The vertical measurements were ShDiffHt, AxDiffHt and WaistDiffHt, the difference in vertical height of the right and left points. ShDiffHt was identified using the technique of Akel et al. (2008). In both horizontal and vertical measures a positive number indicates the right was higher or further from the midline than the left. The size of any spinal curve was measured using 'lateral asymmetry', which is the ISIS2 equivalent of the radiographic Cobb angle (Cobb, 1948) and is measured in degrees (Berryman et al. 2008b,c).

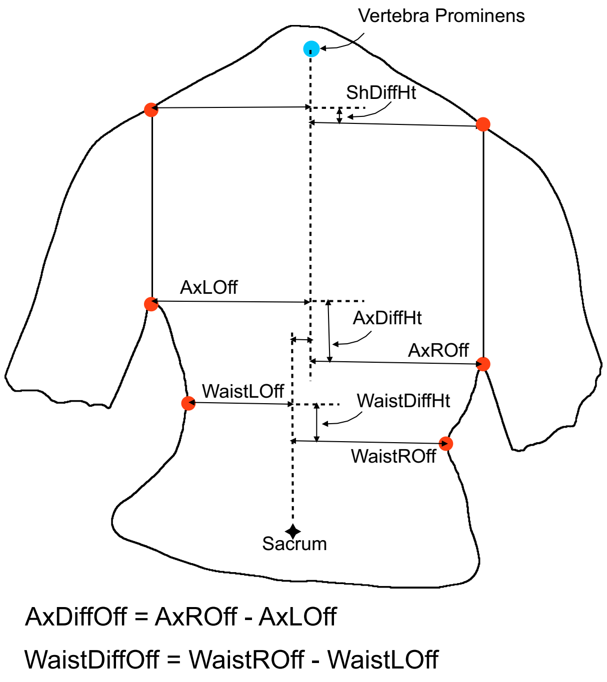

Fig. 1 A graphical representation of the torso and identified points for analysis as defined in Table 1.

The data points were analysed as separate, unrelated points rather than points linked by participants over time. This was felt to be the appropriate way of analysing the data based on both the literature and the data. The literature demonstrates that the shape of the torso changes with age and that longitudinal analysis adds little to the understanding or the ability to predict the shape of the torso in a non-scoliotic population (Nissinen et al. 1989, 1993, 2000). Analysis of our data in a longitudinal fashion demonstrated that the variability of the data over time was such that this form of analysis did not add to the conclusions drawn.

The measured torso parameters are plotted against the spinal curve to demonstrate the data spread around the point of perfect symmetry (the point of no spinal curve and no difference between the right and left sides of the torso). For each pair of variables, the mean point of the data was calculated and the spread of the data were plotted as a data ellipse (Fox & Weisberg, 2011) which represents the 95% confidence interval of the mean. Data ellipses were used to represent the bivariate distributions of the data (Friendly et al. 2013).

Ethical and research governance approvals had been obtained for this study (11/H1207/10). Statistical and graphical analysis was performed using R (R Core Team, 2016).

Results

There were 195 participants in the study group (116 males and 79 females). The mean age of the whole group was 12.5 years (SD 1.8 years, range 9.2–18 years) with the age

Table 1 A table of the torso parameter and their definitions as shown pictorially in Fig. 1.

Orientation	Torso parameter	Definition
Vertical measurements	ShDiffHt	The difference in vertical height between the shoulder points
	AxDiffHt	The difference in vertical height between the axillary points
	WaistDiffHt	The difference in vertical height between the waist points
Horizontal measurements	AxROff	The horizontal distance from the midline to the right axillary point
	AxLOff	The horizontal distance from the midline to the left axillary point
	WaistROff	The horizontal distance from the midline to the right waist point
	WaistLOff	The horizontal distance from the midline to the left waist point
	AxDiffOff	The difference between AxROff and AxLOff
	WaistDiffOff	The difference between WaistROff and WaistLOff

demographics by sex shown in Table 2. The total number of images available for analysis was 642. The participants had all been in the study for a variable length of time and so may have been imaged between one and five times (Table 3). The ethnic origins of the study group were predominantly Caucasian with 3% of the total not Caucasian.

Even though the presence of spinal deformity was an exclusion criterion to the study, there was a degree of spinal curve seen in the majority of the group as measured using the lateral asymmetry parameter of ISIS2. There were two patterns of spinal curve observed which corresponded to patterns of curve observed in scoliosis (Lenke et al. 2001; main thoracic curve with compensatory thoracolumbar curve and main thoracolumbar curve with compensatory thoracic curve). Consequently the analysis of the torso points was performed after subdividing the data in to these groups. There were 387 images in the main thoracic curve group and 227 images in the main thoracolumbar curve group (Table 4). Of the 642 images, 28 were excluded as there was either no spinal curve or a curve pattern that was not classifiable (e.g. a small proximal thoracic curve and thoracolumbar curve but no thoracic curve).

Figures 2 and 3 show the measured torso parameters plotted against the underlying spinal curve pattern. Due to anatomical proximity, ShDiffHt, AxDiffHt and AxDiffOff are plotted against the thoracic curve and WaistDiffHt and WaistDiffOff against the thoracolumbar curve. The mean value and surrounding data ellipse which represents the 95% confidence interval of the mean are plotted for each of the parameters. Positive values in the spinal curve indicate a convexity of the spine to the right and negative values to the left. For the torso parameters, a positive value indicates either the right further from the midline than the left (AxDiffOff and WaistDiffOff) or the right higher than the left (ShDiffHt, AxDiffHt and WaistDiffHt).

When the data were analysed for torso parameter by the anatomically distant curve, no relationship could be found between the size and direction of the curve and the effect on the torso parameter.

Table 2 The age and sex demographics of the dataset.

Sex	*n*	Mean age (years)	SD (years)	Age range (years)
Males	116	12.7	1.8	9.2–18
Females	79	12.0	1.7	9.2–16

Table 3 The number of repeated measurement per participant.

Number of measurements taken	1	2	3	4	5
Number of times participants were measured (males)	9	24	19	29	35
Number of times participants were measured (females)	12	23	13	15	16

Table 4 The size and distribution of the main thoracic and main thoracolumbar components of the main thoracic and main thoracolumbar curve pattern.

Parameter	*n*	Mean (°)	SD (°)	Range (°)
Males				
Main thoracic curve	253	0	6	−11 to 14
Main thoracolumbar curve	134	0	6	−12 to 17
Females				
Main thoracic curve	133	1	6	−12 to 17
Main thoracolumbar curve	94	0	6	−14 to 14

The data were analysed longitudinally by age (a representative plot of one of the parameters is shown in Fig. 4). Although increasing age was associated with a greater width of the torso at both the level of the axillae and waist, this was symmetrical between the right and left sides of the torso and so AxDiffOff and WaistDiffOff did not change. There was no change in ShDiffHt, AxDiffHt or WaistDiffHt with increasing age. There was also no change in curve size with increasing age.

Discussion

This work demonstrates the degree of asymmetry in both the spine and torso seen in a group of children and adolescents who do not have scoliosis. The features that cause the greatest clinical concern in patients with AIS were identified (Donaldson et al. 2007; Zaina et al. 2009; Misterska et al. 2011). From this literature, identifiable and reproducible points around the torso in two dimensions were defined. From these points it has been shown that there is asymmetry between the right and left sides of the body in both the vertical and the horizontal planes. The data also demonstrate that there is a degree of spinal curvature that subdivides into similar patterns to those seen in scoliosis (Lenke et al. 2001), namely a main thoracic curve with a compensatory thoracolumbar curve and a main thoracolumbar curve with a compensatory thoracic curve, although all curves were small.

The main thoracic curve caused the major effect on the height and distance from the midline of the shoulders and axillae, as would be expected due to the anatomical proximity of these structures. Likewise for the main thoracolumbar curve the effect of the curve was seen more clearly in the waist. The compensatory curves showed little effect on the torso points anatomically remote to the curve.

For both the thoracic and thoracolumbar curve patterns, the means of all of the torso parameters and the spinal curves approach zero. The mean of the parameter WaistDiffOff is furthest from zero with WaistDiffOff and

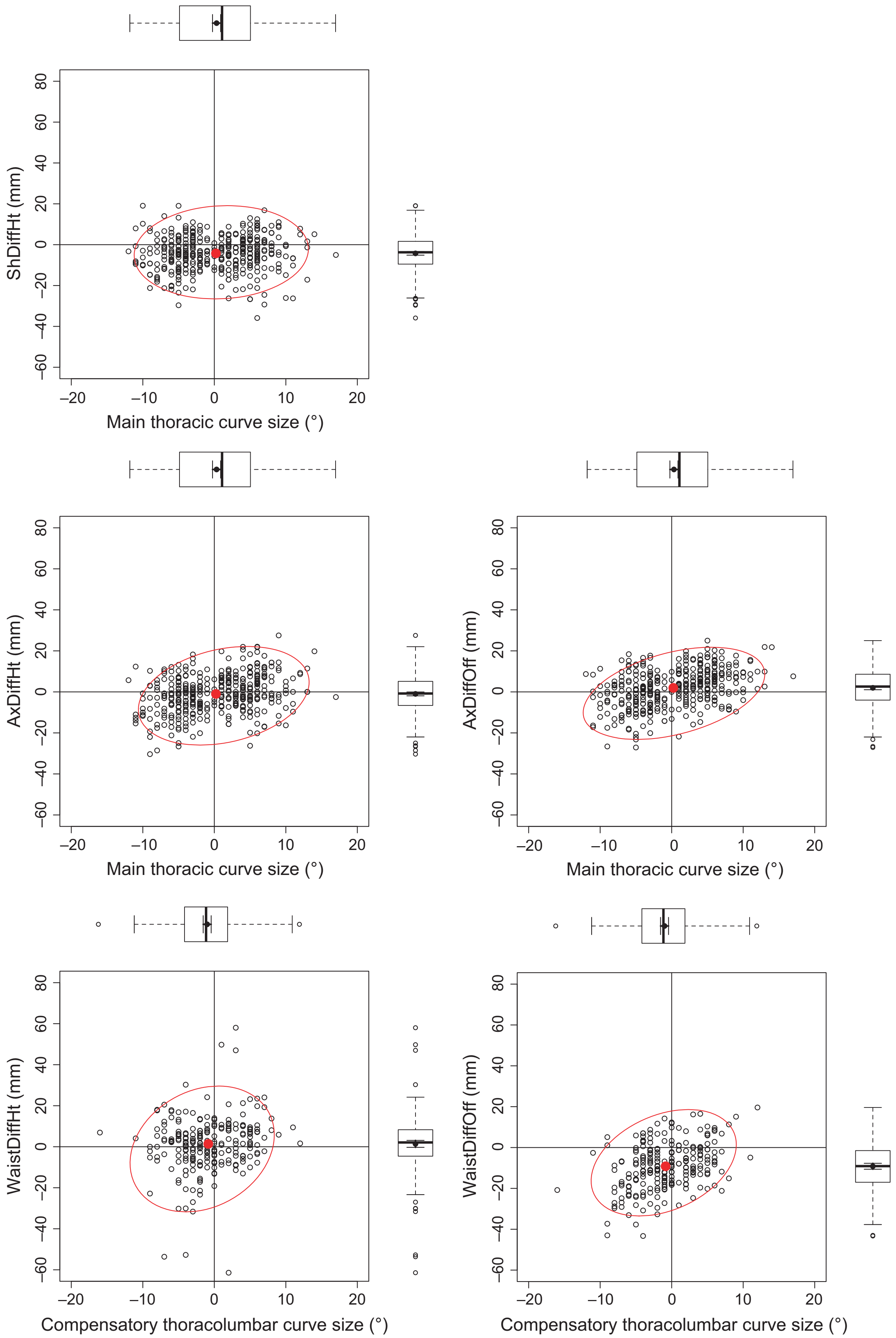
ShDiffHt (mm)
Main thoracic curve size (°)
AxDiffHt (mm)
Main thoracic curve size (°)
AxDiffOff (mm)
Main thoracic curve size (°)
WaistDiffHt (mm)
Compensatory thoracolumbar curve size (°)
WaistDiffOff (mm)
Compensatory thoracolumbar curve size (°)

Fig. 2 Data ellipses for the measured parameters plotted against spinal curve for the main thoracic curve pattern. The mean is the solid red dot with the red ellipse representing the 95% confidence interval of the mean. Box plots represent the distribution of the data for each axis. The median value is the solid line within the box, the latter representing the interquartile range. The whiskers show the range covered by the data points up to 1.5 times the interquartile range, and outliers beyond this range are shown as open circles. The mean and 95% confidence interval of the mean are shown as a solid dot and whiskers around that dot in the box.

WaistDiffHt showing the greatest spread in the distribution of the data. The spread of the data for both the curve and the torso asymmetry around these means show that non-scoliotic children are not symmetrical. This defines a normative dataset for future studies in this area.

Previous work already exists that is similar to that presented here. Akel et al. (2008) took digital photographs of the back for non-scoliotic children at the same time as exposing a radiograph of the chest. The children all believed that their shoulders were level. However, analysis of these photographs demonstrated there was a difference in shoulder height of 7.5 $\pm$ 5.8 mm (mean and standard deviation), with only 18.7% having level shoulders. The methodology used in the Akel et al. (2008) paper to evaluate shoulder height has been reproduced here and therefore their results are directly comparable with ours, which had a mean difference of 4.3 $\pm$ 8.7 mm between the sides. Although differing slightly in methodology to that used by Vercauteren et al. (1982), both our data and that of Akel et al. (2008) are within the differences described as 'physiologic asymmetry' by Vercauteren et al. (1982).

By extending this methodology, the relative height and distance from the midline has also been calculated for the axillae. Again, the mean difference between the right and left sides is small. The data spread around this mean is skewed towards the right being higher than the left with an increasing curve size convex to the right, and this is not seen in the shoulders with the same degree of curve. The difference in distance of the axillae from the midline is very similar to that of the difference in axilla height in terms of the mean, data spread and skew. Overall, this allows the observation that the spinal curve affects the position of the axillae to a greater extent than that of the shoulders, and any difference in height in the axillae due to an underlying spinal curve is countered through the shoulder girdle, thought to be a way to keep the hands at an equal distance from the floor.

The waist point data presented show that there is a greater asymmetry in the heights of the waist between the right and left sides, and in the distance from the midline, compared with the equivalent axillae point data. There are many definitions of what constitutes the waist in the literature depending on the context in which the waist is being identified (Qiu et al. 2010; Guerra et al. 2012; Veitch, 2012; Matamalas et al. 2016). The definition of the waist used here is the 'minimal waist' (Mason & Katzmarzyk, 2016), which is very similar to that of Qiu et al. (2010) and is the definition most suited to a scoliotic population. Of note, the anatomical position of the waist can be less easily identified than that of the axillae and shoulders, especially in younger children who have not developed the torso shape that comes with the adolescent growth spurt. This may decrease the accuracy of the position of the waist points compared to the axillae points with a corresponding increase in data variability.

The data presented here are a combination of both males and females and little reference is made to the effects of growth. Longitudinal analysis of the data showed that age did not affect the mean and variability of the spinal curve and torso points. The absolute width of the torso measured at the level of the axillae and waist increased as the children grew during adolescence. The torso points are the difference between the right and left side and the difference between the sides did not change with increasing age. Subdividing the data into males and females did not alter these observations. Also, analysis by BMI (body mass index) did not alter the results. This is of note as it has been shown that a low BMI can increase the chance of the development of scoliosis (Clark et al. 2014) and thus, by inference, torso asymmetry.

An appreciation and judgment of the shape of the torso in AIS has been published as the cosmetic spinal score (Theologis et al. 1992, 1993), comparing the score to parameters generated from ISIS1 images (Turner-Smith et al. 1988). An association between the cosmetic spinal score and the size of the rib hump was found. The information from the present study concentrates on the features that have been identified as of cosmetic concern in the coronal plane (Donaldson et al. 2007; Zaina et al. 2009; Misterska et al. 2011) and does not assess three-dimensional features such as the rib hump. It is noted that a combination of both two- and three-dimensional features form assessment tools used in AIS such as the Spinal Appearance Questionnaire (Sanders et al. 2007).

Adolescent idiopathic scoliosis (AIS) is known to be a three-dimensional deformity with effects seen in both the coronal and sagittal planes. This paper deals with the torso in the coronal plane, adding to the work of Akel et al. (2008) and Vercauteren et al. (1982). An assessment of sagittal shape in AIS comprises an assessment of the size and shape of the thoracic kyphosis and lumbar lordosis along with the rib hump, rather than an assessment of torso shape through two-dimensional edge points. There is a range of accepted norms for kyphosis and lordosis (Giglio & Volpon, 2007) and rib hump (Nissinen et al. 1989, 1993, 2000). However, investigation of this aspect of back shape was not part of this study.

