RISK MARKERS FOR SELF-INJURIOUS BEHAVIOUR IN CHILDREN WITH INTELLECTUAL DISABILITY

by

Catherine Steenfeldt-Kristensen

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Department of Clinical Psychology
School of Psychology
The University of Birmingham
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OVERVIEW

This thesis consists of two volumes and is submitted by Catherine Steenfeldt-Kristensen for the Clinical Psychology Doctorate at the University of Birmingham. Volume One comprises the research component of the doctorate and contains three papers. The first paper is a meta-analytic review of the prevalence of self-injurious behaviour in autism spectrum disorder. The second paper is an empirical study of the risk markers associated with self-injurious behaviour in children with an intellectual disability or developmental delay. The final paper is an executive summary which provides an overview of the two preceding papers. The executive summary will be used to disseminate the findings of the meta-analysis and empirical paper to families and professionals.

Volume Two of the thesis consists of five clinical practice reports that were completed over the course of the doctorate. The first report describes the assessment and formulation of a young boy with separation anxiety. His difficulties were formulated using cognitive-behaviour and systemic models. The second report describes a service evaluation exploring the experience of staff in adhering to the NICE guidance on mental health and intellectual disabilities. The third report presents a time series analysis conducted to measure change in symptomology in a woman with social anxiety. The fourth report details the assessment, formulation, intervention and evaluation of cognitive-behavioural therapy for a man with generalized anxiety disorder. The final report presents an abstract of an oral presentation case study.

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CHAPTER 1

The Prevalence of Self-Injurious Behaviour in Autism Spectrum Disorder: A Meta-Analytic Study

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1.1 Abstract

Background: Self-injurious behaviour is reported to be common in individuals with autism spectrum disorder (ASD) and is associated with poor outcomes. However, current prevalence estimates of self-injury in ASD are highly varied and the influence of person and study characteristics on prevalence and topography of self-injury in ASD is unknown. Therefore, the present meta-analysis seeks to describe and evaluate the current literature estimating the prevalence of self-injurious behaviour in ASD to produce more robust estimates.

Methods: A literature search identified 37 studies reporting the prevalence of self-injurious behaviour in ASD. Reliable quality criteria were developed and a quality weighting assigned to the studies to weight the prevalence estimates from the most robust studies more heavily. Data from the 37 papers were extracted and included in the meta-analysis. Pooled prevalence estimates for self-injury in ASD and different topographies of self-injury were generated. Meta-regression analyses assessed the impact of study and person characteristics on prevalence rates.

Results: The pooled prevalence of self-injury in ASD was estimated at 42% with confidence intervals between 0.38 and 0.47. Meta-regression revealed no association between study quality, participant intellectual disability or participant age and estimated prevalence rates of self-injury. However, gender was associated with self-injury prevalence, with females obtaining higher prevalence rates than males ($p = .013$). Hand-hitting was the most common topography of self-injurious behaviour, whereas self-cutting was found to be the least common. The presence of an intellectual disability was found to influence prevalence of some topographies of self-injury, specifically hair pulling and self-scratching topographies were

more common when intellectual disability was present. In contrast, gender and age were found to have no effect on prevalence of specific topographies of self-injury.

Discussion: Results are discussed in relation to existing literature, service provision for individuals with ASD with and without intellectual disability and areas for future research.

1.2. Introduction

Autism Spectrum Disorder[1] (ASD) is a neurodevelopmental disorder characterised by persistent deficits in social interaction and communication, as well as restricted or repetitive patterns of thoughts, behaviours and interests (DSM-V, American Psychiatric Association, 2013). Symptoms typically emerge in early childhood and persist across the lifespan. There is some variability in the reported prevalence rates of ASD with recent population estimates ranging from 1 in 100 (Baird, Simonoff, Pickles, Chandler, Loucas, Meldrum *et al*., 2006; Center for Disease Control, 2009) to 1 in 68 (Center for Disease Control, 2014) and higher rates of ASD observed in males than in females (Baird *et al*., 2006; Fombonne, 2009; Saracino, Noseworthy, Steiman, Reisinger & Fombonne, 2010; Volkmar, Szatmari & Sparrow, 1993). Prevalence rates of ASD have increased significantly in a short period of time which some researchers have suggested may be a result of changes in diagnostic and screening methods, increased community awareness and increased knowledge of the risk factors associated with ASD (Newschaffer, Falb, & Gurney, 2005; Rice, Rosanoff, Dawson, Durkin, Croen, Singer *et al*., 2012). Despite differences in the possible reasons for this observed increase in recent years, and the variability in reported prevalence rates of ASD, it is well established that the consequences and co-morbidities of autism pose a significant challenge for individuals, families and services (Altiere & von Kluge, 2009; Hastings & Brown, 2002; Hastings, 2003; Knapp, Romeo, & Beecham, 2009).

Some individuals with ASD are able to lead independent lives, whilst for others the impact of core symptomatology and co-morbidities can be extremely detrimental to their quality of life

[1] For the purpose of this meta-analysis, the author has used diagnostic terminology such as "disorder" and "disability" to describe ASD. These terms are derived from diagnostic manuals; however, it is recognised that these are not the preferred terminology for some individuals with autism. Additionally, whilst for the purposes of this thesis, person-first language is used, it is recognised that within the autism community there is also disagreement about the use of person-first language (for a full review see Kenny *et al.*, 2016).

(Farley, McMahon, Fombonne, Jenson, Miller & Gardner *et al*., 2009; van Heijst & Geurts, 2015). For example, individuals with ASD are at a greater risk of having co-morbid medical and psychiatric conditions such as sleep disorders (Mannion, Leader & Healy, 2013), mental health problems such as anxiety and depression (Bradley, Summers, Wood & Bryson, 2004; Brereton, Tonge & Einfeld, 2006; Ghaziuddin, Tsai, & Ghaziuddin, 1992), and intellectual disability (Matson & Shoemaker, 2009). In addition, the presence of ASD has been associated with increased inpatient hospital admissions (Cowley, Newton, Sturmey, Bouras & Holt, 2005; Emerson, 2000) and increased use of psychotropic medication (Tsakanikos, Costello, Holt, Sturmey & Bouras, 2007). In particular, the high prevalence of co-morbid intellectual disability in those with ASD (Matson & Shoemaker, 2009) means that many individuals require specialist care and support throughout their lifetime. In the UK, such specialist provision incurs significant costs to education and healthcare providers, with estimated annual costs of supporting children and adults with ASD at £2.7 billion and £25 billion, respectively (Knapp, Romeo & Beecham, 2009). Given the substantial impact on quality of life for those with ASD, as well as the costly financial implications, it is important to further our understanding of the factors that contribute most significantly to the development and consequences of co-morbid disorders in ASD, such as symptom severity, presence and/or severity of intellectual disability, age and gender.