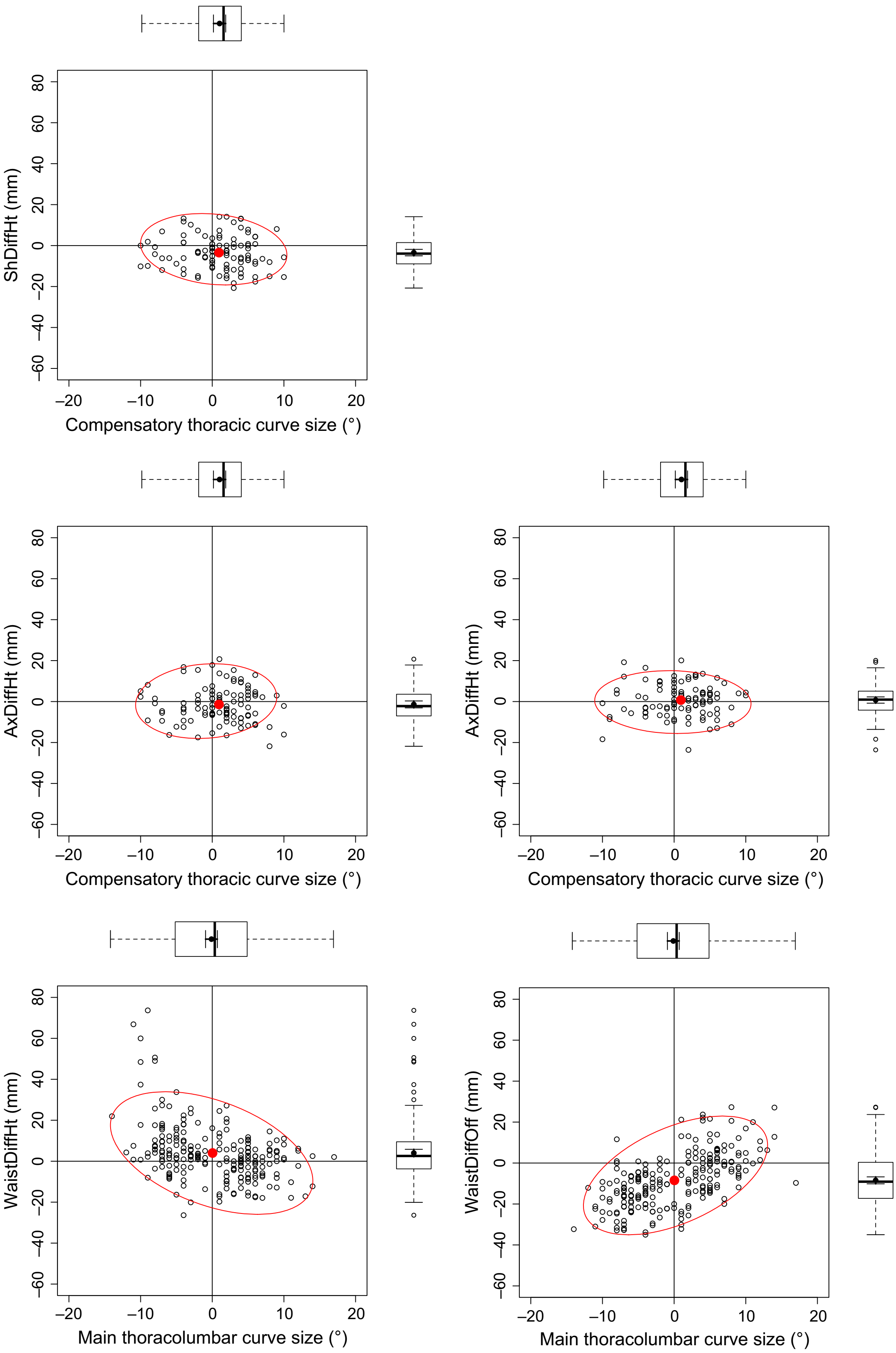

Fig. 3 Data ellipses for the measured parameters plotted against spinal curve for the main thoracolumbar curve pattern (solid red dot, red ellipse and box plots as for Fig. 2).

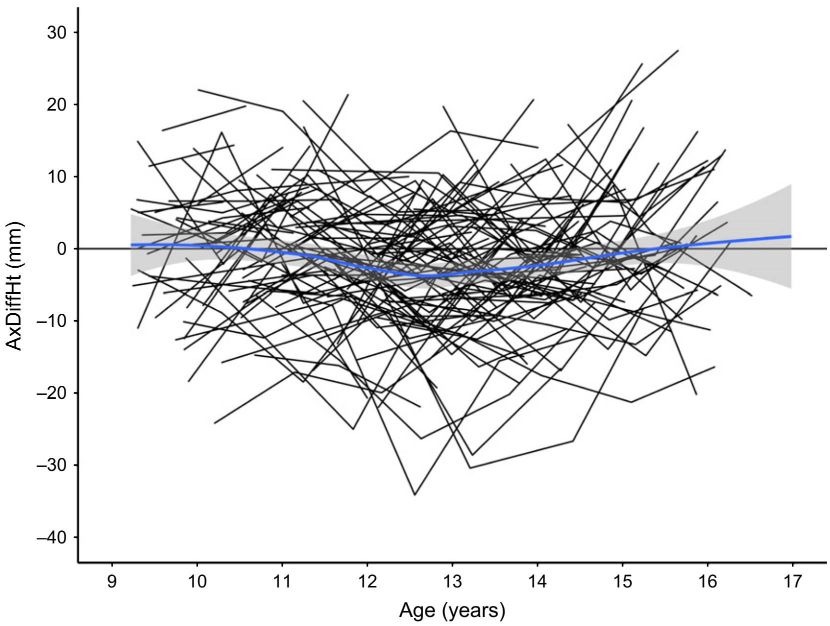

Fig. 4 A plot demonstrating the longitudinal distribution of the AxDiffHt parameter in males over the period of the study. Each line represents one participant as they increase in age during the study. The longitudinal mean is shown for the entire cohort with the 95% confidence intervals of the mean as the thicker blue line and surrounding grey funnel.

It is not possible to be absolutely certain that the participants of this study did not have scoliosis, as only a clinical examination was performed to confirm a non-scoliotic spinal shape. A radiograph or an equivalent (an EOS low-dose radiation image in a standing position or a DEXA (dual energy X-ray absorptiometry) image in a supine position) was not taken. This was due both to concerns over the radiation dose of the investigation, for which there was no direct clinical need (Law et al. 2016; Simony et al. 2016), and the lack of suitable equipment, particularly for an EOS image. The data presented here came from adolescents who did not appear to have scoliosis following clinical examination by experienced clinicians.

These results are useful for defining the variability in position of the shoulders, axillae and waist in a non-scoliotic population. It is also useful information in the management of those with AIS. The correction of the spine from the scoliotic pre-operative to postoperative position is never 100% (Winter et al. 2007). There is always some residual torso asymmetry which can be a source of disappointment to the patient if they are not aware of this in advance of the operation. These data show that there is a range of asymmetries in a non-scoliotic population and that 'normal' is not necessarily perfect symmetry. Anecdotally, patients find this knowledge helpful.

In conclusion, these data define the normative values for spinal shape and how those affect the torso parameters of shoulders, axillae and waist height and the distance of the axillae and waist from the midline on either side of the torso in a normal adolescent population. These data are of use to those managing AIS when counselling patients before and after surgery.

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Conflict of interest

None declared.

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Scoliosis and Spinal Disorders

ERRATUM

Open Access

Erratum to: The use of growth standards and corrective formulae to calculate the height loss caused by idiopathic scoliosis

Adrian Gardner[1*], Anna Price[1], Fiona Berryman[1] and Paul Pynsent[2]

Erratum

After publication of this article [1] the author brought to our attention that the formula of Stokes in Table 1 is incorrect. The correct formula is $y = (1 + 0.066x + 0.0084x^2)/10$ where x represents the mean Cobb angle of the largest two curves in the scoliosis and y the height loss in centimetres.

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Scoliosis and Spinal Disorders

RESEARCH Open Access

The use of growth standards and corrective formulae to calculate the height loss caused by idiopathic scoliosis

Adrian Gardner[1*], Anna Price[1], Fiona Berryman[1] and Paul Pynsent[2]

Abstract

Background: Loss of trunk height caused by scoliosis has been previously assessed using different mathematical formulae. However, these are of differing algebraic construction and will give a range of values for the same size of scoliosis curve. As such, the following study attempted to determine the most valid published formulae for calculating height loss caused by idiopathic scoliosis based on reported growth charts.

Methods: The height and sitting height for a group with idiopathic scoliosis were measured. These were plotted on published growth standards. The size of the coronal curves and the thoracic kyphosis was measured. Height was corrected for the size of the scoliosis using the formulae and replotted on the growth standards. The data spread on the standard was analysed for significant differences between the median and the 5th or 95th centile, and between data outside the 5th and 95th centile.

Results: The sitting to standing height ratio growth standard was used in the analysis as it minimised errors across the different growth standards, given that these standards come from different original populations. In the female group significant differences in the data spread were seen using the formulae of Bjure, Ylikoski and Hwang. Non-significant results were seen for the Kono and Stokes formulae. All formulae caused no significant differences in data spread across the growth standard in the males group.

Conclusions: When assessing against growth standards, the formulae of Kono and Stokes are the most valid at determining height loss caused by idiopathic scoliosis.

Keywords: Scoliosis, Idiopathic, Height, Loss, Formula

Background

Idiopathic scoliosis (IS) is of unknown origin and includes scoliosis seen in the adolescent, between the ages of 10 and 18, and in early adult life once older than 18. It is a growth related deformity of the spine. The deformity leads to a loss of standing height and it is common for surgeons to be asked how much height will be regained when a patient undergoes a corrective scoliosis fusion procedure. Whilst the 'height gained' during surgery is dependent on many factors that cannot reliably be predicted pre-operatively, it is possible to calculate the height that has been lost through formulae that have been published [1–5]. All of the formulae have a different mathematical construction and the aim of this paper is to identify which formula gives the most valid estimation of scoliosis related height loss. This will be performed through an assessment of the concurrent validity of the different formulae with reference to previously published cross-sectional growth standards.

Methods

The standing and sitting heights of a group of patients with IS were measured. All had one curve of at least 10° in the coronal plane to satisfy the definition of structural scoliosis [6] even if the curve pattern was of more than one curve. None of the group had undergone surgical intervention. The measurements were performed with calibrated stadiometers by two research nurses who were not involved in the analysis of the data. The sitting

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height to standing height ratio was calculated. The measurements were all taken at the same time of day to eliminate the change in height that can occur as the day passes [7]. Participants were asked to stand in an 'upright but natural' posture so that the position of the body during height measurement matched the body position during the subsequent radiograph.

The measured data were plotted on the published growth standard. The World Health Organisation (WHO) standard for standing height was used with the data sub divided by sex [8]. This was repeated for sitting height on the Danish sitting height standard and for sitting to standing height ratio, again on the Danish standard [9]. The WHO standing height standard is presented as the median value ± 2 z-scores [8], whereas the Danish sitting height and sitting to standing height ratio is presented as a median value with 5th and 95th centiles [9].

The size of the coronal scoliosis curves and the sagittal kyphosis and lordosis was measured from standing whole spine radiographs accessed digitally. The radiographs were all taken in the same standardised fashion with the sagittal radiograph taken with the arms in the 'fists on clavicle' position to eliminate the effect of arm position on overall sagittal alignment [10].

All coronal curves were measured between the most angled end plates as per the Cobb method [11]. The kyphosis was measured between the superior endplate of T1 and inferior endplate of T12. The lordosis was measured between the superior endplates of L1 and S1. Radiographic measurements were made by an experienced scoliosis surgeon not involved in the measurement of the patient's height. If there was no curve in a part of the spine this was recorded as 0° for that particular segment.

The formulae for calculating height loss are shown mathematically in Table 1 and graphically in Fig. 1. The height loss caused by the scoliosis was calculated using published formulae [1–5] for each participant. The height loss was then added to the standing or sitting height. The new 'corrected' heights were then replotted on the appropriate standard. The sitting to standing height ratio was the most appropriate standard to measure against as it does not include absolute values and thus any bias caused by a standard of a particularly tall population will be minimised. The sitting to standing height ratio is calculated by dividing the sitting height by the standing height.

Two different assessments were made to assess which formulae were most valid for calculating the height loss due to the scoliosis. The first method counted the number of data points on either side of the median but between ± 2 z-scores or between the 5th and 95th centile (from here on defined as the inner data spread) and compared them. Second the number of data points above the + 2 z-scores or the 95th centile were compared with the number of data points below – 2 z scores or the 5th centile (defined as the outer data spread). A statistical comparison was made using a test of equal or given proportions [12]. With data added to the standard, the 'best data fit' would have no significant difference when comparing the inner data spread either side of the median or outer data spread outside ± 2 z-scores or the 5th and 95th centiles. Thus the formula that gives a non-significant result by both analyses would be the most valid available for assessing the height loss from the scoliosis preoperatively. Significant results were defined as a p value ≤ 0.05.

In all of these formulae, y is the calculated height lost and x is the Cobb angle or sum of Cobb angles. In the Hwang [2] formula, z is the kyphosis angle. As not all radiographic series included a sagittal radiograph concordant with the time of height measurement that could be measured for thoracic kyphosis, the total number of data points in the Hwang formula calculation was reduced accordingly.

Bland Altman analysis [13] was also performed. This compared the results of the formulae against each other to calculate the mean and 95 % limits of agreement for the differences between the formulae.

All statistical and graphical analysis was performed using R [14]. The patients in this cohort have been followed up for a minimum of 24 months as part of their medical care although follow up is not part of this paper.

Results

In the scoliosis group, there were 161 females and 44 males. In the analysis for the Hwang formula there were

Table 1 Formulae for height loss caused by the scoliosis (y equalling height loss in cms). The Bjure formula, which is logarithmic, has been changed to a form equivalent to the other formulae for clarity

Name of formula	Formula	Description of formula
Bjure	$y = 10^{0.011x - 0.177}$	x is Cobb angle of the major curve
Kono	$y = (0.6 (x - 30) + 2.6)/10$	x is combined Cobb of all curves
Ylikoski	$y = (0.0062 x + 0.0024 x^2)/10$	x is combined Cobb of all curves
Stokes	$y = (1 + 0.0066 x + 0.0084 x^2)/10$	x is mean Cobb of the largest two curves
Hwang	$y = 0.059 x + 0.012z - 0.919$	x is the major thoracic Cobb, z is T5 to T12 kyphosis

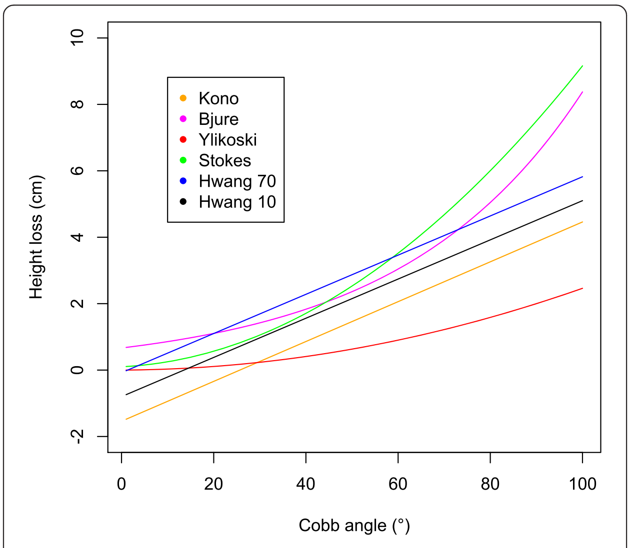

Fig. 1 Height loss formulae plotted as mathematical functions for a single curve pattern. Note: The Hwang formula is depicted as two parallel lines, Hwang 10 and Hwang 70. This allows for a measure of kyphosis as required by the formula. A kyphosis of between 10° and 70° will include all of the data presented in this series which would be found between the two lines

137 females and 37 males. Tables 2 and 3 show the demographics of the groups analysed.

When standing height was plotted on the WHO height standards there was no significant difference between the inner or outer data spread for both the male and female groups (see Fig. 2). A plot of sitting height of the scoliotic females on the Danish sitting height standard shows that the scoliotic females have a lower sitting height than the standard (Fig. 3) [9]. There were significantly more data points between the median and the 5th centile compared to those between the median and the 95th centile ($p < 0.01$), representing an unequal inner data spread. Similarly, there was a significant difference in the number of outliers below the 5th compared to above the 95th centile ($p < 0.01$), an uneven outer data spread. The ratio of sitting height to standing height was also plotted on the Danish standard [9]. Although pictorially the data looks less shifted compared to the sitting height data, again a significant difference in the inner data spread ($p < 0.01$) and outer data spread ($p < 0.01$) for females was seen (Fig. 4). This is repeated, although it is less striking, with the male group, with a significant difference seen in sitting height for both inner ($p < 0.01$) and outer ($p < 0.01$) data spread. For the sitting height to standing height ratio both the inner data spread ($p < 0.01$) and outer data spread ($p < 0.01$) were significant.

Table 2 Demographics of the study group

	Mean age	SD of age (months)	Age range (months)
Males	15 years 7 months	21	11 years 7 months to 20 years and 3 months
Females	15 years 1 month	30.5	8 years and 4 months to 27 years and 11 months

Table 3 Size of scoliosis measured in the study group

	Mean curve size (°)	SD of curve size (°)	Range of curve size (°)
Males	38	22	0–81
Females	44	21	2–96

Corrected standing and sitting height and corrected sitting height to standing height ratios were calculated. This was done by adding the calculated height loss using the different formulae to the original measured data and then replotting on the appropriate standards. For the Stokes method, only the formula for double curves was used, averaging the size of the two curves as in this series there were not enough truly single curves to make analysis of this group meaningful [4].

The table of results (Table 4) shows that for the female group, for the inner data spread, all formulae other than the Kono et al. [3] and Stokes [4] formula caused a significant result, whereas for the outer data spread none of the formulae caused a significant difference in data spread (see Fig. 5). For males, no formulae caused a significant result in data spread for either the inner and outer data spread.

Bland Altman analysis showed that the mean difference between all formulae was less than 3 cm (2.97 cm) in both males and females with 95 % limits of agreement no greater than 5.52 cm [13].

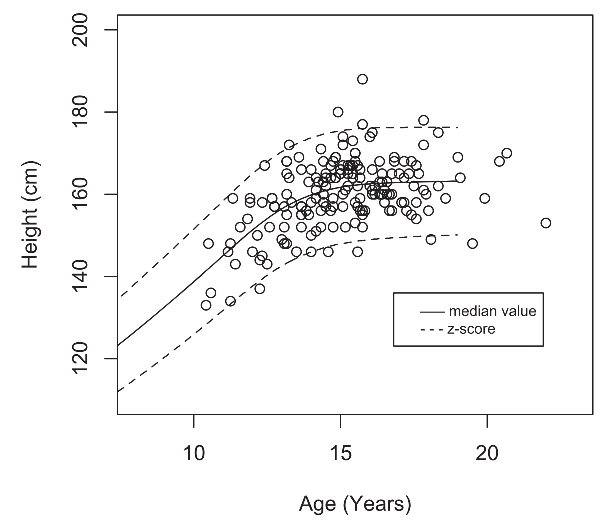

Fig. 2 Standing height of the female group plotted on the WHO standard for standing height

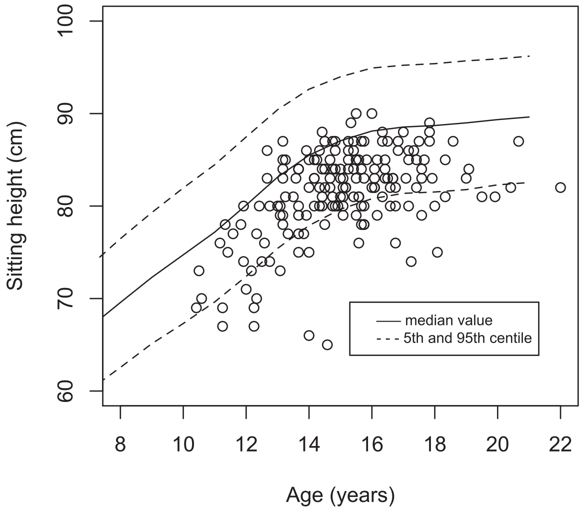

Fig. 3 Sitting height of the female group plotted on the Danish sitting height standard

Discussion

This is the first time that growth standards have been used to reference height loss and corrected height loss in scoliosis to our knowledge. The World Health Organisation (WHO) standing height standards [15] were chosen over those of the Centre for Disease Control (CDC) [16] because, for the over 5 year olds, the data is the same for UK specific standards known as the UK-WHO [17] and would be the most appropriate to the widest worldwide audience. When assessing the growth standards for clinical relevance to this topic in this age group, there is very little difference between any of the standards (Fig. 6) [18].

A sitting height standard is not published by the WHO but the Danish standard used here is easily accessible [9]. The Danish standard also includes growth standards for leg length and sitting height to standing height ratios. It is acknowledged that the Danish are generally an exceptionally tall race and thus placing UK sitting height data onto a Danish standard may generate a false impression of the data [19]. The use of a sitting to standing height ratio eliminates this problem as the absolute value of height is removed from the calculation. Thus, assuming that the ratio of sitting to standing heights is within a similar range between the Danish and UK populations (which given the geographical and historical pasts of both countries is felt to be a reasonable assumption), then the use of this standard is also reasonable. The sitting height standard and sitting height to standing height ratio are presented as median and centiles with the 5th and 95th centile shown here [9]. There is a slight difference between the ± 2 z-scores and 5th and 95th centiles. Two standard deviations are equivalent to 95.45 % of data from the mean in a normal distribution whereas only 90 % is covered between median and 5th or 95th centile. In the setting of this study this difference was not felt to be clinically relevant. The data here demonstrates that there is trunk height loss caused by the presence of a scoliosis as seen on sitting height and sitting height to standing height ratio growth standards. All of these growth standards were created from large numbers of participants minimizing the effects of different populations and outliers to give an accurate description of growth for both standing and sitting height.

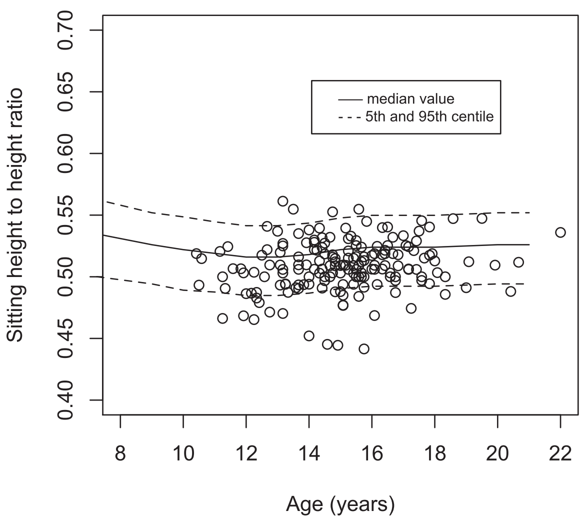

Fig. 4 Sitting height to standing height ratio of the female group plotted on the Danish ratio standard

In clinical practice, patients commonly ask about the height that will be regained following scoliosis surgery. It is very difficult to estimate this pre-operatively as the exact end result of scoliosis correction is dependent on intraoperative factors. The best that can be done is to estimate the amount of height loss secondary to the presence of the scoliosis. This can be achieved pre-operatively using a variety of published formulae that have been detailed here [1–5]. Post-operatively it is also possible to calculate rather than measure the height gained following a scoliosis correction through the use of formulae published by Watanabe and Hosagane [20], Spencer et al. [21] and Sarlak et al. [22] using both pre-operative and intra-operative criteria but these calculation methods are only possible after the event.

The first attempt to calculate height loss caused by a scoliosis was by Bjure et al. [1]. They developed their formula as a way of finding the true height of the thorax in the absence of deformity for the assessment of respiratory function in those with scoliosis. The formula was originally published in 1968 [1] with a typographic error and the formula was corrected to the one used in this paper in 1970 [23]. The weakness of their formula is that it only took into account the major curve in the coronal plane and no assessment was made of the sagittal plane. Further

Table 4 A table of the spread of the inner and outer data spread for males and females

Females	Above 95th	Below 5th	Significance (p value)	Between median and +95th	Between median and +5th	Significance (p value)	Total
Standing height	5	5	1	76	75	1	161
Sitting height	0	36	<0.001*	16	109	<0.001*	161
SH/H ratio	4	24	<0.001*	43	90	<0.001*	161
Standing Bjure	11	3	0.056	91	56	<0.001*	161
Standing Ykilowski	11	2	0.056	90	58	<0.001*	161
Standing Kono	16	2	0.002	98	45	<0.001*	161
Standing Stokes average	19	1	<0.001*	103	38	<0.001*	161
Standing Hwang	7	3	0.334	78	49	<0.001*	137
Sitting Bjure	0	14	<0.001*	49	98	<0.001*	161
Sitting Ylikowski	0	15	<0.001*	52	94	<0.001*	161
Sitting Kono	2	8	0.108	86	65	0.026	161
Sitting Stokes average	3	6	0.498	89	63	0.002	161
Sitting Hwang	0	12	0.001*	32	93	<0.001*	137
Ratio Bjure	10	13	0.665	49	138	<0.001*	161
Ratio Ylikowski	9	12	0.652	53	87	<0.001*	161
Ratio Kono	13	6	0.156	64	78	0.145	161
Ratio Stokes average	11	6	0.319	79	65	0.145	161
Ratio Hwang	9	11	0.816	39	78	<0.001*	137
Males	**Above 95th**	**Below 5th**	**Significance (p value)**	**Between median and +95th**	**Between median and +5th**	**Significance (p value)**	**Total**
Standing height	1	3	0.6088	23	17	0.2844	44
Sitting height	1	10	0.01*	4	29	<0.001*	44
SH/H ratio	0	8	0.009*	11	25	0.005*	44
Standing Bjure	2	2	1	27	13	0.005*	44
Standing Ykilowski	2	2	1	27	13	0.005*	44
Standing Kono	2	2	1	29	11	<0.001*	44
Standing Stokes average	2	2	1	30	10	<0.001*	44
Standing Hwang	1	2	1	24	10	0.002*	37
Sitting Bjure	1	5	0.205	11	27	0.001*	44
Sitting Ylikowski	1	5	205	10	28	<0.001*	44
Sitting Kono	1	2	1	16	25	0.087	44
Sitting Stokes average	1	2	1	22	23	1	44
Sitting Hwang	1	4	0.354	8	24	<0.001*	37
Ratio Bjure	0	4	0.125	16	24	0.134	44
Ratio Ylikowski	0	5	0.065	16	23	0.198	44
Ratio Kono	1	5	0.205	21	17	0.519	44
Ratio Stokes average	2	5	0.431	22	16	0.282	44
Ratio Hwang	0	4	0.123	15	18	0.64	37

Significant results marked with *

criticism has been voiced questioning the accuracy of a logarithmic scale where errors will be greater with a larger Cobb angle, and also with concerns over the lack of standardisation of radiographs for magnification errors [3].

A new formula was proposed by Kono et al. [3] in 2000. They reviewed 140 scoliosis radiographs with both single and double curve patterns, and calculated the true length of the spine in the anteroposterior plane (AP) looking to

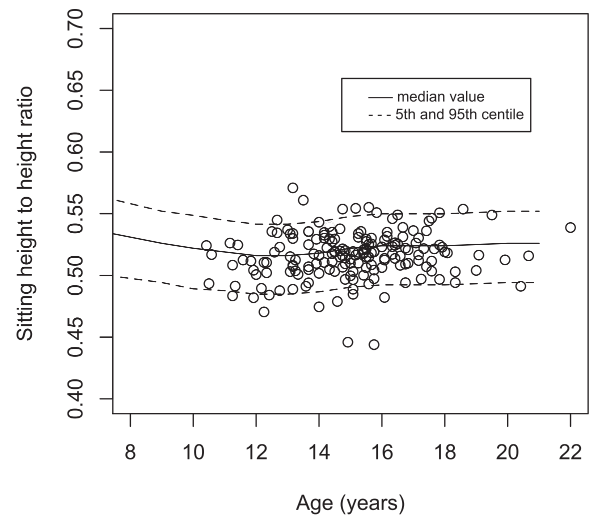

Fig. 5 Female sitting height to standing height ratio corrected with the Ylikoski formula

improve on the Bjure formulae for respiratory function assessment. The calculated loss of height was then analysed with the size of the Cobb angle. The conclusion of the paper was that their new formula should be used instead of the Bjure formula as all of the curves contributed to the height loss rather than just the major curve. However, again, this formula did not take in to account the effect of the sagittal plane and changes in thoracic kyphosis seen with growth and deformity.

In 2003, Ylikoski [5] measured the height of 1500 Finnish girls with scoliosis greater than 10° and compared this to the standing height of the average non-scoliotic girl in Finland. The height loss caused by the scoliosis was measured in the AP plane by subtracting the direct distance between the upper endplate of the T4 vertebral body and the lower endplate of the L4 vertebral body from the measured distance using a flexible wire through all of the vertebral bodies from T4 to L4. The thoracic kyphosis was measured in the same way between the upper endplate of T4 and the lower endplate at the distal end of the kyphosis most commonly seen at T12. A normal value of 29° of thoracic kyphosis was taken from previous work. In his analysis of the first 30 patients, kyphosis greater than 29° led to an addition and less than 29° a subtraction from the overall end height. The conclusion stated that the amount added or subtracted due to thoracic kyphosis did not affect the height of patients with scoliosis when compared to the heights of girls with a normal kyphosis of the same age in a non-scoliotic group; the sagittal plane was therefore excluded from the final formula. There was no assessment of lumbar lordosis for this formula and the effects of the lumbar levels were ignored. In addition, this formula, if applied strictly, is only applicable to females, having been constructed from a female group.

Stokes [4] published a retrospective radiographic review of 387 patients with adolescent or juvenile scoliosis between 9 and 20 years of age comprising 182 single curves and 205 double curves. The size of the scoliotic curves was measured. Spinal length was calculated through the addition of the heights of the vertebral bodies and discs between T5 and L5 from previously stored three dimensional coordinate data of the position of the spine in space. Spinal height was measured from standardised radiographs. Height loss was defined as the difference between spinal height and spinal length. Analysis of height loss with the degree of spinal curvature led to the development of formulae for single and double curve patterns. Stokes [4] stated that it would be appropriate to include the compensatory curve in the calculation of

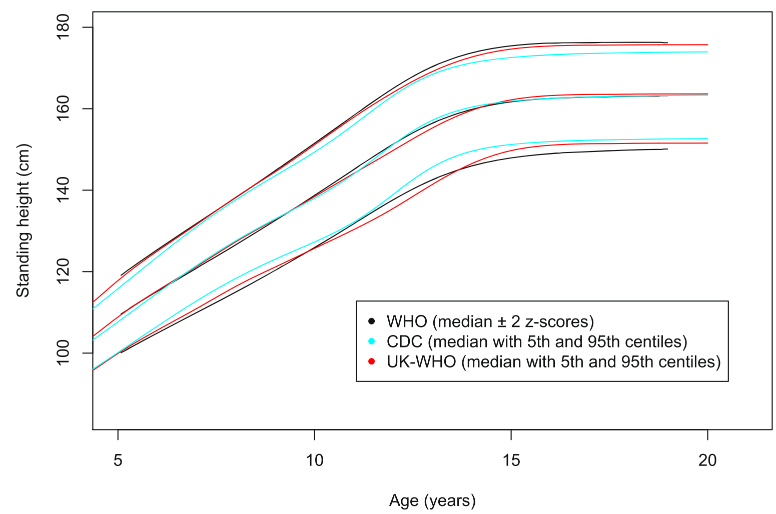

Fig. 6 A plot comparing the growth standards for standing height for WHO [7], CDC [15] and UK-WHO [16] for females

height loss even if it was not structural by averaging the two Cobb measurements. This analysis again only looked at the coronal and not the sagittal plane.

Hwang et al. [2] retrospectively looked at a group of 447 patients with Lenke types 1, 2, and 3 curves in both males and females having undergone only posterior scoliosis procedures. Their formula concluded that height gained is related to the amount of coronal curve and the size of the kyphosis in the thoracic spine. The authors accept that this formula only explains some of the height loss secondary to a scoliosis as it is derived from the post-operatively height. It is likely that this is because a deformity correction is never 100 % and the post-operative spine will not represent the true non-deformed spinal height [24].

The Hwang formula [2] is the only formula to include an assessment of the three dimensional nature of a scoliotic deformity in the calculated height loss. It is known that IS is a lordotic deformity in the sagittal plane, thus there will be less thoracic spine kyphosis when compared to population norms. This may well result in an addition to vertical height rather than the subtraction caused by the coronal plane deformity [25, 26]. Kyphosis in the Hwang [2] paper was measured using the Cobb method between T2 and T12. In this paper, the Hwang formula has been used with the size of the main thoracic curve interpreted as the 'major thoracic Cobb' [2]. The proximal thoracic curve is not included as in the Hwang paper's multivariate analysis, thoracic curve magnitude is quoted rather than proximal or main thoracic curve magnitude [2]. It can be difficult to identify whether a proximal curve is a true structural curve and for consistency only the main thoracic curve was included in this analysis.

The assessment of which of the formulae gives the most valid calculation of height loss secondary to scoliosis has been performed using a test of equal or given proportions on the spread of data points above or below the median or 50th centile line, and outside ± 2 z-scores or the 5th and 95th centile [12]. This has been defined as either the inner or outer data spread. The assumption behind this analysis is that there is an equal distribution in the growth standard at any one age point and the amount above and below the median at that age point will be equal. The formula that changes the data, from initially having a significant difference in spread to being non-significant around the median or as outliers on a growth standard will therefore represent the formula which gives the most valid calculation of height loss. This analysis has been performed on the sitting height to standing height ratio standard to eliminate any effects of the difference in total height between the Danish and UK population.

The uniform spread of data points across the WHO standing height standard, even with a loss of height caused by a scoliosis, demonstrates that the spread of data between ± 2 standard deviations from the median is too large to demonstrate the generally small changes in height caused by most scoliotic curves. This is because the loss of height caused by the scoliosis when viewed as a fraction of total body height in the standing position is small and makes little difference to the whole. In this series the mean height loss across all formulae was 3.38 cm for females and 2.86 cm for males. When expressed as a fraction of sitting height, the change is more obvious and can be seen on the sitting height standard. This then suggests that both sitting and standing height standards should be used to chart height in those with scoliosis to identify the subtle loss from the spinal curve, agreeing with the previous literature [27].

Using the definition of most valid as no significant differences in the number of data points for either the inner or outer data spread when plotted on the sitting to standing height ratio, it is seen that the Kono [3] or Stokes [4] formulae are the most valid for females. In the male group, all of the formulae by either definition are equally valid as they are not significant. As is reflected by a 10 to 1 ratio of females to males with AIS, the female group is larger than the male group. It may well be that with a larger number in the male group the results may change and be closer to those seen in the female group.

Bland-Altman analysis for all pairs of formulae allows a comparison of the differences between them [13]. In this paper, as the absolute height or length of the spine has not been measured, an analysis against 'the truth' is not possible. By sequentially comparing the results of one formula against all of the others the appropriate analysis can be performed. The differences seen in the Bland Altman analysis are small and it is felt would not be deemed to be of clinical significance.

Four of these formulae for calculating loss of height were compared against a measured loss of spine height on an individual basis by Tyrakowski et al. [28]. The 'true height loss' caused by the scoliosis was calculated as the difference between the measured vertical height of the spine between the endplates of T1 and S1 and the measured length of the spine between the endplates of T1 and S1 but through the centroids of all vertebral bodies between. This was then compared to the calculated height loss using four of the five formulae used in this paper [1, 3–5]. The authors conclude that none of the formulae agree about the height loss secondary to the scoliosis. They also note that patients with the same curve sizes but a different overall height will have the same height loss by any of the formulae, but this does not take into account the initial trunk height which may then over or under predict the individual height loss. The use of growth standards versus an individual radiograph in this paper is a different approach in defining the 'true height' to compare with a calculated height. The advantage of a radiograph is that the measurement is of just the spine excluding the head, neck and pelvis. The disadvantage of the radiograph method is that it is individual to that particular patient which, depending on whether the child is tall or small for age may, as described above, over or under predict height loss. Growth standards on the other hand, like the formulae themselves, represent a population. The flaws inherent in a mathematical description of a biological process will be minimized through this approach.