In addition to co-morbid intellectual disability and psychiatric diagnoses, the presence of behaviours that challenge (i.e. aggression, destruction and self-injurious behaviour) in individuals with ASD have a significant impact on quality of life (Baghdadli, Pascal, Grisi & Aussilloux, 2003; Duerden, Oakley, Mak-Fan, McGrath, Taylor *et al*., 2012; McTierman, Leader, Healy & Mannion, 2011; Moyal, Lord & Walkup, 2014; Murphy *et al*., 2009; Rattaz, Michelon & Baghdadli, 2015; Richards, Oliver, Nelson & Moss, 2012). The term ‘behaviours

that challenge' typically refers to a behaviour that is of such an intensity, frequency or duration as to threaten the quality of life and/or the physical safety of the individual or those around them. In particular, research highlights that those with ASD display more behaviours that challenge than their typically developing peers (McClintock *et al.*, 2003), than those with a psychiatric diagnosis only (Matson, Wilkins & Macken, 2009) and than those with intellectual disability of heterogeneous aetiology (Davies & Oliver, 2013; Holden & Gitlesen, 2006; McClintock, Hall & Oliver, 2003; Oliver, Murphy & Corbett, 1987).

Notably, self-injurious behaviour is particularly common in individuals with ASD (Matson *et al.*, 2009). Self-injurious behaviour refers to a series of aggressive behaviours that an individual directs towards themselves that have the potential to result in physical injury, typically in the form of tissue damage (Matson & Turygin, 2012). Common forms of self-injurious behaviour include head banging, self-biting, skin scratching, hair pulling and hitting oneself against hard objects (Cooper, Smiley, Allan, Jackson, Mantry *et al*., 2009; Iwata, Pace, Dorsey, Zarcone, Vollmer, Smith *et al*., 1994; Minshawi, Hurtwitz, Fodstad, Biebl, Morris *et al*., 2014; Weiss, 2002). Such behaviours vary in intensity and in the most severe cases can lead to life threatening injuries and even death (Baghdadli *et al.*, 2003; Rojahn, Schroeder & Hoch, 2008; Schroeder, Mulick & Rojahn, 1980). The presence of self-injurious behaviour has been associated with higher rates of psychiatric hospitalisation (Mandell, 2008), more reactive physical interventions (e.g. use of protective equipment, emergency medication use, physical holds and seclusions; Allen, Lowe, Brophy& Moore, 2009), lower quality of life (Beadle-Brown, Murphy & DiTerlizzi, 2009), exclusion from mainstream services (Knapp, Comas-Herrera, Astin, Beecham & Pendries, 2005), and is the primary cause of emergency room visits among children with ASD (Kalb, Stuart, Freedman, Zablotsky & Vasa, 2012). Furthermore, the detrimental impact of self-injurious behaviour extends beyond the individual with

consequences for the whole family. For example, quality of life has been found to be lower in parents of children with ASD due to heightened rates of behavioural problems, most notably, self-injurious behaviour (Baghdadli., Pry, Michelon & Rattaz, 2014; Greenberg, Seltzer, Hong & Orsmond, 2006; LeCavalier, Leone & Wiltz, 2006;). Finally, a recent study demonstrated that self-injurious behaviour is highly persistent in individuals with ASD, with 77.8% of individuals continuing to show self-injury over three years (Richards *et al*., 2016). In summary, self-injury is common and persistent in ASD, and the effects of self-injurious behaviour are highly detrimental to both the individual and their families. Thus, self-injury remains a behaviour of significant clinical importance in ASD.

Despite the significant negative consequences of self-injurious behaviour, and the emerging evidence of persistence of the behaviour in ASD, prevalence estimates for self-injurious behaviour in ASD remain highly heterogeneous. Estimated prevalence rates vary from 33% to 71% (e.g. Baghdadli *et al*., 2003; Bartak & Rutter, 1976; Bodfish *et al*., 2000; Cooper *et al*., 2009; Dominick *et al.*, 2007; Gulsrud, Lin, Park, Hellemann & McCracken, 2018). This variation is likely due to a combination of differences in sample sizes and study methodologies, differences in the way that ASD has been assessed and different definitions of self-injurious behaviour (Fombonne, 2005). Moreover, participant characteristics such as age and presence and/or severity of co-morbid intellectual disability vary between studies making it difficult to estimate robustly the prevalence of self-injurious behaviour in ASD. It is important that these differences are taken into account when attempting to synthesise published prevalence data on self-injurious behaviour in ASD in order to determine the extent to which these factors influence prevalence rates.

Our understanding of the factors that influence the presence and severity of self-injury within ASD must be integrated with evidence based models of the function and psychological mechanisms underpinning self-injury. For individuals with an intellectual disability, many of whom have co-morbid ASD, the most influential and evidence-based explanatory model of self-injurious behaviour is derived from operant learning theory (Summers, Shahrami, Cali, D'Mello, Kako, Palikucin-Reljin *et al.*, 2017). Within this model, self-injury is understood to occur as a result of positive or negative reinforcement and can be mediated by contingencies in the individual's social, sensory and material environment. Evidence arising from applied behaviour analytic studies demonstrates that self-injury can be reduced by introducing adaptive behaviours that displace self-injury, therefore providing evidence for self-injury as a functional behaviour (Oliver & Richards, 2015). Consequently, interventions for behaviours that challenge are often predicated on the principles of operant learning theory, such as those recommended in the NICE guidance on behaviours that challenge (NICE, 2015).

However, there is preliminary emerging evidence that the presence of self-injury in those with ASD *without* an intellectual disability may be best understood through reference to different psychological models than those applied to self-injury in those with co-morbid intellectual disability. Self-injury in those with ASD *without* an intellectual disability may in fact be more similar to behaviours observed in those of typical development with mental health difficulties (Maddox, Trubanova & White, 2017). Often referred to as self-harm or non-suicidal self-injury (NSSI) in the general population, common topographies include cutting, carving and burning which are often classified as distinct from the more repetitive and rhythmic self-injurious behaviours typically seen in those with an intellectual disability, such as head banging and self-hitting. Similarly to distinction in the form and function observed in those with mental health difficulties, the purpose of self-harm/NSSI within the ASD population is believed to regulate

negative affect and reduce emotional distress in those who engage in the behaviour (Maddox *et al*., 2017).

In light of this distinction between self-injurious behaviour in those with ASD and self-injurious behaviour in those with ASD *and* co-morbid intellectual disability, one initial step in furthering our understanding of this purported difference in categorisation is to evaluate the topographies of self-injurious behaviours in those with ASD with and without intellectual disability; putative differences in underpinning psychological models may be associated with differences in form. For example, more rhythmic stereotypic topographies (e.g., head banging) may be associated with the presence of intellectual disability, whereas less rhythmic topographies (e.g., self-burning) may be associated with the *absence* of intellectual disability. More broadly, it is important to establish which features of ASD and which demographic variables may influence the presence and/or topography of self-injurious behaviour in order to inform more accurate causal models in the future.