The assumption behind the use of growth standards in this setting is that the scoliotic spine is the same length as the non-scoliotic spine when all other variables are equal when at the same chronological age. This may be flawed as there is some evidence to suggest that scoliosis is an effect of an 'over long' spine or rapid early growth [29]. Further research states that scoliotic children are taller than their non-scoliotic counterparts [26] and that those with more severe curves are taller than those with smaller curves or curves secondary to a leg length discrepancy and pelvic tilt [25]. One hypothesis suggests that this effect may represent the uncoiling of thoracic kyphosis which is greater in a large scoliosis or a different pattern of growth and growth velocity [25, 29–32]. The only way to be absolutely sure would be to measure the length of the spine in three dimensions, possibly from an MRI scan or using an EOS three dimensional scanner, and relate this length to the growth standards and confirm the validity of these formulae against a measured true spinal length.

Conclusion

The height loss seen in the presence of scoliosis is best documented on sitting height and sitting to standing height ratio growth charts. This height loss can be calculated pre-operatively and the most valid result will be obtained with the formulae described by Kono et al. [3] or Stokes [4] when compared to cross-sectional growth standards.

Research ethics committee approval

Ethical approval from the Research Ethics Committee has been obtained for this study reference 15/EM/0283.

Competing interests

The authors declare that they have no competing interests.

Authors' contributions

AG conceived the study, collected data and drafted the manuscript. AP performed the literature search. FB collected data and helped draft the manuscript. PB performed statistical analysis and helped draft the manuscript. All authors read and approved the final manuscript.

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Measuring back shape with ISIS2 in non-scoliotic children

Protocol

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Dr. Fiona Berryman
Sister De Baker
Prof. Jeremy Fairbank

Version: 1.1

Dated: 8th March 2011

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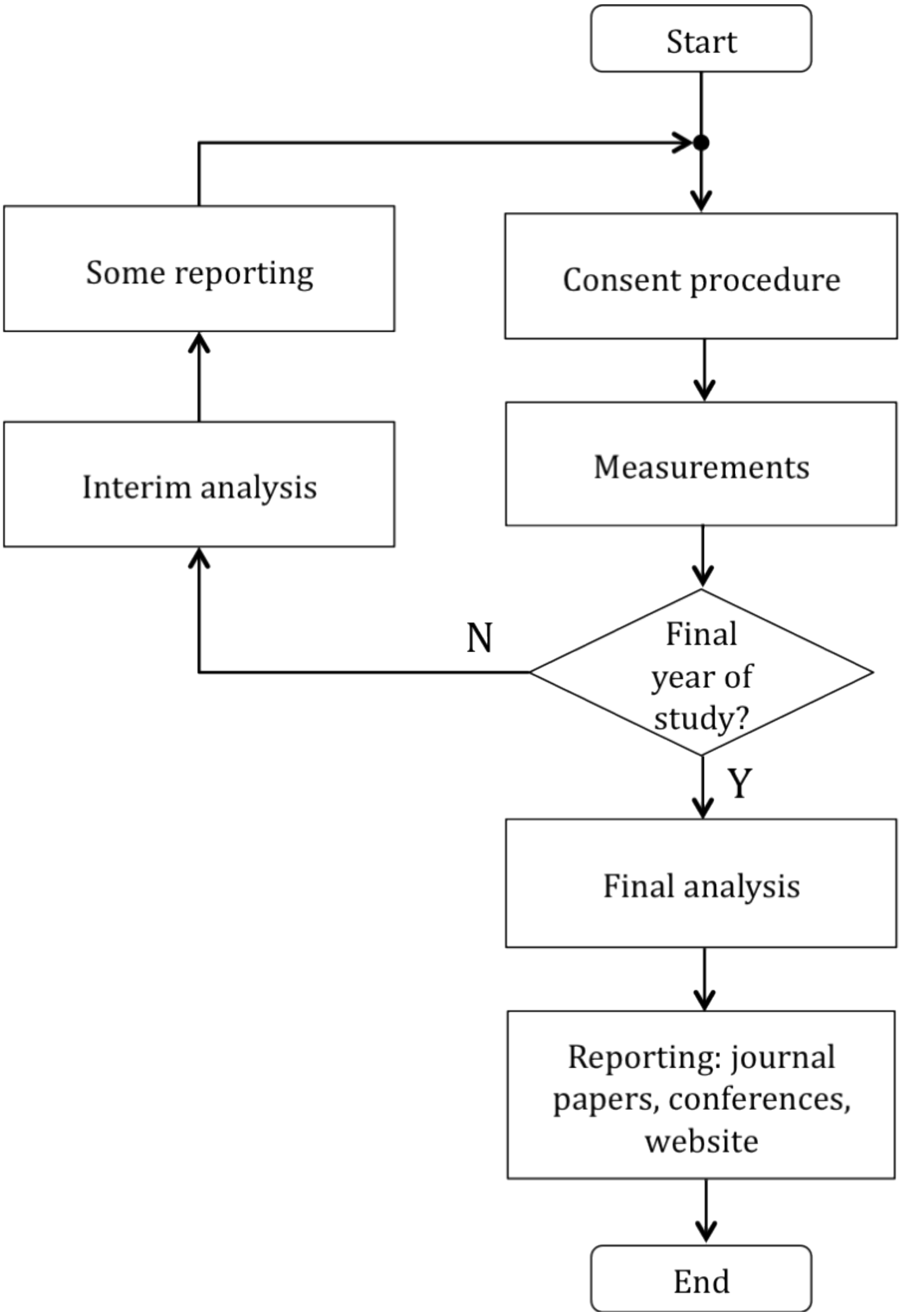
STUDY FLOW DIAGRAM



1. BACKGROUND AND RATIONALE

1. BACKGROUND

The aim of this project is to measure the shape of the back during adolescent growth in normal children. The spine in a normal back when viewed from behind is straight and the two sides of the back are symmetrical. For patients with scoliosis, the spine is curved; this curvature causes the ribs to rotate often giving a hump on the back on one side. In assessing the degree of deformity in patients with scoliosis, it would be very helpful to be able to compare their back shape with the “norm” for their age and build. Although there is now much data on the shape of backs in patients with scoliosis there is very little information about the normal shapes of the spine and chest wall, and how much this varies between individuals or with age during the adolescent growth spurt. This project will give a benchmark for comparison with children with scoliosis. The shape of the back can be measured simply by taking a photograph using ISIS2, a surface topography system developed at the Royal Orthopaedic Hospital. This means an investigation can be carried out on the population without using harmful radiation or contacting methods that take a long time. A second important aspect is measuring the same backs annually over several years. This will show how an individual’s back changes shape with growth. This can then be used as a benchmark for interpreting changes in patients with scoliosis.

The project therefore aims to measure the back shapes of approximately 200 school children every year for seven years using the ISIS2 surface topography system.

1. STUDY RATIONALE

1.2.1 Justification for volunteer population

The recruitment for this study is from children from Bromsgrove School in year groups 5 to 11 (aged between 9 and 15) from both sexes. The age range of 9-15 is chosen because this encompasses the adolescent growth spurt in both sexes. Bromsgrove School has been chosen as there is very little change in the student body between years which will limit the drop out rate of the study. It is a large school with a potential pool of 700 students to measure. We aim to recruit as many children as possible from the initial year group 5 as this group will provide full longitudinal monitoring over a seven year period. Another 25 children will be recruited in all the other year groups to provide additional measurements for each age group. This means approximately 200 pupils will be measured per year (~50 in the initial year group 5 and ~25 in the other six year groups). By the end of the study approximately 1400 measurements will have been made of backs with pupils in the age range 9 to

15. This will provide adequate numbers for calculating statistics on back shape.

1.2.2 Justification for study design

The purpose of this research is to study how the spine and posterior chest wall change with growth in normal children, especially through the adolescent growth spurt.

Surface topography using the ISIS2 system creates a contour map representation of the whole surface of the back including the spinal area and rib cage. The system allows recording and analysis of all aspects of shape, dimensions and asymmetry in a risk free, quick and easy photograph.

ISIS2 will therefore be used to take surface topography measurements of the backs of a group of children who do not have a spinal deformity. Each child will be measured annually over a seven year period. This will give about 200 measurements each year over the age range 9 to 15 years, a total of 1400 measurements. This will enable us to understand the shape changes of the growing child with no deformity. In clinical practice, this will help us distinguish between deformity and growth changes when monitoring children with scoliosis.

Previous studies have looked at back shape using either measurements across a chest radiograph [1] or scoliometer readings [2] [3]. However, these methods of analysis of the shape of the back of the chest are one off measurements of one particular set of parameters, either the transverse diameter of the chest on a chest radiograph or the trunk asymmetry at one place on the back using the scoliometer. None of these previous studies has been longitudinal. This study can produce a range of parameters covering more aspects of the whole back shape than previously published. Furthermore, this current study will use modern statistical methods of shape analysis which have not previously been applied to backs.

Longitudinal measurements of growth of school children have been carried out in the past [4] studying height, weight, limb length, sitting height and a number of other parameters. This project aims to provide similar longitudinal data for measures of normal back shape in adolescents.

1.2.3 Potential risks for participants

A minor risk associated with this study is allergy to the stickers used to mark the bony landmarks. The participants will be informed about the possibility of allergy to the stickers in the information leaflet accompanying the consent form. The participants will be asked about skin sensitivity to the stickers each time they are measured.

There may be some inconvenience in taking part in the study with a loss of school time; however, by working with the school, the photographs will be taken at suitable times to minimise any loss of lesson time. With advance notice, this would then not lead to a loss in education.

There is also the possibility that a few participants may be identified as having a spinal deformity which later turns out not to be the case (false positives) thus leading to some needless worry before their fears are allayed. Where the surface topography results indicate a possible spinal deformity, a confidential letter will be written by the Chief Investigator recommending that the participant and his/her parents consult their GP. This may result in a referral to a spinal deformity clinic. To ensure low rates of false positives only curves of greater than 20 degrees and/or volumetric asymmetry of greater than 20 (ISIS2 parameters measured by surface topography) will be referred to the GP.

1.2.4 Potential benefits for participants

There is no direct benefit to the participants in this study. However, there are three indirect benefits:

- The knowledge that the participants are helping to advance the care of children with spinal deformity.
- Any students with unrecognised spinal deformities will be identified. It is quite common for an adolescent to present in the NHS with a reasonably large and previously unrecognised spinal deformity as children in this age group are independent and are not seen by their parents or peers in a state of undress or because the deformity is slow to evolve it is not noticed by those who are around the child on a day to day basis.
- The participants in the older age groups will be able to take part in statistical analysis of the (anonymised) data thus gaining some experience of the use of advanced statistics in a real life situation.

1.2.5 Potential conflict of interests

If any children are found to have a spinal deformity which has been previously unrecognised, then they will receive appropriate care as would any other child in the NHS.

Although ISIS2, the surface topography system to be used for measuring back shape in this project, was developed by two of the investigators, the system is neither patented nor commercialised and is supplied to any interested users at cost price.

No parties will gain financially from the research to be carried out in this project.

2. AIMS, OBJECTIVES AND ENDPOINTS

2.1 AIMS AND OBJECTIVES

The aim of this research is to study how the spine and posterior chest wall (the surface of the back) change with growth in normal children, especially through the adolescent spurt.

The primary objective:
To measure the statistical variation in back shape with age during adolescence in normal children.

The secondary objectives:
To measure the longitudinal changes in back shape in the same cohort of children over seven years.

2.2 ENDPOINTS

Primary endpoint
The study will end when seven annual measurements on approximately 200 adolescent children each year have been completed and analysed.

Secondary endpoints
None

3. TRIAL DESIGN

This is a longitudinal observational study of children aged between 9 and 15 years old from one school. The study will involve annual measurements of all children in year groups 5 to 11 who are willing to volunteer for the study. One group (the initial year group 5 set) will be followed throughout the study period. The children in the other year groups may change from year to year.

The purpose of this research is to study how the spine and posterior chest wall change with growth especially through the adolescent growth spurt.

Surface topography using the ISIS2 system creates a contour map representation of the whole of the back including the spine and rib cage. The system allows recording and analysis of all aspects of shape, dimensions and asymmetry in a risk free, quick and easy photograph. ISIS2 was developed at the Royal Orthopaedic Hospital in association with the Nuffield Orthopaedic Centre, Oxford [5]. ISIS2 is currently used on a regular basis in NHS clinical practice at several centres.

ISIS2 will be used to make surface topography measurements of the backs of a group of children who do not have a spinal deformity. The children will be measured annually over a seven year period. This will give about 200 measurements each year over the age range 9 to 15 years. This will enable us to understand the shape changes of the growing child with no deformity. In clinical practice, this will help us distinguish between deformity and growth changes when monitoring children with scoliosis.

The photographs taken using the ISIS2 system are analysed using Fourier transform profilometry to calculate the three-dimensional shape of the back from the two-dimensional photograph. The software incorporates innovative mathematical methods with the algorithms designed to minimise the errors found in earlier topography systems [5-8]. The system presents graphical information about the back shape in the transverse, coronal and sagittal planes. It also calculates a range of clinical parameters from the shape including back length, pelvic rotation, flexion/extension angle, coronal imbalance, lateral asymmetry (the ISIS2 estimate of spinal curve in the coronal plane – Cobb angle proxy), volumetric asymmetry, kyphosis and lordosis angles. All numerical and graphical results are stored to a database so that later statistical analysis can be carried out on the measured parameters. Height, sitting height and weight will also be collected and input to the database.

The measured parameters will be plotted against age; regression methods will be used to estimate an analytical curve to fit the observed parameters as a function of age and other covariates. 95% confidence intervals will also be calculated. These results will be compared against similar data for patients with spinal deformities available from the clinical database. The null hypothesis is that there is no significant difference between the ISIS2 parameters measured on adolescents with normal backs compared to adolescents with spinal deformities. The analysis of the normal adolescent data will be carried out each year after the measurements, although low numbers in each age group in the early years of measurement may mean that statistically sound conclusions will only be available in later years as the power of the study increases. Procrustes and Euclidean Distance Matrix Analysis (EDMA) methods of analysing the whole back shape will be carried out and compared with similar data from patients with spinal deformity. Procrustes analysis is a statistical method of analysing the distribution of a set of shapes based on landmark information, considering the degree of rotation, translation and scaling needed to produce the best fit for all shapes. EDMA also uses landmark data from the surfaces but provides a coordinate free calculation method involving ratios of landmark locations.

4. ELIGIBILITY

1. INCLUSION CRITERIA

The recruitment for this study is from children from Bromsgrove School in year groups 5 to 11 (aged between 9 and 15) from both sexes. The age range of 9-15 is chosen because this encompasses the adolescent growth spurt in both sexes. Bromsgrove School has been chosen as there is very little change in the student body between years which will limit the drop out rate of the study. It is a large school with a potential pool of 700 students to measure. We aim to recruit as many children as possible from the initial year group 5 as this group will provide full longitudinal monitoring over a seven year period. Another 25 children, approximately, will be recruited in all the other year groups to provide additional measurements for each age group. This means approximately 200 pupils will be measured per year (~50 in the initial year group 5 and ~25 in the other six year groups). By the end of the study, approximately 1400 measurements will have been taken of backs with pupils in the age range 9 to 15.

The inclusion criterion is every child who wishes to take part from year groups 5 to 11. We will particularly encourage a large cohort from the initial year group 5 so that the group providing full longitudinal monitoring over the study period is as large as possible.

4.2 EXCLUSION CRITERIA

The exclusion criteria are:

- Any child whose parents are not fluent in English and are unable to understand the information allowing informed consent to be given.
- Any child who cannot stand unsupported for five minutes for whatever cause.

The data from some children will be excluded from the statistical analysis of the normal back data if:

- The child has an established spine or rib cage deformity.
- The child has previously undergone treatment for a spine or chest wall deformity.
- The child has previously undergone a thoracotomy or median sternotomy.
- The child is found to have an undiagnosed spinal deformity.
- The child’s bony landmarks cannot be found by palpation.

Such children will still be allowed to take part/continue in the study so that they do not feel that they have been differentiated from their peer group unnecessarily by a medical condition.

5. SCREENING, RECRUITMENT AND CONSENT

1. SCREENING

Prior to study entry, each volunteer's exclusion criteria will be checked.

2. RECRUITMENT PHASE

All children will be approached via a joint letter from the study group and the school outlining the study and inviting all to a talk on the study which will educate and inform parents and children about scoliosis and the study, and which will include a demonstration of the technique. There will be an opportunity to ask any questions with regards to the study. Information leaflets for parents and children, together with consent and assent forms, will also be sent out with this letter.

A website will be developed including a video of a measurement so that potential participants can see exactly what is involved even if they do not attend the demonstration.

Members of the study team will also be present at parents' evenings prior to the measurements so that potential participants and their parents can ask any questions directly to them if they do not want/cannot attend the demonstration.

3. CONSENT/ASSENT

The consent (parents) and assent (children) forms will be taken back prior to the study commencing. There will be a window of at least two weeks between the meeting and the start of the study. Details of how to contact the study team will be given in the information leaflets. Thus, should queries arise after the meeting(s) but before the study commences, an opportunity to seek further information is given. Consent will be taken from all parents for any child eligible for enrolment in the study. All children will also be offered the opportunity to sign an assent form.

2. RANDOMISATION

Randomisation is not required for this observational study.

3. MEASUREMENT DETAILS

1. SUMMARY

ISIS2 will be used to take surface topography measurements of the backs of a group of children who do not have a spinal deformity. Measurement simply involves taking a normal digital photograph of the child's back. The back must be exposed from head to hips. The photographs taken are only of the back; the face is never seen. Figure 1 shows an example of an ISIS2 photograph.