In summary, self-injurious behaviour is common, persistent and associated with poor outcomes. However, current prevalence estimates are highly varied and the influence of person and study characteristics on prevalence and topography of self-injury in ASD is unknown. Thus, in order to identify existing service need for individuals with ASD, and in turn shape future interventions and service delivery for this population, it is necessary to determine robust prevalence estimates for self-injurious behaviour. Similarly, there is a need to empirically synthesise differences in study and/or participant characteristics and quantitatively evaluate the effects of these factors on the data reporting self-injurious behaviour in those with ASD. Therefore, the present meta-analysis seeks to describe and evaluate the current literature estimating the prevalence of self-injurious behaviour in ASD in order to:

1) Synthesise prevalence rates of self-injurious behaviour in ASD in order to generate robust estimates.
2) Evaluate how study characteristics (self-injurious behaviour definition, ASD definition) and participant characteristics (age, gender, presence/severity of ID, presence/severity of ASD) influence the reported prevalence rates.
3) Evaluate how participant characteristics influence the reported prevalence rates of individual topographies of self-injurious behaviour.

1.3. Methods

1.3.1 Search Strategy

In order to ensure the literature search was as inclusive as possible, a broad list of search terms was created by scanning relevant articles and adopting the search terms used by the previous systematic reviews (Edmondson, Brennn & House 2015; Richards, Jones, Groves, Moss & Oliver, 2015; Victor & Klonsky, 2014). Literature searches were then conducted in Ovid PsychINFO, Ovid Medline and Ovid Embase by combining all variations of the autism and/or intellectual disability and self-injury search terms. The autism search terms included: Autism spectrum disorder*/autis* spectrum disorder*, Intellectual disabilit*, Autis*, ASD, PDD-NOS, PDDNOS, Unspecified PDD, Pervasive developmental disorder*, Pervasive developmental disorder not otherwise specified, Asperger* and Asperger* syndrome. The self-injury search terms included: Self-injur*, Self-injurious behav*, Self-injurious behaviour, Self-harm*, Self harm*, Selfharm*, Deliberate harm*, NSSI. Non-suicidal self-injur*, Nonsuicidal self-injur*, Self-mutilat*, Mutilat*, Self-cut*, Self-mutilative behav*, Deliberate self-harm*, DSH, Self-inflicted wound*, Self-destructive behav*, Self-destruct*, Parasuicid* and Non-fatal deliberate self-harm.

The literature search was conducted on 28th May 2018 and included all English language papers published from 1967 up to the date of the literature search. In addition to the literature search, a hand search of the reference lists of the returned articles was conducted and any relevant identified papers were included alongside those from the literature searches.

1.3.2. Quality review

Each article was reviewed based on five quality criteria (see Table 1.1) which control for key threats to validity. The quality criteria were generated by reviewing standardised quality criteria for intervention (e.g. Downs & Black, 1998) and prevalence studies (Shamliyan, Kane, Ansari,

Raman, Berkman, Grant *et al.*, 2011; Richards *et al.*, 2015; Surtees, Oliver, Jones, Evans & Richards, 2018). For visual ease of interpretation, the criteria for each article were coded as red for a score of 0, yellow for a score of 1, amber for a score of 2 and green for a score of 3. This method provides a simple visual matrix that evidences the quality of each paper. The quality weighting for each paper was then calculated by dividing the total quality score by the maximum possible total of fifteen. All studies which met the inclusion criteria were reviewed by the author and rated for quality using these criteria. Inter-rater reliability was established by a second researcher independently reviewing a subset of studies (N=8) to ascertain the reliability of the criteria. Inter-rater reliability for total quality weighting was good (r =.923; p=.001).

Table 1.1. Quality Criteria for sample identification, ASD assessment, measurement of ID and measurement of SIB/self-harm.

	0 Poor	1 Adequate	2 Good	3 Excellent
Sample Identification	Not specified/reported	Single restricted or non-random sample e.g., a specialist clinic or previous research study Single regional sample e.g., one ASD school or adult service/group home	Multiple restricted or non-random samples e.g., multi-region specialist clinics National non-random sampling e.g., national parent support groups	Random or total population sample
Assessment of ASD	Not specified/reported Clinician judgement only	Screening instrument e.g., SCQ, M-CHAT Clinician judgement against specified diagnostic criteria e.g., DSM-IV or ICD-10	Diagnostic instrument e.g., ADI-R, DISCO, ADOS, 3Di	Consensus from multiple assessments, including at least one diagnostic instrument
Measurement of intellectual disability: IQ	Not specified/reported Clinician judgement only	Parent/carer report Recruited from a specialist ID school	Parent/carer report with a well validated measure	Formal IQ test (e.g. Weschler Intelligence Scale for Children)
Measurement of intellectual disability: adaptive functioning	Not specified/reported Clinician judgement only	Parent/carer report	Parent/carer report with a well validated measure	Formal measure of adaptive functioning (e.g. Vineland Adaptive Behaviour Scales)
Measurement of self-injurious behaviour/self-harm	Not specified/reported	Parent/carer report	Direct observation of behaviour by clinician/researcher OR use of a formal, validated scale (SIB -Q, SIQ, CBQ)	Consensus from multiple assessments including at least one direct observation or formal, validated scale (**SIB -Q, SIQ, CBQ** to assess for presence of SIB /self-harm in combination with direct observation of behaviour

1.3.3. Selection Strategy

A total of 3,350 papers were identified by searching the databases. These papers were assessed for suitability for inclusion using the follow stages:

1.3.3.1 Stage 1: Screening

The titles and abstracts of all papers were screened by the researcher. Table 1.2 outlines the inclusion and exclusion criteria used to assess for suitability at this stage. In cases where suitability was unclear a second researcher reviewed the paper and consensus was derived.

Table 1.2. Inclusion and exclusion criteria for screening**.**

Inclusion Criteria	Exclusion Criteria
Empirical papers	Conference proceedings, magazines, dissertations, review articles and books
Papers published are available in English	Papers published in a language other than English
Sample includes participants with idiopathic ASD	Sample included participants with ASD of known genetic cause, for example, fragile X Syndrome, tuberous sclerosis complex etc [2]
Abstract indicates that the paper reports on the prevalence and/or topography of self-injurious behaviour/self-harm within the ASD group	Topography and/or prevalence of self-injurious behaviour/self-harm was not reported within the ASD group
Cohort study	Case series and case studies

1.3.3.2 Stage 2: Eligibility

The full texts of the screened papers were then read to assess the eligibility of the data for inclusion in the meta-analysis. The same inclusion and exclusion criteria were used at the

[2] Samples that included ASD of known genetic cause such as fragile X Syndrome and tuberous sclerosis complex were excluded from this meta-analysis on the basis that it would not be possible to delineate self-injurious behaviour in ASD from self-injurious behaviour associated with the genetic syndrome.

screening and eligibility stages; however, the following additional criteria were applied to assess eligibility (see Table 1.3).