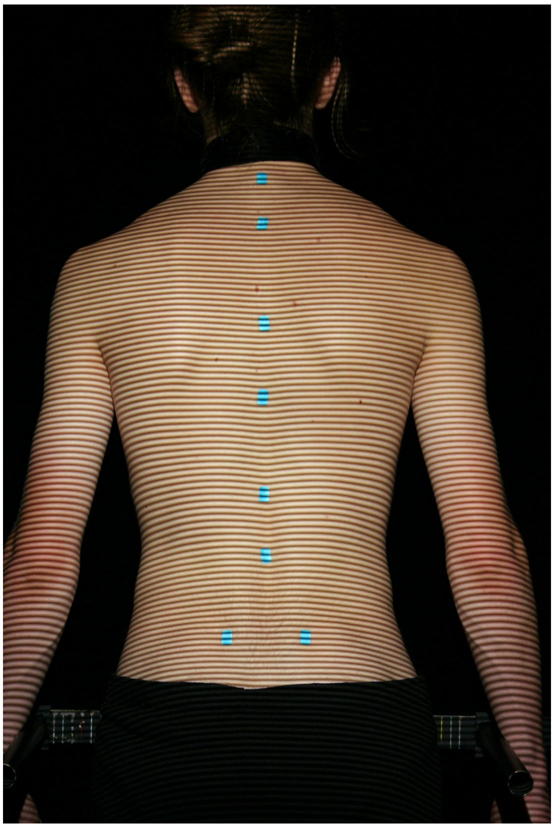


Figure 1: Example of an ISIS2 photograph

The procedure for taking the photographs is as follows:

- Clothes will need to be removed from the top half of the body and trousers/skirt will need to be loosened so that the back is exposed from the neck to the bottom of the back.

- Long hair will be tied up so that it does not cover the back.

- A special black neck band and apron are worn to provide clear limits for the area of the photograph to be analysed.

- For females a special gown is provided to cover the subject's front.

- Some small coloured paper stickers are placed on the back marking certain bony landmarks.
- A photograph of the back is taken.
- The picture is analysed and a report created. The subject will be given a copy if they wish.
- A second photograph may be taken to check for differences in stance.
- The stickers are removed and the subject can dress again.
- Basic stature measurements of height, sitting height and weight will be recorded.

It will take approximately ten minutes to complete all of the above stages.

The computer software is part of the ISIS2 equipment so analysis of the photograph to provide surface topography data and clinical parameters is done immediately the photograph is taken.

An example of the report generated by the system is shown in Figure 2.

This measurement procedure will be followed for each volunteer annually over a seven year period. This will give about 200 measurement each year over the age range 9 to 15 years, a total of about 1400 measurements.

The photographs will be taken by Dr. Fiona Berryman, clinical scientist, and Sister Delia Baker, clinical outcomes sister. Both women have enhanced CRB clearance for working with children.

We anticipate the process to be spread over one week per year although each child's involvement will only be approximately ten minutes.

All the participants will be anonymised to all staff involved in administration of the study. For the longitudinal aspect of the work, to be able to relate the results of one child year on year, there will need to be a unique code allocated to each volunteer. A key relating the unique codes to the names of the participants will be kept by the school and will not be accessible to the study staff. The only pieces of personal information that will be required will be the date of birth and gender of the participant because the age at the date of measurement and gender will be needed for the statistical analysis.

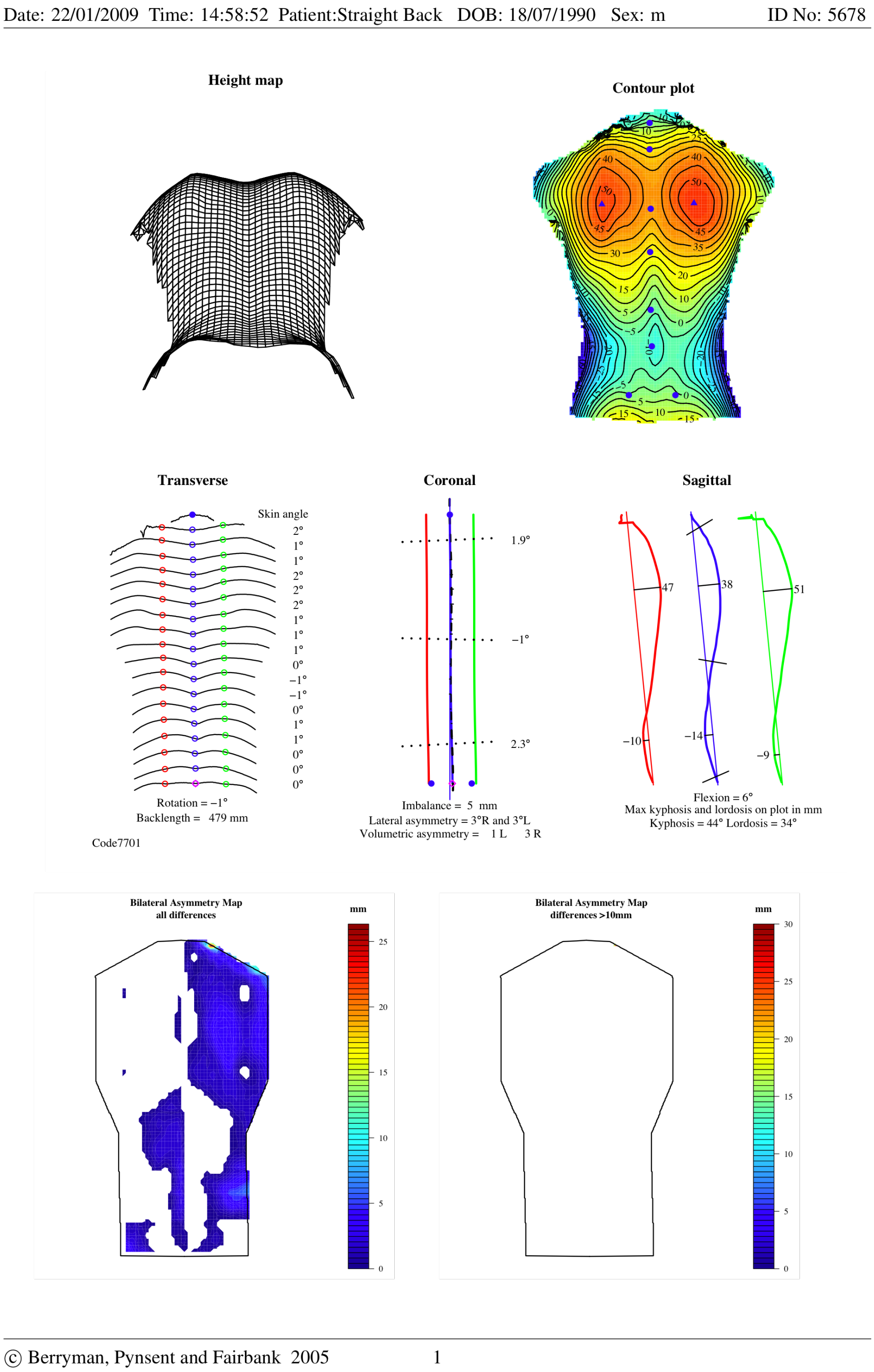


Figure 2: Example of report from ISIS2

<u>Should a medical problem that requires treatment be identified in one of the participants, a confidential letter about the problem will be written by the Chief Investigator and sent to the parents recommending a review with the GP. The code for identification will be broken to identify the child by the Chief Investigator via the school nurse. Only the Chief Investigator and the school</u>

nurse will thus know the identity of the child. The letter will be sent directly to the parents of the child in question.

2. INVESTIGATIONAL THERAPY ARM

Not applicable for this project.

8. ASSESSMENTS/DATA COLLECTION

1. VISIT SCHEDULE

The participants will be photographed once per year. The ISIS2 system will be transported to the school and set up there so that measurements can be carried out with minimal disruption to the normal school day. Gathering the data for each participant takes approximately ten minutes. It is expected that a week will be spent on site at the school each year, gathering the data from all the pupils participating in the study, approximately 200 each year.

2. SERIOUS ADVERSE EVENTS

As this study is purely an observational study it is unlikely that there will be adverse effects to report.

The only known risk to the participants from taking part in this study is the possibility of skin sensitivity to the stickers used to identify the bony landmarks. The participants will be asked about skin sensitivity at every measurement and may refuse to proceed with the measurement if he/she feels that the skin is too sensitive for the use of stickers.

9. DATA MANAGEMENT

1. DATA COLLECTION

All data will be collected on an ROH computer running the ISIS2 software at Bromsgrove School. The data will be stored in the ISIS2 database and the computer will be password protected and encrypted.

9.2 DATABASE MANAGEMENT AND QUALITY CONTROL

All participants will be anonymised to the staff involved in administration of the study. However, for the longitudinal aspect of the work, to be able to relate the results of one child year on year, there will need to be a unique number allocated to each volunteer. A key relating the unique numbers to the names of the participants will be kept by the school and will not be accessible to the study staff.

The database will contain the back surface data, clinical parameters associated with the surface, the photographs, height, sitting height, weight and basic information about the volunteer namely gender, date of birth and the unique code. Access to this database will only be granted to the researchers named in this document.

On completion of the study, the data will be stored in encrypted files at the Royal Orthopaedic Hospital with access only to Professor Pynsent. The encrypted password will be kept in a sealed envelope in the Royal Orthopaedic Hospital safe. The envelope will only be opened by a member of the study team in the event of changes in Professor Pynsent's circumstances.

All data will be handled, computerised and stored in accordance with the Data Protection Act 1998. Quality control will be maintained through adherence to ICH GCP and the R&D Quality Assurance Process and approved by the Caldicott Guardian (Mr A. Thomas).

10. STATISTICAL CONSIDERATIONS

10.1 PLANNED RECRUITMENT RATE

The recruitment for this study is from children from Bromsgrove School in year groups 5 to 11 (aged between 9 and 15) from both sexes. The age range of 9-15 is chosen because this encompasses the adolescent growth spurt in both sexes. Bromsgrove School has been chosen as there is very little change in the student body between years which will limit the drop out rate of the study. It is a large school with a potential pool of 700 students to measure. We aim to recruit as many children as possible from the initial year group 5 as this group will provide full longitudinal monitoring over a seven year period. We estimate that this will be approximately 50 children. Another 25 children will be recruited in all the other year groups to provide additional measurements for each age group. This means approximately 200 pupils will be measured per year (~50 in the initial year group 5 and ~25 in the other six year groups). Each subsequent year, another 25 children will be recruited from the new year group 5 and those who have reached year group 12 will drop out. By the end of the study, approximately 1400 measurements (~200 per year group) will have been taken of pupils in the age range 9 to 15. This will have involved approximately 350 volunteers in total over the full study period.

10.2 SAMPLE SIZE

This is a new application of statistical methods where it is not clear what variability will be found. We believe the number recruited (~200 per year group over the full study) will be sufficient to make inferences although we cannot yet guarantee what the variability in the parameters in a normal population will be. However, if we assume that the covariates of height, sitting height, weight and sex are related to the variance of shape and shape change, then our study will have sufficient power to minimise type II errors.

11. STATISTICAL ANALYSIS

11.1 INTERIM ANALYSIS

In the initial year there will be ~50 measurements for the year group 5 set and only 25 for the other year groups. Gradually over the period of the study these numbers will increase to ~ 200 per year group.

The number of participants needed to measure mean differences in ISIS2 parameters between the normal and the scoliotic backs with a power of 0.95 ranges from 40–110 depending on the measured parameter and the difference in its mean that is sought. The number of children participating in the project will therefore be adequate to provide significant statistical results with a power of 0.95. (The sample size/power calculations were carried out using the power.t.test function in R [9].)

The measured parameters will be plotted against age for male and female participants; regression methods will be used to estimate an analytical curve to fit the observed parameters as a function of age. 95% confidence intervals will also be calculated.

Procrustes and EDMA methods of analysing the whole back shape will also be carried out and compared with similar data from patients with spinal deformity. No sample size calculations can be carried out in this case because the project is a new application for this type of analysis. We believe, however, that the numbers recruited will be sufficient.

11.2 STATISTICAL ANALYSIS

Clearly in this longitudinal study, time (age) is the key independent variable with sex as the second covariate. Thus all models explaining shape change will be based on this assumption. These models will be developed and refined as the study progresses. However, the stated null hypothesis of this study is that there is no difference in shape and shape change in normal and scoliotic backs. Thus, when we are confident that our models of shape and shape change in normal backs are correct then these models will be applied to longitudinal data from our spinal deformity database.

All analyses will be carried out at the 5% level of significance.

12. DATA MONITORING

Data will be monitored for completeness and quality by the R&D Directorate via the Quality Assurance Process. All safety data will be considered by the R&D Directorate.

13. ETHICS AND GOOD CLINICAL PRACTICE

The trial will be performed in accordance with the recommendations guiding physicians in biomedical research involving human subjects, adopted by the 18th World Medical Assembly, Helsinki, Finland 1964, amended at Edinburgh in 2000. The study will be submitted to and approved by the appropriate independent Research Ethics Committees, prior to entering patients into the study. The trial will be conducted in accordance with ICH GCP.

14. CONFIDENTIALITY

The data collected about each participant will be gender, date of birth, height, sitting height, weight and a photograph of the back. The participants will be anonymised to the staff involved in administration of the study. The researchers will never know the names of the participants. For the longitudinal aspect of the work, however, the results from one child year on year must be followed; therefore, there will need to be a unique code allocated to each volunteer. The school will allocate unique codes to each participant and keep a key relating these codes to the names of the participants. This key will not be accessible to the study staff.

Should a medical problem that requires treatment be identified in one of the participants, a confidential letter about the problem will be written by the Chief Investigator and sent to the parents recommending a review with the GP. The code for identification will be broken to identify the child by the Chief Investigator via the school nurse. Only the Chief Investigator and the school nurse will thus know the identity of the child. The letter will be sent directly to the parents of the child in question.

Agreement to the anonymisation process and the action that will be taken if a medical problem is detected is part of the consenting procedure.

Summing up, the school will have the unique codes and names of all participants but no data associated with them; the research team will have the numbers and data but no names to identify the participants.

The handling of the photographs and data will comply with all aspects of the Data Protection Act 1998.

Any information which would allow individual participants or clinicians to be identified, will not be released into the public domain.

15. PUBLICATION

The results of the analysis of the data generated by the study will be published in hospital reports, conference presentations and journal papers. We intend to publish information about the results of the research on the ROH Orthopaedic Charity website (www.orthsurg.org.uk).

16. REFERENCE LIST

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Appendix 1

The Royal Orthopaedic Hospital NHS
NHS Trust

Research and Teaching Centre, Royal Orthopaedic Hospital, Northfield, Birmingham, B31 2AP

Study: Normal back shape in adolescents

Parent Information Sheet

We would like to invite your child to take part in a study investigating the normal shape of the back in adolescents. Before you decide if you would like your child to take part you need to understand why the research is being done and what it would involve. Please read the following information carefully. Talk to others about the study if you wish. Ask us if there is anything that is not clear or if you would like more information.

What is the purpose of this study?

Figure 1: X-ray showing a curved spine.

Scoliosis is curvature of the spine. When the spine bends it causes the ribs to rotate, producing a hump on the back. Although the curvature of the spine can be measured using X-rays (see Figure 1), this does not give any information about the surface of the back. The shape of the surface of the back can be measured using surface topography which produces a picture similar to an Ordnance Survey contour map. We are using surface topography regularly in our spinal clinic to monitor back shape changes in patients with scoliosis but we have very little information about the shape and variability in normal adolescent backs. This information is important as surgical correction of a scoliosis alters the shape of the back of the chest; surgeons need to have accurate information about normal backs to get the best possible results for the patient.

We would therefore like to measure back shape in a large group of normal adolescents, annually over a period of seven years. This will give us statistical information about the shape and variability in normal backs. The measurements over several years will enable us to investigate how an individual's back changes shape as he/she grows through the pubertal growth spurt. We can then use this data as a benchmark for changes in patients with scoliosis.

We will measure the shape of the back using a surface topography system called ISIS2 which was developed at our hospital.

What is ISIS2?

ISIS stands for Integrated Shape Imaging System; the '2' is because this is a modern version of technology that was first developed in the 1980s. The ISIS2 system can calculate the shape of the back from a simple digital photograph. This is accomplished by shining a grid of horizontal lines onto the back (Figure 2), photographing the back and then using special mathematical methods to convert the distortions of the grid into a three dimensional map of the back (Figure 3). Various clinical parameters are then calculated from the surface data. All data collected are stored electronically and can be retrieved at subsequent measurements. This allows the researcher to see how the back has changed with time.

Figure 2: Back photograph of child with scoliosis showing the stickers and projected grid

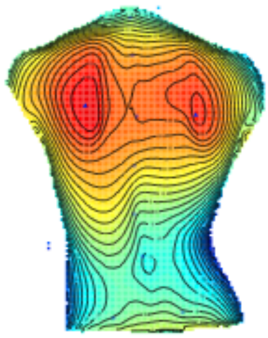


Figure 3: Contour map

The development of ISIS2 enables us to measure back shape simply by taking a photograph. This means we can carry out an investigation on the population without using harmful radiation or contacting physical methods that take a long time.

Why have we chosen your child?

The age range of interest is 9–15 years because this is the period when a child grows most quickly. In scoliosis this is the time that a curve is most likely to progress. Bromsgrove School has been chosen because there is little change in the pupils attending from year to year which should limit the drop out rate of the study. So long as your child can stand unsupported for a few minutes for the photograph to be taken, we would like him/her to take part.

Does your child have to take part?

No. It is up to you and your child to decide whether he/she would like to take part in the study. If you and your child decide that he/she would like to take part, you will be asked to sign a consent form and your child will be asked to sign an assent form. If you decide that your child will take part now, you can still change your mind later and withdraw your child from the study at any time.

What will participants need to do?