Table 1.3. Additional inclusion and exclusion criteria for eligibility assessment**.**

Inclusion Criteria	**Exclusion Criteria**
Participants were recruited without any specific bias that could have influenced self-injury e.g. presence of epilepsy	Participants were recruited because they showed additional special characteristics that may influence self-injury e.g. epilepsy
Study reports on a unique sample (or a potentially overlapping sample, but the proportion of overlap cannot be readily determined)	Study reports on exactly the same sample as reported in a previous study.

1.3.3.3. Stage 3: Quality Criteria

The quality of the remaining papers was then assessed according to the quality criteria outlined in Table 1.3. The flowchart presented below uses the PRISMA model (Moher, Tetzlaff & Altman, 2009) to outline the number of papers excluded at each stage of review.

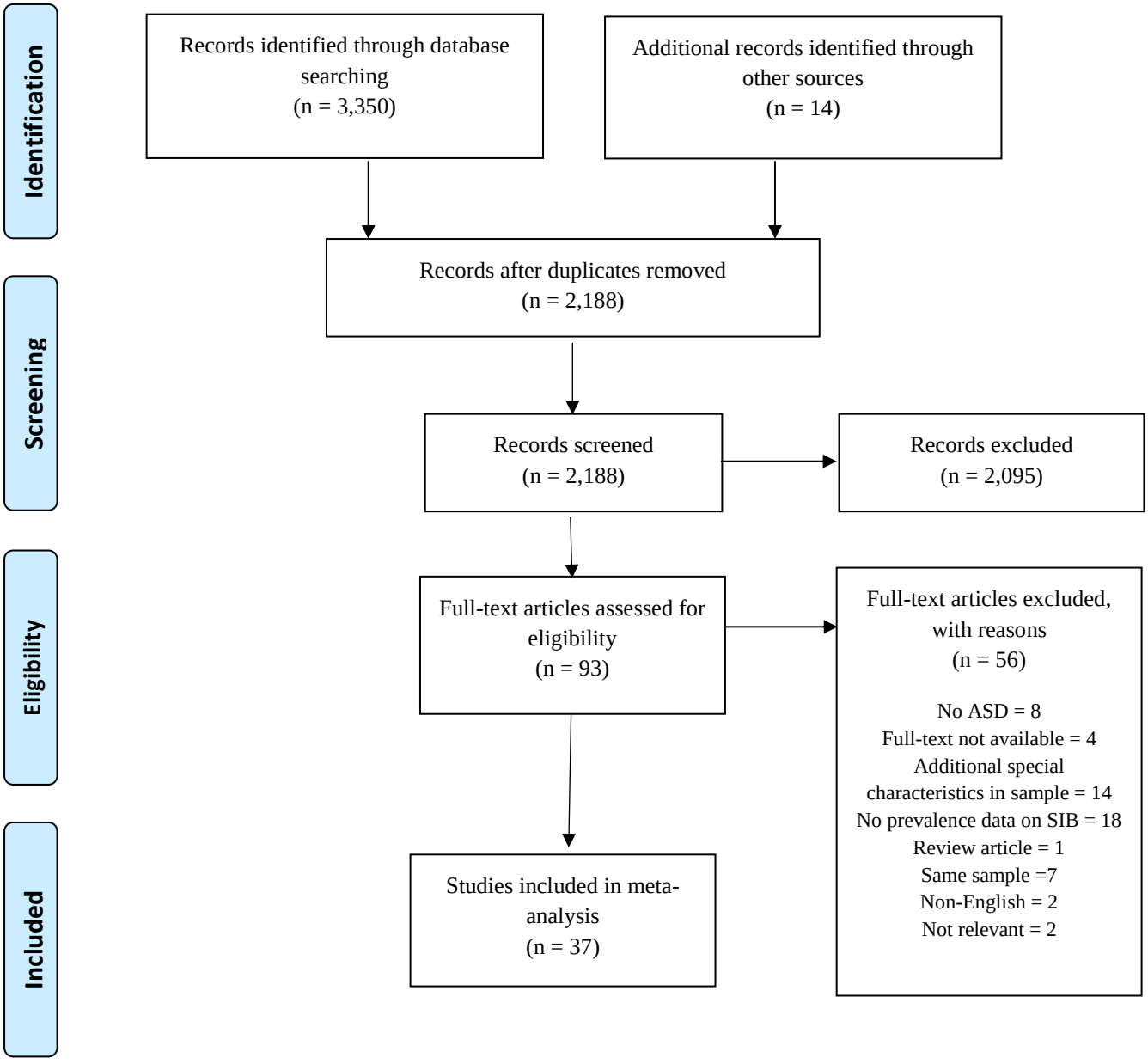

Figure 1.1 PRISMA flow chart showing the number of papers included and excluded at each stage of screening and review (Moher *et al*., 2009).

1.3.4 Data Analysis

In order to describe the prevalence of self-injurious behaviour in ASD, the number of participants in the sample that displayed self-injurious behaviour[4] was extracted from each paper. These data were analysed to generate pooled prevalence estimates. Fixed-effects models of pooled prevalence assume that that the true effect size for all studies under review are identical and that any variation in prevalence estimates are due to sampling error (Barendregt & Doi, 2011). Given the substantial heterogeneity in the extracted prevalence rates between studies, a random-effects model was determined to be more appropriate and was used to calculate the pooled prevalence estimate. Unlike the fixed-effects model which attributes variation in prevalence rates to sampling error, the random-effects model assumes two sources of variability that could account for the heterogeneity between studies; one from sampling error and one from differences in study level characteristics, which are controlled for in the weighting assigned to each study. However, the random-effects model does not control for variability that arises due to differences in the methodological quality of the studies. Therefore, further sub-group analyses were also conducted in order to assess the impact of these differences upon heterogeneity.

In order to explore the influence of individual participant characteristics such as age, gender and presence of intellectual disability on prevalence rates of self-injurious behaviour, a series of meta-regression analyses were conducted. Due to insufficient data,

[4] Where a study reports more than one prevalence figure for self-injurious behaviour, a decision on which figure to include in the meta-analysis was made on an individual basis. Reasons for the decision are included as a footnote for each study in Table 1.4.

it was not possible to meta-analyse the influence of ASD severity on prevalence rates, however, those studies that did report these data are described.

Finally, in order to describe the prevalence rates of different topographies of self-injurious behaviour, the number of participants engaging in each topography was extracted from the papers that reported on topography. These data were analysed using the random-effects model to generate pooled prevalence estimates for each topography of behaviour.

1.4 Results

1.4.1 Prevalence of Self-Injury in ASD

A total of 37 primary studies reporting a total of 14,379 participants were identified as suitable for inclusion in the current meta-analysis. In order to assess the first aim of the meta-analysis, each study was evaluated against the quality criteria and data describing the study, sample characteristics (e.g. age, gender, ASD characteristics, and presence of an intellectual disability) and total prevalence of self-injurious behaviour/self-harm were extracted and analysed to generate pooled prevalence estimates (see Table 1.4.).

Across all studies, only five (13.5%) papers met criteria for the highest quality rating for sample identification, seven (18.9%) for ASD assessment, 11 (29.7%) for assessment of IQ, 13 (35.1%) for assessment of adaptive functioning and three (8.1%) for measurement of self-injurious behaviour. Whilst the majority of papers (N=25; 67.5%) reported the proportion of the sample that had an intellectual disability, assessment of intellectual disability received the highest number of poor ratings with 14 (37%) studies for assessment of IQ and 18 studies (48.6%) for assessment of adaptive functioning being rated as poor. In addition to reporting the prevalence of self-injurious behaviour, 16 studies (43.2%) reported prevalence of different topographies of behaviour. No studies were excluded on the basis of quality as doing so would result in the loss of valuable data obtained from large samples of participants. Instead, analyses of the effect of methodological quality was conducted to determine the possible influence of poorer quality studies on prevalence estimates.

Sample sizes varied considerably between studies with one notable study by Soke, Rosenberg, Hamman, Fingerlin Robinson, Carpeter *et al.*, (2016) reporting data on a very

large sample of participants with ASD (N=8065). Additionally, this study obtained a rating of excellent for sample identification, suggesting that the prevalence data reported in this large scale study were obtained in a representative sample. However, this study received ratings of poor for assessment of intellectual disability (IQ and adaptive functioning) and measurement of self-injurious behaviour, which may represent a justifiable trade-off between sample size and measurement precision. Just over 50% of studies received a good (N=19; 51.3%) rating for sample identification suggesting that the prevalence data reported in these studies were obtained from representative samples.

Table 1.4. Quality criteria, study and sample characteristics and outcome data for studies reporting the prevalence/topography of SIB/SH in the ASD population.

	Quality Criteria					ASD Study and Sample Characteristics							Outcome Data	
Author	SAMPLE	ASD	ID:IQ	ID: AF	SIB/SH	N	% Male	Mean Age (SD) *Range*	ASD measure	% ASD categories	% with ID	Prevalence of SIB across categories/measures	**Prevalence of SIB/SH**	**Quality Weighting**
Akram *et al.*, 2017						83	72.2	11.77 (3.59) *8.0-18.0*	CARS[5]	ASD=100.0	Not reported	-	33.0	0.2
Ando & Yoshimura (1979a; 1979b) [6]						47	83.0	*6.0-14.0*	Not reported	Not reported	95.7[7]	-	42.6	0.4
Baghdadli *et al.*, (2003)						222	4.7:1 (male: female)	5.0 (1.2) *2.0-7.0*	ICD-10, ADI-R[8] & CARS[9]	Not reported	96.2[10]	-	53.0[11]	0.73
Ballaban-Gil *et al.*, (1996)[12]						Adolescents 54 Adults 45	3:1 (male: female)	6.8 *9.5m-20.3y*	DSM-IV	Not reported	Adolescents 36.0[13] Adults 64.0[14]	-	Adolescents 26.0[15] Adults 38.0[16]	0.13

[5] Childhood Autism Rating Scale
[6] The same sample was used for both studies therefore only reported in the table once
[7] IQ score distribution as measured by the WISC: 92% <50, 4% 51-67, 2% 68-84 and 2% >85.
[8] Autism Diagnostic Interview-Revised
[9] The CARS was used to measure ASD severity
[10] 20.3% had a mild ID, 70% had a severe ID and 5.9% had a profound ID
[11] 21.5% displayed mild SIB, 17.1% moderate SIB and 14.6% severe SIB.
[12] Baseline data reported
[13] Percentage within each level of ID: Normal/near normal 31%; Mild-Moderate ID 17%; Severe ID 19%; Indeterminate 33%
[14] Percentage within each level of ID: Normal/near normal 12%; Mild-Moderate ID 14%; Severe ID 50%; Indeterminate 24%
[15] SIB displayed by 9% of participants within the Normal/Near normal group, 45% of the Mild-Moderate ID group and 30% of the severe ID group. Participants in the indeterminate group were including in the mild-moderate group for analysis of SIB.
[16] SIB displayed by 31% of participants within the Normal/Near normal group, 9% of the Mild-Moderate ID group and 32% of the severe ID group. Participants in the 'indeterminate' group were including in the mild-moderate group for analysis of SIB.

Quality Criteria						ASD Study and Sample Characteristics							Outcome Data	
Author	SAMPLE	ASD	ID:IQ	ID: AF	SIB/SH	N	% Male	Mean Age (SD) *Range*	ASD measure	% ASD categories	% with ID	Prevalence of SIB across categories/measures	**Prevalence of SIB/SH**	**Quality Weighting**
Bartak & Rutter (1976)[17]						19 (ASD) 17 (ASD+ID)	100.0	No mean/SD <11	Not reported	Not reported	0.0 100.0	-	31.6 70.6	0.53
Billstedt *et al.*, (2005)[18]						10	70.0	Not reported	DSM-III-R; clinical interview, HBSS[19], CARS; ABC[20].	ASD = 65.0 Autistic-like/ atypical autism = 35.0	81.7	-	50.0	0.73
Bodfish *et al.*, (2000)						32	75.0	33.1 No SD or range	DSM-IV, CARS, ABC	Not reported	94.0[21]	-	69.0	0.33
Bradley *et al.*, (2004)						12	66.7	16.3 (2.2) No range	ADI-R	100.0 = Autism	100.0[22]	-	58.0	0.8
Buono *et al.*, (2010)						49	60.0	13.1 (8.5) No range	Not reported	Not reported	100.0[23]	-	70.0	0.13
Cassidy *et al.*, (2018)						164	40.0	Not reported for total sample[24]	Diagnosis self-reported	HFA/AS[25]=83.0 Classic autism=2.0 ASC=9.0 PDD-NOS=1.2	1.5	-	78.0	0.4

[17] Two samples included in this study, one with IQ > 70, and one with IQ< 70. Results from both samples are reported here.
[18] ID and diagnostic classifications based upon total sample at T1 (N=120), but SIB prevalence based upon sample at T2 (N=108)
[19] Handicaps, Behaviour and Skills Schedule
[20] Autistic Behaviour Checklist
[21] 44% of participants classed as having severe ID and 50% as having profound ID
[22] All participants had severe ID
[23] 18% of participants had mild ID, 14% moderate ID, 44% severe ID and 24% profound ID
[24] Males: 41.52 (11.73); females: 38.89 (10.47)
[25] High functioning autism/Asperger's Syndrome