Once a year a photograph will be taken of your child's back using the ISIS2 system while he/she remains in the 9–15 years age group. This will involve the following procedure:

- Your child will need to remove the clothes from the upper body and loosen his/her trousers or skirt so that the back is exposed from the neck to the bottom of the back.
- Hair may need to be tied up so that it does not cover the back.
- The child will wear a black neck band and apron to provide clear limits for the area of the photograph to be analysed.
- For female participants, a special gown will be provided to cover their front.
- Some small coloured paper stickers will be placed on the back marking certain bony landmarks.
- The child will be photographed with his/her back to the camera.
- The computer will analyse the photograph and present clinical information about the back on screen and on paper. A copy of the report will be given to your child to keep for your records.
- A second photograph may be taken and analysed to check for variability in stance.
- The stickers will be removed from your child's back and he/she will get dressed again.

The whole procedure is likely to take about ten minutes. A video of such a measurement is available at the ROH Orthopaedic Charity website, www.orthosurg.org.uk.

All photographs and the data calculated about the shape of your back will be stored in a database. This database will be confidential and it will be stored and used in accordance with the Data Protection Act.

What are the possible benefits of taking part?

The main benefit is the knowledge that your child is helping advance the care of children with spinal deformity.

If there were unrecognised spinal deformities in the children photographed, these would be identified. Where surface topography results indicate a possible spinal deformity, a confidential letter will be written by the Chief Investigator recommending that the child and his/her parents consult their GP.

We intend to involve the senior students in some statistical analysis of the anonymised data so that they can gain some experience of the use of advanced statistics in a real situation.

What are the disadvantages or risks in taking part?

The main risk associated with the measurements is allergy to the stickers used to mark the bony landmarks. Your child will be asked every time a photograph is taken about skin sensitivity.

There may be some inconvenience with loss of school time; however, by working with the school the photographs will be taken at suitable times to minimise any loss of lesson time. With advance notice, this would then not lead to a loss in education.

There is also the possibility that a few participants may be identified as having a spinal deformity which later turns out not to be the case (false positives) thus leading to some needless worry before their fears are allayed. This may result in a referral to an NHS spinal deformity clinic. To ensure low rates of false positives only curves of greater than 20 degrees will be referred to the GP.

Will my child's participation in this study be kept confidential?

All information collected about your child in the course of the research will be kept confidential. The school will allocate a unique code to each child who participates so that data sets from the same subject can be compared from year to year. The database will identify the participant by this code, his/her initials, sex and date of birth (both sex and date of birth are needed for the subsequent statistical analysis). Even if a child cannot tell us at later measurements what his/her unique code is, we will be able to use initials and date of birth to retrieve the code from the system without actually recording full names. The data is thus fully anonymised. The database will be stored on an encrypted computer at the Royal Orthopaedic Hospital. Professor Pynsent will be its custodian, only allowing access to the data to the researchers involved in the project.

What will happen to the results of the study?

The researchers will publish their results at scientific conferences worldwide and in respected scientific journals. We will also publish information about the results of the research on the ROH Orthopaedic Charity website (www.orthosurg.org.uk).

Who is organising and funding the research study?

We have received a grant from the ROH Orthopaedic Charity. The principal researchers are employed by the Royal Orthopaedic Hospital. ISIS2 was developed at the Royal Orthopaedic Hospital in association with the Nuffield Orthopaedic Centre, Oxford but it is not a commercialised system and is supplied at cost price to any other interested users. No parties will gain financially from its use in this project.

Who has reviewed the research study

We have obtained the views of independent referees in setting up this study. All research in the NHS is also examined by an independent group of people called a Research Ethics Committee. This project has been reviewed and given a favourable opinion by the ???? Research Ethics Committee.

Who will be involved in conducting the study?

The research team involved in this project comprises:

- Mr. Adrian Gardner, Spinal Surgeon, Royal Orthopaedic Hospital.
- Dr. Fiona Berryman, Clinical Scientist, Royal Orthopaedic Hospital.
- Sister Delia Baker, Clinical Outcomes Sister, Royal Orthopaedic Hospital.
- Prof. Paul Pynsent, Clinical Scientist, Royal Orthopaedic Hospital.

We have also discussed the project with Prof. Jeremy Fairbank, Spinal Surgeon, Nuffield Orthopaedic Centre and taken advice from him.

For more information contact:

- Mr Adrian Gardner Tel: 0121-685-4083 E-mail: adrian.gardner@roh.nhs.uk
- Dr Fiona Berryman Tel: E-mail:

If you have any concerns or complaints which Mr Gardner and Dr Berryman cannot deal with, then please contact the Complaints Department:

- Telephone: 0121-685-4016
- E-mail: complaints@roh.nhs.uk
- Letter: Chief Executive, Royal Orthopaedic Hospital, Bristol Road South, Northfield, Birmingham, B31 2AP

The Royal Orthopaedic Hospital NHS
NHS Trust

Research and Teaching Centre, Royal Orthopaedic Hospital, Northfield, Birmingham, B31 2AP

Study: Normal back shape in adolescents

Information Sheet for children aged 9–15 years old

We would like to invite you to take part in a study measuring the shape of the back in children. The purpose of the research is to help people who have scoliosis, a condition where the back is twisted. We need you to read this information sheet before you decide if you would like to take part. You can ask questions and talk to your family and friends about the study before you decide. Your mum or your dad will also have an information sheet to read and they will also have to give their permission if you want to join the study.

Why are we doing this research?

Scoliosis is medical condition where the spine or backbone curves and twists so that the shape changes from what it should be (see Figure 1). When the spine twists it often makes one side of the back stick out more than the other. We measure the back shape of children who come to the hospital with scoliosis because the shape helps us to decide what treatment these children should have. However, we have very little information about the shape in normal children's backs and how that changes as they are growing. We would like to measure your back shape each year until you are 16 to help us improve treatment for other children with scoliosis. If you agree to join the study, we will measure the shape of your back by taking a digital photograph of it using a system called ISIS2.

Figure 1: X-ray showing a curved spine.

What is ISIS2?

ISIS stands for Integrated Shape Imaging System; the '2' is because this is a modern version of technology that was first developed in the 1980s. The ISIS2 system can calculate the of the back from a simple digital photograph. We do this by shining a pattern of horizontal lines onto the back (Figure 2), photographing the back and then doing some special analysis to give a three dimensional map of the back (Figure 3). This figure is similar to an Ordnance Survey map of hills.

Figure 2: Back photograph of child with scoliosis showing the stickers and projected lines

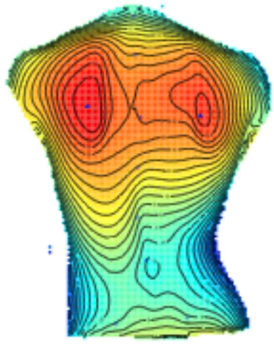


Figure 3: Contour map

ISIS2 lets us measure back shape simply by taking a photograph. This means we can measure lots of children very quickly.

Why have we chosen you?

We want to measure children aged 9–15 years old which is the age you are. We have chosen Bromsgrove School because the pupils attending from year to year do not change very much. If you can stand for a few minutes for the photograph to be taken, we would like you to take part.

Do you have to take part?

No, you don't. It is up to you. You can decide to take part and then change your mind later if you want to. No one will mind if you do not take part or if you change your mind later.

What will happen if you do take part?

Once a year a normal digital photograph will be taken of your back while you are aged between 9 and 15 years old. The camera we use is just like one you may have at home.

This is what happens when the photograph is taken:

- You remove the clothes from your top half (waist upwards) and loosen your trousers or skirt so that we can see all of your back.
- If you have long hair we will ask you to tie it up so that it does not cover any of your back.
- You will wear a black neck band and apron to make it easy for the computer to analyse the photograph.
- Girls will also wear a special gown which covers their front.
- We will stick some small coloured paper stickers on your back.
- We will take a photograph of your back while you stand in a special stand with your arms away from your sides.
- The computer will analyse the photograph and present the results on the computer screen and on paper. You can have a copy of the photograph and the results if you want.
- We may take a second photograph sometimes to check that everything is working properly.
- We will take the stickers off your back and you will get dressed again.

The whole procedure will take about ten minutes. You can look at a child having this photograph taken on the ROH Orthopaedic Charity website, www.orthosurg.org.uk. Your photograph will be taken in private. The only people in the room with you will be the two researchers who run the measuring system. They are both mums and will make sure that you are well looked after. They will also measure your height and weight.

Do you get anything if you join in?

No, you don't. However, you will know that you are helping improve the care of children with spinal deformity in the future.

When you are in the senior school you will be able to learn about statistics using real data from the measurements done in this study.

What if we find that you have a curve in your spine?

We can still use your photographs because we need to compare results from children with straight spines and children with curved spines. If we find you have a curved spine that you did not know about then the doctor in charge of the research, Mr Gardner, will write a letter to your mum and dad suggesting that they take you to see your family doctor. Nobody else will see this letter apart from you and your mum and dad.

Are there any risks in taking part?

You might have sensitive skin which gets irritated by the stickers. We will ask you about this every time we take your photograph. If you feel your skin is too sensitive you can always decide not to take part.

Will anyone know that you are taking part?

Obviously your mum and dad, your teacher and your friends in your class will all know that you are taking part in the research. We will not tell anyone else. All the information collected about you in the course of the research will be kept confidential. The school will give you a special code which we will use in the database when we save your photograph. So we will know that all the photographs with that code are from the same person. We will be able to plot how your back changes through the years as you are growing but we will not know your name. We will need to know your birth date because we need to be able to calculate how old you are when each photograph is taken. We may also use your initials in case you forget what your code is.

What will happen to my photographs and back shape data?

The researchers will put the results from everyone together and analyse them using statistics. We will write reports for meetings and scientific magazines so that we can tell others about what we find out. No one who reads these reports will know what comes from you because all the backs from all the children taking part will be mixed up together.

Who is in charge of the research study?

Mr Gardner is the orthopaedic surgeon who is leading this research. He will be running the study for the ROH Orthopaedic Charity which is helping to pay for the research. Dr Berryman and Sister Baker will be taking the photographs.

Who decides if researchers can do studies like this

The ROH Orthopaedic Charity has asked some independent referees for their opinions about the study. All research in the NHS is also examined by an independent group of people called a Research Ethics Committee. This project has been reviewed and given a favourable opinion by the ???? Research Ethics Committee. We are only allowed to go ahead with studies like this when these people confirm that they think it is worthwhile.

What if something goes wrong?

There is not really anything that can go wrong in this study but if you are worried about anything then you or your mum and dad can talk to Mr Gardner. His telephone number is 0121-685-4083.

Appendix 2

The Royal Orthopaedic Hospital **NHS**
NHS Trust

Parent/Guardian Consent Form

Study Title: Normal back shape in adolescents

Project team: Mr A Gardner, Prof P Pynsent, Dr F Berryman, Sister D Baker, Prof J Fairbank

Please initial box

1. I confirm that I have read and understood the information sheet Version __________ dated __________ for the above study. I have had the opportunity to consider the information, to ask questions and have had my questions answered satisfactorily. ☐
2. I understand that my child's participation is voluntary and that he/she is free to withdraw at any time, without giving any reason. ☐
3. I agree that my child will take part in the above study. ☐
4. I understand that photographs of my child's back will be stored in a database. ☐
5. I understand that my child's photographs and data may be looked at by responsible individuals from the Royal Orthopaedic Hospital. I give permission for these individuals to have access to these records. ☐
6. I give permission that anonymised photographs of my child's back (from neck to hips, thus not identifiable) may be used in teaching, hospital reports, research papers and presentations to show the results of this study. ☐

Name of participant ____________________

Name of parent/guardian ____________________

Signature __________________ Date __________

Name of staff member who took consent ____________________

Signature __________________ Date __________

Version 1.0 17 June 2010 Page 1 of 1

Assent Form (Children 9–15 years old)

Study Title: Normal back shape in adolescents

Project team: Mr A Gardner, Prof P Pynsent, Dr F Berryman, Sister D Baker, Prof J Fairbank

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.

Have you read about the study on normal back shape (or has someone read something about it to you)?
Yes/No

Has someone explained to you what the project is about?
Yes/No

Do you understand what the project is about?
Yes/No

Have you been given a chance to ask questions?
Yes/No

Have you asked all the questions you want to ask?
Yes/No

Has someone answered all your questions?
Yes/No

Did you understand the answers?
Yes/No

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

Did you understand that the photographs are associated with a number in the database and not your name so that the researchers will not know who you are?
Yes/No

Would you like to join the study in measuring normal back shape?
Yes/No

Are you happy to have the photograph of your back taken every year until you reach 16 years old?
Yes/No

Are you happy for the researchers to use your photographs and data to calculate statistics about normal back shape?
Yes/No

Are you happy for your photographs to be used to illustrate reports, presentations and teaching materials (just from neck to hips, heads removed)?
Yes/No

Write your name here if you would like to join the study on normal back shape and you have circled yes to all the questions.

Your name ______________________________

Today's date ____________________________

If you would like to join the study on normal back shape and you have circled yes to all the questions but you would prefer your mum or dad to sign, ask one of them to sign here.

Parent's name ______________________________

Today's date ____________________________

The person who told you about the study has to sign here too.

Name of staff member who took consent _________________________

Signature _____________________ Date __________

Thank you very much for helping us

Appendix 3

GP Letter

Not required for this project.

Appendix 4

Declaration of Helsinki

WORLD MEDICAL ASSOCIATION DECLARATION OF HELSINKI

Ethical Principles
for

Adopted by the 18th WMA General Assembly
Helsinki, Finland, June 1964
and amended by the
29th WMA General Assembly, Tokyo, Japan, October 1975
35th WMA General Assembly, Venice, Italy, October 1983
41st WMA General Assembly, Hong Kong, September 1989
48th WMA General Assembly, Somerset West, Republic of South Africa, October 1996
and the
52nd WMA General Assembly, Edinburgh, Scotland, October 2000

A. INTRODUCTION

1. The World Medical Association has developed the Declaration of Helsinki as a statement of ethical principles to provide guidance to physicians and other participants in medical research involving human subjects. Medical research involving human subjects includes research on identifiable human material or identifiable data.

2. It is the duty of the physician to promote and safeguard the health of the people. The physician's knowledge and conscience are dedicated to the fulfilment of this duty.

3. The Declaration of Geneva of the World Medical Association binds the physician with the words, "The health of my patient will be my first consideration," and the International Code of Medical Ethics declares that, "A physician shall act only in the patient's interest when providing medical care which might have the effect of weakening the physical and mental condition of the patient."

4. Medical progress is based on research which ultimately must rest in part on experimentation involving human subjects.

5. In medical research on human subjects, considerations related to the well-being of the human subject should take precedence over the interests of science and society.

6. The primary purpose of medical research involving human subjects is to improve prophylactic, diagnostic and therapeutic procedures and the understanding of the aetiology and pathogenesis of disease. Even the best

proven prophylactic, diagnostic, and therapeutic methods must continuously be challenged through research for their effectiveness, efficiency, accessibility and quality.

7. In current medical practice and in medical research, most prophylactic, diagnostic and therapeutic procedures involve risks and burdens.

8. Medical research is subject to ethical standards that promote respect for all human beings and protect their health and rights. Some research populations are vulnerable and need special protection. The particular needs of the economically and medically disadvantaged must be recognized. Special attention is also required for those who cannot give or refuse consent for themselves, for those who may be subject to giving consent under duress, for those who will not benefit personally from the research and for those for whom the research is combined with care.

9. Research Investigators should be aware of the ethical, legal and regulatory requirements for research on human subjects in their own countries as well as applicable international requirements. No national ethical, legal or regulatory requirement should be allowed to reduce or eliminate any of the protections for human subjects set forth in this Declaration.

B. BASIC PRINCIPLES FOR ALL MEDICAL RESEARCH

10. It is the duty of the physician in medical research to protect the life, health, privacy, and dignity of the human subject.

11. Medical research involving human subjects must conform to generally accepted scientific principles, be based on a thorough knowledge of the scientific literature, other relevant sources of information, and on adequate laboratory and, where appropriate, animal experimentation.

12. Appropriate caution must be exercised in the conduct of research which may affect the environment, and the welfare of animals used for research must be respected.

13. The design and performance of each experimental procedure involving human subjects should be clearly formulated in an experimental protocol. This protocol should be submitted for consideration, comment, guidance, and where appropriate, approval to a specially appointed ethical review committee, which must be independent of the investigator, the sponsor or any other kind of undue influence. This independent committee should be in conformity with the laws and regulations of the country in which the research experiment is performed. The committee has the right to monitor ongoing trials. The researcher has the obligation to provide monitoring information to the committee, especially any serious adverse events. The researcher should also submit to the committee, for review, information regarding funding, sponsors, institutional affiliations, other potential conflicts of interest and incentives for subjects.

14. The research protocol should always contain a statement of the ethical considerations involved and should indicate that there is compliance with the principles enunciated in this Declaration.

15. Medical research involving human subjects should be conducted only by scientifically qualified persons and under the supervision of a clinically competent medical person. The responsibility for the human subject must always rest with a medically qualified person and never rest on the subject of the research, even though the subject has given consent.

16. Every medical research project involving human subjects should be preceded by careful assessment of predictable risks and burdens in comparison with foreseeable benefits to the subject or to others. This does not preclude the participation of healthy volunteers in medical research. The design of all studies should be publicly available.

17. Physicians should abstain from engaging in research projects involving human subjects unless they are confident that the risks involved have been adequately assessed and can be satisfactorily managed. Physicians should cease any investigation if the risks are found to outweigh the potential benefits or if there is conclusive proof of positive and beneficial results.

18. Medical research involving human subjects should only be conducted if the importance of the objective outweighs the inherent risks and burdens to the subject. This is especially important when the human subjects are healthy volunteers.

19. Medical research is only justified if there is a reasonable likelihood that the populations in which the research is carried out stand to benefit from the results of the research.

20. The subjects must be volunteers and informed participants in the research project.

21. The right of research subjects to safeguard their integrity must always be respected. Every precaution should be taken to respect the privacy of the subject, the confidentiality of the patient’s information and to minimise the impact of the study on the subject's physical and mental integrity and on the personality of the subject.