Quality Criteria						ASD Study and Sample Characteristics							Outcome Data	
Author	SAMPLE	ASD	ID:IQ	ID: AF	SIB/SH	N	% Male	Mean Age (SD) *Range*	ASD measure	% ASD categories	% with ID	Prevalence of SIB across categories/measures	**Prevalence of SIB/SH**	**Quality Weighting**
Cooper *et al.*, (2009)						77	Not reported	Not reported	C21st Healthcheck; Assessed by Consultant Psychiatrist	Not reported	Not reported	-	13.0	0.66
Dominick *et al.*, (2007)[26]						54	87.0	91.2 (29.8) No range	DSM-IV, ADI-R & ADOS-G[27]	Not reported	Mean IQ 81.0	-	32.7	0.6
Duerden *et al.*, (2012)[28]						241[29]	84.8[30]	88.33 No SD *21.74-231.51*	ADOS-G & ADI-R	ASD = 92.8 AS/PDD-NOS= 7.2	27.9[31]	52.3 (n =241; ADI-R) 64.9 (n=171; RBS-R)	52.3[32]	0.73
Folch *et al.*, (2018)						158[33]	Not reported	Not reported	CARS	All had ASD symptoms	100.0	-	37.3[34]	0.53
Gal *et al.*, (2009)						56	75.0	9.71 (1.86) No range	CARS/DSM-IV	Not reported	50.0	-	64.3	0.4
Gulsrud *et al.*, (2018)						144	81.0	9.3 (8.6) *2.5 – 60.1*	ADI-R & ADOS-2	Not reported	Mean Full Scale IQ 91.3 (23.7)	Current=29.1 Past=13.8	29.1[35]	0.73

[26] Age data reported in months
[27] Autism Diagnostic Observation Schedule—Generic
[28] Age data reported in months
[29] Total sample N= 250, however, SIB data collected for N=241.
[30] Proportion of male participants for total sample (N=250)
[31] Ability was measured in 231 participants with ID defined as IQ <80
[32] The prevalence figure obtained from data collected from the total sample was used in the meta-analysis
[33] Total sample consisted of 833 participants but only ASD group (N=158) is included in this meta-analysis
[34] Total prevalence of self-injury reported as 37.3%; mild-moderate ASD symptoms is 33.8%; severe ASD symptoms is 39.8%
[35] Prevalence of current SIB was selected as the prevalence figure to report as the majority of studies report on current SIB

Quality Criteria						ASD Study and Sample Characteristics							Outcome Data	
Author	SAMPLE	ASD	ID:IQ	ID: AF	SIB/SH	N	% Male	Mean Age (SD) *Range*	ASD measure	% ASD categories	% with ID	Prevalence of SIB across categories/measures	**Prevalence of SIB/SH**	**Quality Weighting**
Handen *et al.*, (2018)						302	79.0	12.9 (3.4) *4.0 – 20.0*	ADOS-2	Not reported	Mean Non-Verbal IQ 75.15 (28.74)[36]	Home SIB 49.2% Home/Hospital 24.8%	74.2[37]	0.8
Hattier *et al.*, (2011)						633	Not reported for ASD group	Not reported for ASD group	Not reported	ASD = 55.0 PDD-NOS = 45.0	Not reported	22.9 (one endorsement) 6.6 (endorsed all items)	22.9[38]	0.2
Janicki *et al.*, (1983)						314	72.0	≥ 21	Not reported	Autism	85.0[39]	-	20.0	0.13
Kamio (2002)						165	77.5	15.5 (No SD) *12.0-18.0*	Not reported	Not reported	95.8[40]	Severe SIB =1.8 Mild SIB =23.0[41]	23.0[42]	0.33
Kats *et al.*, (2013)						438	73.0[43]	42.0 (8) *30.0 -59.0*	Not reported	Not reported	100.0[4445]	-	39.0	0.2

[36] Data only reported for 252 subjects
[37] Total prevalence of self-injury reported as 74.2%; 49.2% displayed self-injury at home only and 24.8% displayed self-injury at home and in hospital
[38] In order to be an inclusive as possible, the prevalence figure used in the meta-analysis relates to those who endorsed one form of SIB
[39] Categorised as: 10% Mild Mental Retardation, 17% Moderate mental retardation, 30% Severe Mental Retardation and 43% Profound Mental Retardation.
[40] Categorised as: 7.9% Mild ID, 18.8% Moderate ID, 32.1% Severe ID and 37% Profound ID.
[41] When the criteria for SIB was broadened, prevalence rates increased from 1.8% to 23%.
[42] In order to be representative of the majority of SIB cases, the prevalence figure relating to mild SIB was used in the meta-analysis
[43] Missing data for two subjects
[44] Categorised as: 27% Mild ID, 37% Moderate ID, 37% Severe ID. Those with Profound ID were excluded due to complexity in diagnosing ASD within this group.
[45] Data on ID was missing for 38 subjects.

Quality Criteria						ASD Study and Sample Characteristics							Outcome Data	
Author	SAMPLE	ASD	ID:IQ	ID: AF	SIB/SH	N	% Male	Mean Age (SD) *Range*	ASD measure	% ASD categories	% with ID	Prevalence of SIB across categories/measures	**Prevalence of SIB/SH**	**Quality Weighting**
Lecavalier (2006)						487[46]	82.6	Not reported	Not reported	PDD= 33.0 ASD=67.0	66.0	-	Not reported[47]	0.46
Maddox *et al.*, (2017)						42	57.0	Not reported for total sample[48]	Diagnosis confirmed by previous evaluation and written report	ASD 100.0	0.0	-	50.0	0.33
Matson *et al.*, (1996)						185	64.0	37.94 (12.84) No range	DASH-II	PDD Autism	100.0[49]	-	34.0	0.4
McTiernan *et al.*, (2011)						174	82.0	8.0 (2.38) *3.0 - 14.0*	DSM-IV-TR	Not reported	87.0[50,51]	-	48.9	0.2
Murphy *et al.*, (2009)						157	82.8	8.5 (2.17) *3.0 – 14.2*	Not reported	Not reported	79.8[52,53]	-	45.2	0.13
Poustka & Lisch (1993)						61	82.6	15.3[54] *5.00-33.00*	ADI score ADOS	Autism	78.3	-	52.5	0.46

[46] Data missing for 16 subjects; Total N=487; Teacher ratings N=437; Parent ratings N=353
[47] It was not possible to calculate prevalence of SIB, however, the paper does report prevalence of different topographies of SIB, therefore, has been included
[48] ASD/NSSI group: 25.29 (7.77) range 18-47; ASD only group: 29.19 (10.87) range 18-56.
[49] Categorised as: 33% Severe Mental Retardation and 67% Profound Mental Retardation.
[50] ID data only reported for 63.2% (N=110) of sample
[51] Males: 39.6% mild ID; 25.3% moderate ID, 18.7% severe ID; 2% profound ID; Females: 47.4% mild ID, 15.8% moderate ID; 10.5% severe ID; 0% profound ID.
[52] ID data only reported for 69.4% (N=109) of sample
[53] Males: 24.6% mild ID; 18.5% moderate ID; 13.1% severe ID; 0.8% profound ID; Females: 33.3% mild ID; 7.4% moderate ID; 7.4% severe ID
[54] Median age

Quality Criteria						ASD Study and Sample Characteristics							Outcome Data	
Author	SAMPLE	ASD	ID:IQ	ID: AF	SIB/SH	N	% Male	Mean Age (SD) *Range*	ASD measure	% ASD categories	% with ID	Prevalence of SIB across categories/measures	**Prevalence of SIB/SH**	**Quality Weighting**
Rattaz *et al.*, (2015)						152	82.2	15.0 (1.6) No range	ADI-R & CARS	Childhood autism 79.6 Atypical autism 20.4	Not reported	-	35.8[55]	0.86
Richards *et al.*, (2012)						149	88.6	9.98 (4.86) *4.0–39.0*	Professional diagnosis; SCQ score	Not reported	Not reported	-	50.0	0.6
Richards *et al.*, (2017)						Child 208 Adult 216	78.5	24.10 (13.01) *6.0-61.0*	Clinician report	ASD	Not reported	-	Child 45.7 Adult 49.1	0.26
Richler *et al.*, (2007)						165	84.4	29.41 *(4.86)* No range	ADI-R score; ADOS	Autism 71.0 PDD-NOS 29.0	Mean IQ = 49.44	-	29.7	0.73
Seltzer *et al.*, (2010)						86	79.1	24.7 *(7.24)* *18.0-53.0*	Professional diagnosis; ADI-R score	AD AS[56] PDD-NOS	57.0	-	24.0	0.73
Siegel *et al.*, (2015)						147	73.4	12..6 (3.42) *4.0 – 20.0*	SCQ, ADOS-2, DSM-5	Not reported	42.6	-	26.5	0.93
Slingsby *et al.*, (2017)						41	81	6 (No SD) *2.0 – 18.0*	Parent report	Autism	Mean IQ = 49.44	-	51.0	0.13
Soke *et al.*, (2016)						8065	82.24	8.0 No SD/Range	DSM-IV-TR	Not reported	29.75 (IQ <70)	-	27.7[57]	0.26

[55] 16.6% low self-injury; 19.2% high self-injury
[56] Asperger's Syndrome
[57] Data collected over three time points and averaged to get prevalence of 27.7%

Quality Criteria						ASD Study and Sample Characteristics							Outcome Data	
Author	SAMPLE	ASD	ID:IQ	ID: AF	SIB/SH	N	% Male	Mean Age (SD) *Range*	ASD measure	% ASD categories	% with ID	Prevalence of SIB across categories/measures	**Prevalence of SIB/SH**	**Quality Weighting**
Soke *et al.*, (2018)						691	81.91	55.7.0[58] (6.83) No range	ADOS, ADI-R, SCQ, VABS-2, DSM-IV-TR	Not reported	Mean IQ 66.87 (20.0)	-	29.4	0.8
Woodman *et al.*, (2015)						406	75.0	21.72 (9.45) *10-49.0*	ADI-R	94.6 = ASD 5.4 = AS/PDD-NOS	69.0	43.0 (T1) 34.0 (T5)	43.0	0.6

[58] Age reported in months

In order to achieve the first aim of the meta-analysis, prevalence rates of self-injurious behaviour in ASD were synthesised and a random-effects pooled prevalence estimate was generated based on the 37 studies that met the inclusion criteria (see Figure 1.2).