22. In any research on human beings, each potential subject must be adequately informed of the aims, methods, sources of funding, any possible conflicts of interest, institutional affiliations of the researcher, the anticipated benefits and potential risks of the study and the discomfort it may entail. The subject should be informed of the right to abstain from participation in the study or to withdraw consent to participate at any time without reprisal. After ensuring that the subject has understood the information, the physician should then obtain the subject's freely given informed consent, preferably in writing. If

the consent cannot be obtained in writing, the non-written consent must be formally documented and witnessed.

23. When obtaining informed consent for the research project the physician should be particularly cautious if the subject is in a dependent relationship with the physician or may consent under duress. In that case the informed consent should be obtained by a well-informed physician who is not engaged in the investigation and who is completely independent of this relationship.

24. For a research subject who is legally incompetent, physically or mentally incapable of giving consent or is a legally incompetent minor, the investigator must obtain informed consent from the legally authorized representative in accordance with applicable law. These groups should not be included in research unless the research is necessary to promote the health of the population represented and this research cannot instead be performed on legally competent persons.

25. When a subject deemed legally incompetent, such as a minor child, is able to give assent to decisions about participation in research, the investigator must obtain that assent in addition to the consent of the legally authorized representative.

26. Research on individuals from whom it is not possible to obtain consent, including proxy or advance consent, should be done only if the physical/ mental condition that prevents obtaining informed consent is a necessary characteristic of the research population. The specific reasons for involving research subjects with a condition that renders them unable to give informed consent should be stated in the experimental protocol for consideration and approval of the review committee. The protocol should state that consent to remain in the research should be obtained as soon as possible from the individual or a legally authorized surrogate.

27. Both authors and publishers have ethical obligations. In publication of the results of research, the investigators are obliged to preserve the accuracy of the results. Negative as well as positive results should be published or otherwise publicly available. Sources of funding, institutional affiliations and any possible conflicts of interest should be declared in the publication. Reports of experimentation not in accordance with the principles laid down in this Declaration should not be accepted for publication.

C. ADDITIONAL PRINCIPLES FOR MEDICAL RESEARCH COMBINED WITH MEDICAL CARE

28. The physician may combine medical research with medical care, only to the extent that the research is justified by its potential prophylactic, diagnostic or therapeutic value. When medical research is combined with medical care, additional standards apply to protect the patients who are research subjects.

29. The benefits, risks, burdens and effectiveness of a new method should be tested against those of the best current prophylactic, diagnostic, and therapeutic methods. This does not exclude the use of placebo, or no treatment, in studies where no proven prophylactic, diagnostic or therapeutic method exists.

30. At the conclusion of the study, every patient entered into the study should be assured of access to the best proven prophylactic, diagnostic and therapeutic methods identified by the study.

31. The physician should fully inform the patient which aspects of the care are related to the research. The refusal of a patient to participate in a study must never interfere with the patient physician relationship.

32. In the treatment of a patient, where proven prophylactic, diagnostic and therapeutic methods do not exist or have been ineffective, the physician, with informed consent from the patient, must be free to use unproven or new prophylactic, diagnostic and therapeutic measures, if in the physician's judgement it offers hope of saving life, re-establishing health or alleviating suffering. Where possible, these measures should be made the object of research, designed to evaluate their safety and efficacy. In all cases, new information should be recorded and, where appropriate, published. The other relevant guidelines of this Declaration should be followed.

The Royal Orthopaedic Hospital NHS Trust

Research and Teaching Centre, Royal Orthopaedic Hospital, Northfield, Birmingham, B31 2AP

Study: Normal back shape in adolescents

Parent Information Sheet

We would like to invite your child to take part in a study investigating the normal shape of the back in adolescents. Before you decide if you would like your child to take part you need to understand why the research is being done and what it would involve. Please read the following information carefully. Talk to others about the study if you wish. Ask us if there is anything that is not clear or if you would like more information.

What is the purpose of this study?

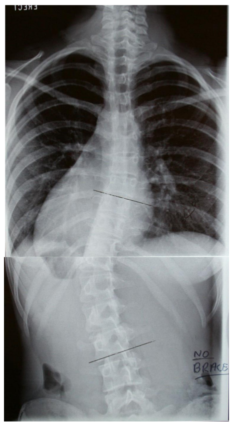


Figure 1: X-ray showing a curved spine.

Scoliosis is curvature of the spine. When the spine bends it causes the ribs to rotate, producing a hump on the back. Although the curvature of the spine can be measured using X-rays (see Figure 1), this does not give any information about the surface of the back. The shape of the surface of the back can be measured using surface topography which produces a picture similar to an Ordnance Survey contour map. We are using surface topography regularly in our spinal clinic to monitor back shape changes in patients with scoliosis but we have very little information about the shape and variability in normal adolescent backs. This information is important as surgical correction of a scoliosis alters the shape of the back of the chest; surgeons need to have accurate information about normal backs to get the best possible results for the patient.

We would therefore like to measure back shape in a large group of normal adolescents, annually over a period of seven years. This will give us statistical information about the shape and variability in normal backs. The measurements over several years will enable us to investigate how an individual's back changes shape as he/she grows through the pubertal growth spurt. We can then use this data as a benchmark for changes in patients with scoliosis.

We will measure the shape of the back using a surface topography system called ISIS2 which was developed at our hospital.

What is ISIS2?

ISIS stands for Integrated Shape Imaging System; the '2' is because this is a modern version of technology that was first developed in the 1980s. The ISIS2 system can calculate the shape of the back from a simple digital photograph. This is accomplished by shining a grid of horizontal lines onto the back (Figure 2), photographing the back and then using special mathematical methods to convert the distortions of the grid into a three dimensional map of the back (Figure 3). Various clinical parameters are then calculated from the surface data. All data collected are stored electronically and can be retrieved at subsequent measurements. This allows the researcher to see how the back has changed with time.

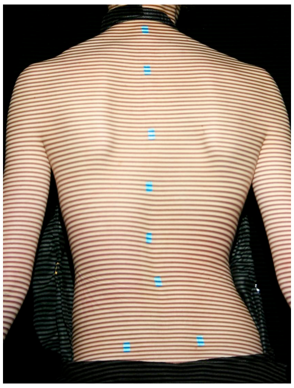


Figure 2: Back photograph of child with scoliosis showing the stickers and projected grid

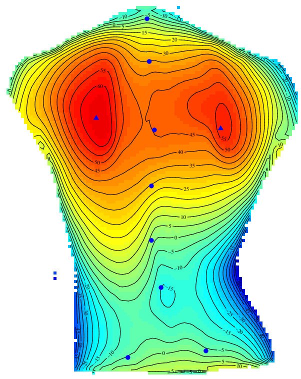


Figure 3: Contour map

The development of ISIS2 enables us to measure back shape simply by taking a photograph. This means we can carry out an investigation on the population without using harmful radiation or contacting physical methods that take a long time.

Why have we chosen your child?

The age range of interest is 9–15 years because this is the period when a child grows most quickly. In scoliosis this is the time that a curve is most likely to progress. Bromsgrove School has been chosen because there is little change in the pupils attending from year to year which should limit the drop out rate of the study. So long as your child can stand unsupported for a few minutes for the photograph to be taken, we would like him/her to take part.

Does your child have to take part?

No. It is up to you and your child to decide whether he/she would like to take part in the study. If you and your child decide that he/she would like to take part, you will be asked to sign a consent form and your child will be asked to sign an assent form. If you decide that your child will take part now, you can still change your mind later and withdraw your child from the study at any time.

What will participants need to do?

Once a year a photograph will be taken of your child's back using the ISIS2 system while he/she remains in the 9–15 years age group. This will involve the following procedure:

- Your child will need to remove the clothes from the upper body and loosen his/her trousers or skirt so that the back is exposed from the neck to the bottom of the back.
- Hair may need to be tied up so that it does not cover the back.
- The child will wear a black neck band and apron to provide clear limits for the area of the photograph to be analysed.
- For female participants, a special gown will be provided to cover their front.
- Some small coloured paper stickers will be placed on the back marking certain bony landmarks.
- The child will be photographed with his/her back to the camera.
- The computer will analyse the photograph and present clinical information about the back on screen and on paper. A copy of the report will be given to your child to keep for your records.
- A second photograph may be taken and analysed to check for variability in stance.
- The stickers will be removed from your child's back and he/she will get dressed again.

The whole procedure is likely to take about ten minutes. A video of such a measurement is available at the ROH Orthopaedic Charity website, www.orthosurg.org.uk.

All photographs and the data calculated about the shape of your back will be stored in a database. This database will be confidential and it will be stored and used in accordance with the Data Protection Act.

What are the possible benefits of taking part?

The main benefit is the knowledge that your child is helping advance the care of children with spinal deformity.

If there were unrecognised spinal deformities in the children photographed, these would be identified. Where surface topography results indicate a possible spinal deformity, a confidential letter will be written by the Chief Investigator recommending that the child and his/her parents consult their GP.

We intend to involve the senior students in some statistical analysis of the anonymised data so that they can gain some experience of the use of advanced statistics in a real situation.

What are the disadvantages or risks in taking part?

The main risk associated with the measurements is allergy to the stickers used to mark the bony landmarks. Your child will be asked every time a photograph is taken about skin sensitivity.

There may be some inconvenience with loss of school time; however, by working with the school the photographs will be taken at suitable times to minimise any loss of lesson time. With advance notice, this would then not lead to a loss in education.

There is also the possibility that a few participants may be identified as having a spinal deformity which later turns out not to be the case (false positives) thus leading to some needless worry before their fears are allayed. This may result in a referral to an NHS spinal deformity clinic. To ensure low rates of false positives only curves of greater than 20 degrees will be referred to the GP.

Will my child's participation in this study be kept confidential?

All information collected about your child in the course of the research will be kept confidential. The school will allocate a unique code to each child who participates so that data sets from the same subject can be compared from year to year. The database will identify the participant by this code, his/her initials, sex and date of birth (both sex and date of birth are needed for the subsequent statistical analysis). Even if a child cannot tell us at later measurements what his/her unique code is, we will be able to use initials and date of birth to retrieve the code from the system without actually recording full names. The data is thus fully anonymised. The database will be stored on an encrypted computer at the Royal Orthopaedic Hospital. Professor Pynsent will be its custodian, only allowing access to the data to the researchers involved in the project. If a previously unrecognised spinal deformity is identified, then Mr Gardner will obtain the name of the child in question from the school nurse and a letter will be sent directly to the parents recommending a review with their GP.

What will happen to the results of the study?

The researchers will publish their results at scientific conferences worldwide and in respected scientific journals. We will also publish information about the results of the research on the ROH Orthopaedic Charity website (www.orthosurg.org.uk).

Who is organising and funding the research study?

We have received a grant from the ROH Orthopaedic Charity. The principal researchers are employed by the Royal Orthopaedic Hospital. ISIS2 was developed at the Royal Orthopaedic Hospital in association with the Nuffield Orthopaedic Centre, Oxford but it is not a commercialised system and is supplied at cost price to any other interested users. No parties will gain financially from its use in this project.

Who has reviewed the research study

We have obtained the views of independent referees in setting up this study. All research in the NHS is also examined by an independent group of people called a Research Ethics Committee. This project has been reviewed and given a favourable opinion by the South Birmingham Research Ethics Committee (Reference: 11/H1207/10).

Who will be involved in conducting the study?

The research team involved in this project comprises:

- Mr. Adrian Gardner, Spinal Surgeon, Royal Orthopaedic Hospital.
- Dr. Fiona Berryman, Clinical Scientist, Royal Orthopaedic Hospital.
- Sister Delia Baker, Clinical Outcomes Sister, Royal Orthopaedic Hospital.
- Prof. Paul Pynsent, Clinical Scientist, Royal Orthopaedic Hospital.

We have also discussed the project with Prof. Jeremy Fairbank, Spinal Surgeon, Nuffield Orthopaedic Centre and taken advice from him.

For more information contact:

- Mr Adrian Gardner Tel: 0121-685-4083 E-mail: adrian.gardner@nhs.net
- Dr Fiona Berryman Tel: [redacted] E-mail: [redacted]

If you have any concerns or complaints which Mr Gardner and Dr Berryman cannot deal with, then please contact the Complaints Department:

- Telephone: 0121-685-4016
- E-mail: roh-tr.complaints@nhs.net
- Letter: Chief Executive, Royal Orthopaedic Hospital, Bristol Road South, Northfield, Birmingham, B31 2AP

The Royal Orthopaedic Hospital NHS
NHS Trust

Research and Teaching Centre, Royal Orthopaedic Hospital, Northfield, Birmingham, B31 2AP

Study: Normal back shape in adolescents

Information Sheet for children aged 9–15 years old

We would like to invite you to take part in a study measuring the shape of the back in children. The purpose of the research is to help people who have scoliosis, a condition where the back is twisted. We need you to read this information sheet before you decide if you would like to take part. You can ask questions and talk to your family and friends about the study before you decide. Your mum or your dad will also have an information sheet to read and they will also have to give their permission if you want to join the study.

Why are we doing this research?

Scoliosis is medical condition where the spine or backbone curves and twists so that the shape changes from what it should be (see Figure 1). When the spine twists it often makes one side of the back stick out more than the other. We measure the back shape of children who come to the hospital with scoliosis because the shape helps us to decide what treatment these children should have. However, we have very little information about the shape in normal children's backs and how that changes as they are growing. We would like to measure your back shape each year until you are 16 to help us improve treatment for other children with scoliosis. If you agree to join the study, we will measure the shape of your back by taking a digital photograph of it using a system called ISIS2.

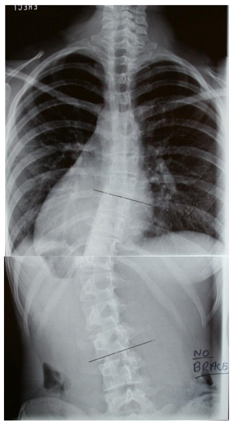


Figure 1: X-ray showing a curved spine.

What is ISIS2?

ISIS stands for Integrated Shape Imaging System; the '2' is because this is a modern version of technology that was first developed in the 1980s. The ISIS2 system can calculate the shape of the back from a simple digital photograph. We do this by shining a pattern of horizontal lines onto the back (Figure 2), photographing the back and then doing some special analysis to give a three dimensional map of the back (Figure 3). This figure is similar to an Ordnance Survey map of hills.

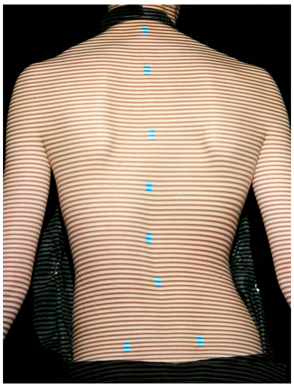


Figure 2: Back photograph of child with scoliosis showing the stickers and projected lines

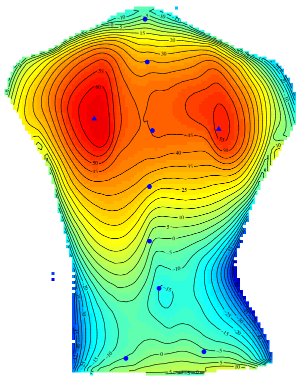


Figure 3: Contour map

ISIS2 lets us measure back shape simply by taking a photograph. This means we can measure lots of children very quickly.

Why have we chosen you?

We want to measure children aged 9–15 years old which is the age you are. We have chosen Bromsgrove School because the pupils attending from year to year do not change very much. If you can stand for a few minutes for the photograph to be taken, we would like you to take part.

Do you have to take part?

No, you don't. It is up to you. You can decide to take part and then change your mind later if you want to. No one will mind if you do not take part or if you change your mind later.

What will happen if you do take part?

Once a year a normal digital photograph will be taken of your back while you are aged between 9 and 15 years old. The camera we use is just like one you may have at home.

This is what happens when the photograph is taken:

- You remove the clothes from your top half (waist upwards) and loosen your trousers or skirt so that we can see all of your back.
- If you have long hair we will ask you to tie it up so that it does not cover any of your back.
- You will wear a black neck band and apron to make it easy for the computer to analyse the photograph.
- Girls will also wear a special gown which covers their front.
- We will stick some small coloured paper stickers on your back.
- We will take a photograph of your back while you stand in a special stand with your arms away from your sides.
- The computer will analyse the photograph and present the results on the computer screen and on paper. You can have a copy of the photograph and the results if you want.
- We may take a second photograph sometimes to check that everything is working properly.
- We will take the stickers off your back and you will get dressed again.

The whole procedure will take about ten minutes. You can look at a child having this photograph taken on the ROH Orthopaedic Charity website, www.orthosurg.org.uk. Your photograph will be taken in private. The only people in the room with you will be the two researchers who run the measuring system. They are both mums and will make sure that you are well looked after. They will also measure your height and weight.

Do you get anything if you join in?

No, you don't. However, you will know that you are helping improve the care of children with spinal deformity in the future.

When you are in the senior school you will be able to learn about statistics using real data from the measurements done in this study.

What if we find that you have a curve in your spine?

We can still use your photographs because we need to compare results from children with straight spines and children with curved spines. If we find you have a curved spine that you did not know about then the doctor in charge of the research, Mr Gardner, will write a letter to your mum and dad suggesting that they take you to see your family doctor. Nobody else will see this letter apart from you and your mum and dad.

Are there any risks in taking part?

You might have sensitive skin which gets irritated by the stickers. We will ask you about this every time we take your photograph. If you feel your skin is too sensitive you can always decide not to take part.

Will anyone know that you are taking part?

Obviously your mum and dad, your teacher and your friends in your class will all know that you are taking part in the research. We will not tell anyone else. All the information collected about you in the course of the research will be kept confidential. The school will give you a special code which we will use in the database when we save your photograph. So we will know that all the photographs with that code are from the same person. We will be able to plot how your back changes through the years as you are growing but we will not know your name. We will need to know your birth date because we need to be able to calculate how old you are when each photograph is taken. We may also use your initials in case you forget what your code is.

What will happen to my photographs and back shape data?

The researchers will put the results from everyone together and analyse them using statistics. We will write reports for meetings and scientific magazines so that we can tell others about what we find out. No one who reads these reports will know what comes from you because all the backs from all the children taking part will be mixed up together.

Who is in charge of the research study?

Mr Gardner is the orthopaedic surgeon who is leading this research. He will be running the study for the ROH Orthopaedic Charity which is helping to pay for the research. Dr Berryman and Sister Baker will be taking the photographs.

Who decides if researchers can do studies like this

The ROH Orthopaedic Charity has asked some independent referees for their opinions about the study. All research in the NHS is also examined by an independent group of people called a Research Ethics Committee. This project has been reviewed and given a favourable opinion by the South Birmingham Research Ethics Committee. We are only allowed to go ahead with studies like this when these people confirm that they think it is worthwhile.

What if something goes wrong?

There is not really anything that can go wrong in this study but if you are worried about anything then you or your mum and dad can talk to Mr Gardner. His telephone number is 0121-685-4083.

The analysis of back shape in scoliosis using ISIS2 surface topography

Protocol

Chief Investigator: Mr. Adrian Gardner

Co-Investigators: Prof. Paul Pynsent
Prof. Joanne Wilton
Dr. Fiona Berryman

Version: 2.0
Dated: 31 August 2017

Table of Contents

1. BACKGROUND AND RATIONALE

1. BACKGROUND

The aim of this project is to measure the shape of the back in patients with Adolescent Idiopathic Scoliosis (AIS) in their adolescent or early adult years and compare it to that of children without scoliosis. In this setting and from here on after "the back" refers to the spine and posterior chest wall from the nape of the neck to the top of the pelvis. Although there is already data published on spinal shape in scoliosis using a variety of surface topography techniques, less is known about the posterior chest wall in the presence of scoliosis or in the normal growing population.

The hospital data will be of both pre operative and post operative subjects and will allow longitudinal analysis prior to and after surgery. This will allow the development of the imaging technique to replace the requirements for serial radiographs in the treatment of scoliosis.

This data will then be analysed with equivalent data from an already running approved research project examining the surface shape in normal children without spinal deformity (ref: 11/H1207/10). This would allow an assessment of what the normal amount of asymmetry in the back is and when this normal asymmetry becomes abnormal or the point when the pathology becomes apparent.

The knowledge of what is 'normal' will also help to guide the surgical correction of scoliosis from the external visual appearance which is the point of most interest and concern to this patient population. Correcting a scoliosis curve to give a straight spine on a radiograph may leave the child with a visually unappealing rib hump and a poor outcome from surgery. Knowledge of the surface shape would allow an alteration in technique to give the best possible cosmetic appearance and thus outcome.

This project aims to analyse retrospectively the ISIS2 surface topography data already collected from patients with scoliosis as part of routine clinical practice at the Royal Orthopaedic Hospital and then compare it to similar data already collected on normal adolescents from another research project.

1. STUDY RATIONALE

1. Justification for patient population

The patient population for this study is all patients who have undergone ISIS2 surface topography for AIS either whilst in their adolescent years or in early adult life, differentiating between AIS in adulthood and adult degenerative scoliosis.

Patients who have been treated with the use of a brace will be excluded from this population as this may affect the surface shape through pressure being applied to the chest wall.

At the last count in October 2014 there were already measured and stored 1777 measurements in 1088 patients.

2. Justification for study design

The study design is retrospective. The data that exists currently includes both pre and post operative surface topography measurements in patients between 9 and 40 years of age. This data will be analysed for individual and group longitudinal trends stratified by age. Throughout this project, more data will be acquired from the ongoing clinical work at the Royal Orthopaedic Hospital adding to the number of measurements available for analysis.

3. Potential risks for participants

There are no risks to participants from the project as they will have already undergone the surface assessment as part of their ongoing clinical care. They will already be aware of their results.

4. Potential benefits for participants

There are no direct benefits for participants in this study for patients whose data is already stored. For younger patients who have already had surface topography measurements taken and who will be having ongoing imaging whilst at the Royal Orthopaedic Hospital as part of their care this research will help to reduce the total radiation dose over the course of their treatment.

2. AIMS, OBJECTIVES AND ENDPOINTS

2.1. AIMS AND OBJECTIVES

The aim of this research is to study the shape of the back in a group of patients with AIS through longitudinal analysis in both a pre and post operative stage and compare it with similar data from adolescents with normal backs.

The primary objective:

To measure the statistical variation of back shape in a group of scoliosis patients and compare it with similar data from adolescents with normal backs.

The secondary objectives:

To measure how the back shape changes with changes in the severity of scoliosis, the effects of surgery on that shape and any changes seen after surgery.

2.2. ENDPOINTS

Primary

endpoint

The study will end at the end of the study of normal backs (noted above).

Secondary endpoints

None

3. TRIAL DESIGN

This is a retrospective longitudinal observational study of patients with scoliosis from the Royal Orthopaedic Hospital. The study will involve the analysis of surface topography measurements taken in comparison with measurements from whole spine radiographs taken in the same clinic visit.

The purpose of this research is to study how the spine and posterior chest wall change in the presence of a developing scoliosis and then subsequent treatment of that scoliosis.

Surface topography using the ISIS2 system creates a contour map representation of the whole of the back including the spine and rib cage. The system allows recording and analysis of all aspects of shape, dimensions and asymmetry in a risk free, quick and easy photograph. ISIS2 was developed at the Royal Orthopaedic Hospital in association with the Nuffield Orthopaedic Centre, Oxford [1]. ISIS2 is currently used on a regular basis in NHS clinical practice at several centres.

The photographs taken using the ISIS2 system are analysed using Fourier transform profilometry to calculate the three-dimensional shape of the back from the two-dimensional photograph. The software incorporates innovative mathematical methods with the algorithms designed to minimise the errors found in earlier topography systems [1-4]. The system presents graphical information about the back shape in the transverse, coronal and sagittal planes. It also calculates a range of clinical parameters from the shape including back length, pelvic rotation, flexion/extension angle, coronal imbalance, lateral asymmetry (the ISIS2 estimate of spinal curve in the coronal plane – Cobb angle proxy), volumetric asymmetry, kyphosis and lordosis angles. All numerical and graphical results are stored to a database so that later statistical analysis can be carried out on the measured parameters. Height, sitting height and weight will also be collected and input to the database. By using all whole spine imaging taken at the same clinic visit, measured ISIS2 parameters can be compared to the current gold standard for the measurement of the magnitude of a scoliosis which is the Cobb angle [5].

The measured parameters will be plotted against age; regression methods will be used to estimate an analytical curve to fit the observed parameters as a function of age and other covariates. 95% confidence intervals will also be calculated. Measured parameters will be analysed longitudinally and with reference to surgical treatment.

4. ELIGIBILITY

4.1. INCLUSION CRITERIA

Any patient who has AIS and has had ISIS2 surface topography as part of their routine clinical care whilst under the Royal Orthopaedic Hospital is eligible for inclusion.

4.2. EXCLUSION CRITERIA

The exclusion criteria are:

- If the patient is known to have been treated with a brace which could by its mechanism of action lead to a deformation of the chest shape independent of the scoliosis.
- A patient who has had surface topography imaging who is known to have a subtype of adolescent scoliosis which is not idiopathic related to other diagnoses.

The data will be excluded from the statistical analysis if it is established subsequent to surface topography imaging that:

- The patient has previously undergone a thoracotomy or median sternotomy prior to their episode of spinal care which through its effects on chest growth could alter the shape of the chest independent of the scoliosis (thoracogenic scoliosis).

5. SCREENING, RECRUITMENT AND CONSENT

5.1. SCREENING

Prior to study entry, each patient's inclusion/exclusion criteria will be checked.

5.2. RECRUITMENT PHASE

All suitably anonymised eligible surface topography measurements will be recruited.

5.3. CONSENT

Written consent will not be taken as this is a retrospective analysis of data already obtained as part of the routine clinical assessment of patients with scoliosis (analogous to the retrospective review of a series of radiographs).

6. RANDOMISATION

Randomisation is not required for this observational study.

7. MEASUREMENT DETAILS

7.1. SUMMARY

Background information in to the process of image and data capture

ISIS2 measurement simply involves taking a normal digital photograph of the child's back. The back must be exposed from head to hips. The photographs taken are only of the back; the face is never seen. Figure 1 shows an example of an ISIS2 photograph.

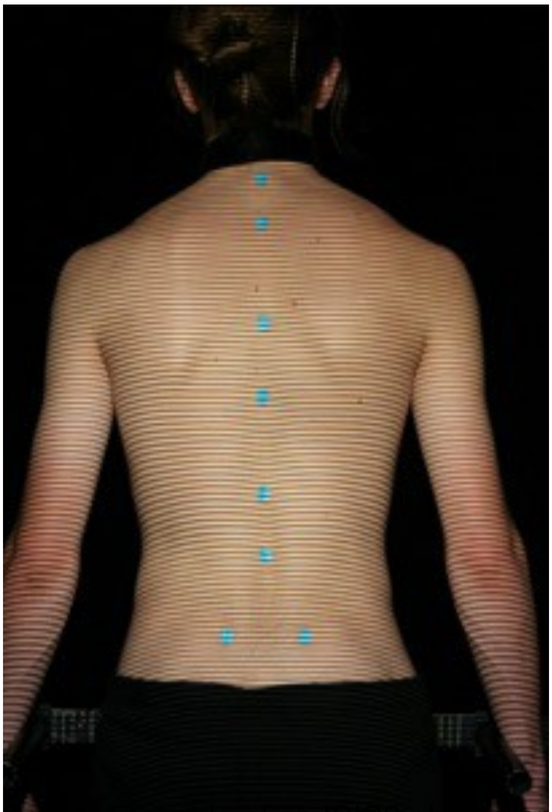


Figure 1: Example of an ISIS2 photograph

The procedure for taking the photographs is as follows:

- Clothes will need to be removed from the top half of the body and trousers/skirt will need to be loosened so that the back is exposed from the neck to the bottom of the back.

- Long hair will be tied up so that it does not cover the back.

- A special black neck band and apron are worn to provide clear limits for the area of the photograph to be analysed.

- For females a special gown is provided to cover the subject's front.

- Some small coloured paper stickers are placed on the back marking certain bony landmarks.

- A photograph of the back is taken.

- The picture is analysed and a report created. The subject will be given a copy if they wish.

- A second photograph may be taken to check for differences in stance.

- The stickers are removed and the subject can dress again.

- Basic stature measurements of height, sitting height and weight will be recorded.

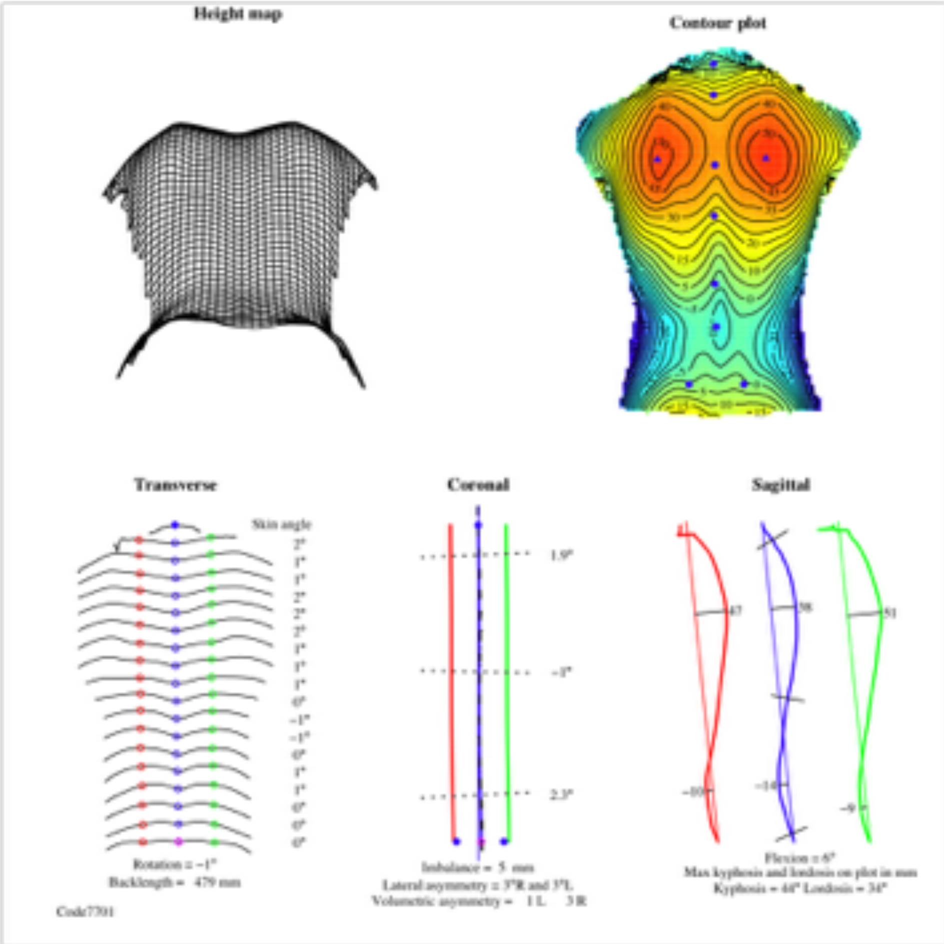
It will take approximately ten minutes to complete all of the above stages.

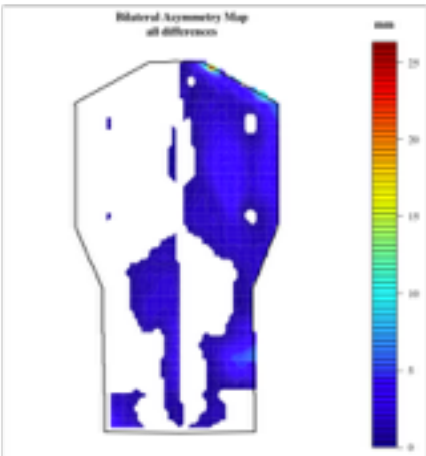
The computer software is part of the ISIS2 equipment so analysis of the photograph to provide surface topography data and clinical parameters is done immediately the photograph is taken.

An example of the report generated by the system is shown in Figure 2.

The measurements are currently taken in a specific location within the Royal Orthopaedic Hospital and stored securely on encrypted storage within the hospital.

Date: 22/01/2009 Time: 14:58:52 Patient:Straight Back DOB: 18/07/1990 Sex: m ID No: 5678





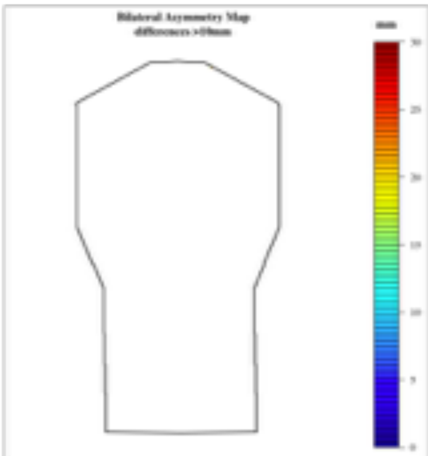


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Figure 2: Example of report from ISIS2

Further use of stored pictures and data for this project

The data is currently stored in two forms. First there is a spreadsheet file containing numerical values which represent the measured parameters which have previously deemed to be of clinical importance. Second is information about the surface which is created from the digital photograph and allows the generation of the numerical data.

Whilst initial analysis will focus on the already measured and stored parameters it is likely that there will be a need to investigate as yet unquantified aspects of the shape of the back. This can be done by regenerating and then reanalysing the shape of the back from the stored surface information and photograph having identified a point of interest.

This project will require access to both forms of the stored information (data points and photographs for recreating the surface for further analysis) to fully assess the shape of the back in comparison with similar data from adolescents with normal backs.

There will also be a need to measure all whole spine imaging series in a longitudinal manner to be able to compare the 'gold standard' of the Cobb angle and other imaging parameters to the surface topography parameters [5]. This will allow for the analysis and comparison of the different imaging techniques.

7.2. INVESTIGATIONAL THERAPY ARM

Not applicable to this study.

8. ASSESSMENTS/ DATA COLLECTION

8.1. VISIT SCHEDULE

Data will be collected when surface topography measurements are made when a patient returns to clinic for review. By necessity the time between these will vary depending on the individual case

8.2. SERIOUS ADVERSE EVENTS

As this is a retrospective study of already stored data, an adverse event cannot occur.

9. DATA MANAGEMENT

9.1. DATA COLLECTION

The data are already collected and stored on the Royal Orthopaedic Hospital Research and Teaching department computer system where the ISIS2 system is located. All radiographic data is already stored on the hospitals PACS system. Access to both systems is password controlled.

9.2. DATABASE MANAGEMENT AND QUALITY CONTROL

The data of all possible participants are currently stored under identifying R numbers (Royal Orthopaedic Hospital hospital numbers) with names and dates of birth. This is also true of any radiographs. These identifiers are required to be able to marry surface topography and radiographic data. Once done the database will be anonymised for analysis. To allow for longitudinal analysis each individual will have a unique code number, a catalogue of which will be kept securely at the Royal Orthopaedic Hospital.

The database will contain the back surface data, clinical parameters associated with the surface, the photographs, height, sitting height, weight and basic information about the volunteer namely gender, date of birth and the unique code.

On completion of the study, the data will be stored in encrypted files at the Royal Orthopaedic Hospital with access only to the study team. All data will be handled, computerised and stored in accordance with the Data Protection Act 1998. Quality control will be maintained through adherence to ICH GCP and the R&D Quality Assurance Process.