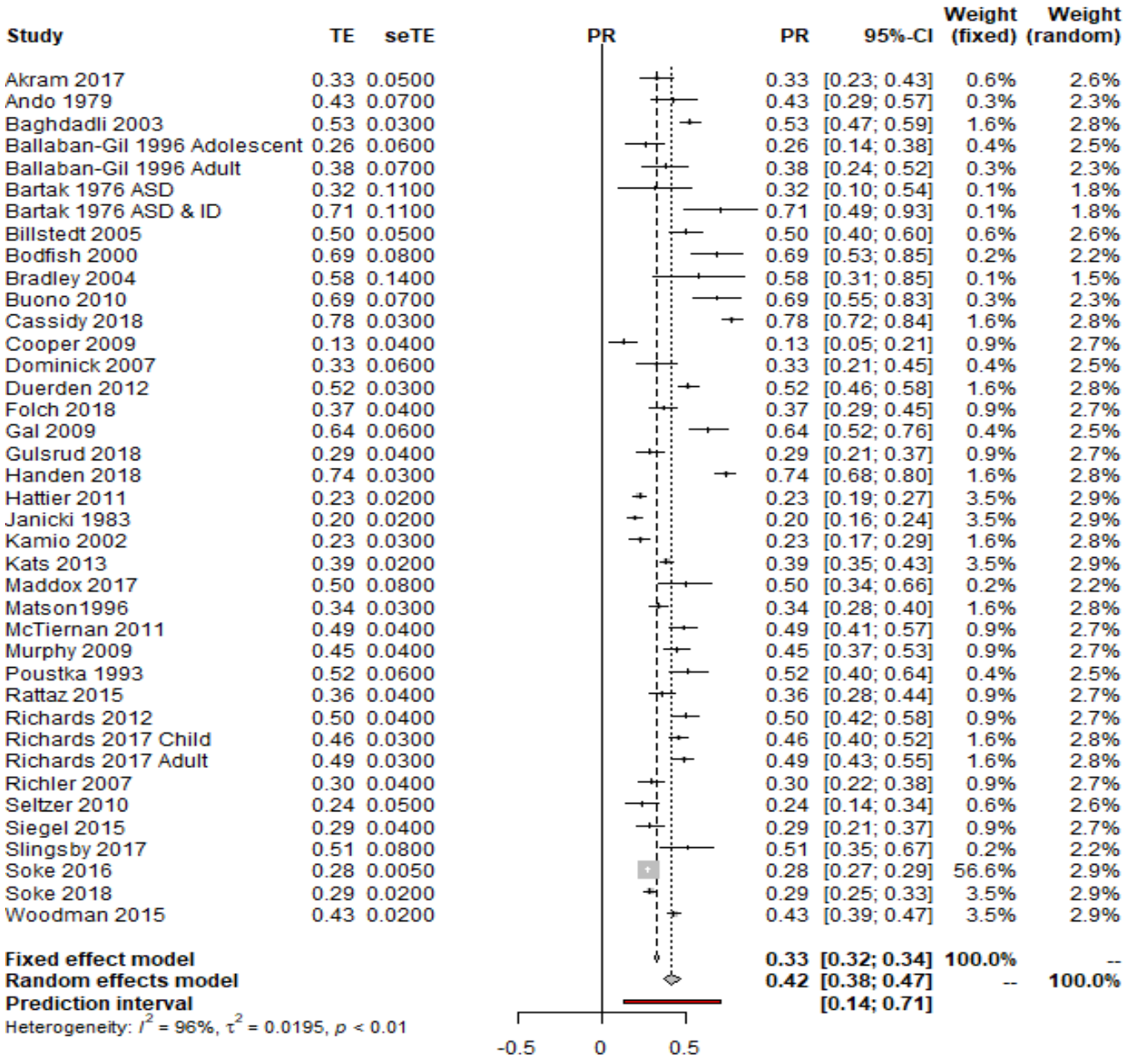

Figure 1.2. Pooled prevalence estimates for self-injurious behaviour in ASD using a random-effects model. Treatment effect (TE), standard error of the treatment effect (seTE), prevalence rate, confidence intervals and weighting by the random-effects model are reported.

The random-effects model was calculated using the generic inverse variance method and generated a weighted prevalence estimate of 42% (z = 17.71, p = < 0.001; CI 38-47 %) for self-injurious behaviour in ASD.

An unacceptable level of heterogeneity between the prevalence rates reported in the primary studies was observed ($tau^2 = 0.019$, Higgin's $I^2 = 96\%$; Q = 958.84, p < 0.001), which suggests that the estimates of the primary studies are biased by the presence of uncontrolled and potentially confounding factors.

1.4.2 Heterogeneity due to study level characteristics

In order to assess the impact of study level characteristics upon heterogeneity, a series of subgroup analyses were conducted on the prevalence rates of self-injurious behaviour based on the quality ratings of poor, adequate, good and excellent for each of the five types of methodological bias.

Table 1.5. Sub-group analysis of the prevalence rates (%) of SIB based on the quality ratings for each type of methodological bias. A mixed effects ANOVA was computed to test the difference between the proportions (Q) with associated probability levels (p).

	Poor	Adequate	Good	Excellent	Q	p
Sample Identification	45.0	40.0	45.0	32.0	6.24	0.10
Assessment of ASD	46.0	39.0	47.0	36.0	2.32	0.50
Measurement of ID: IQ	40.0	50.0	43.0	39.0	0.90	0.82
Measurement of IQ: Adaptive	41.0	57.0	31.0	43.0	1.74	0.62
Measurement of SIB/SH	31.0	39.0	43.0	57.0	6.48	0.09

The results of the sub-group analysis indicated that none of the indices of risk of bias evidenced significant differences in the estimated prevalence of self-injurious behaviour. However, despite being non-significant, overall trends in the data can be seen. For sample identification and assessment of ASD, prevalence estimates decrease as quality of these

study characteristics increase. Additionally, prevalence estimates are seen to increase when more robust methods are used to assess self-injurious behaviour.

1.4.3 Impact of influential studies

Baujat, Mahé, Pignon & Hill (2002) have proposed a method to explore heterogeneity that considers the impact of influential studies on the outcome of interest, in this instance, prevalence rates. On the x-axis the contribution of each study to the overall heterogeneity statistic is plotted. On the y-axis the standardised difference of the overall treatment effect with and without each study is plotted; this quantity describes the influence of each study on the overall effect (see Figure 1.3).

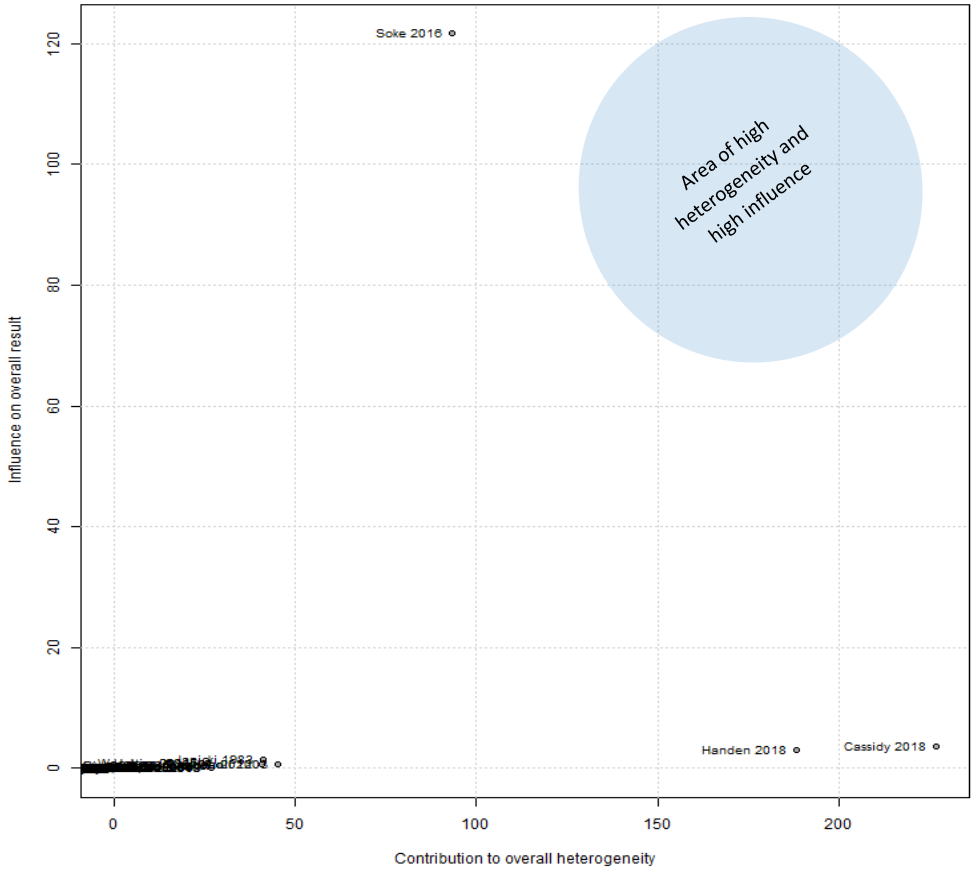

Figure 1.3. Baujat chart of sources of heterogeneity**.**

To examine whether any particular study or studies exerted a disproportionately high influence on the overall meta-analytic effect, a "leave-one-out" analysis, in which the

random effects model was calculated with each of the primary studies removed in turn, was conducted (see Figure 1.4). If omitting a study results in an effect that lies outside of the 95% CI for the complete meta-analysis then that study is deemed to have a disproportionate influence and is removed from the omnibus test. The study by Soke *et al*., (2016) strongly influenced the overall result but was not markedly heterogeneous to the other studies. The reason why it exerted influence over the overall result was due to sample size (N=8065) and therefore this study was not considered to be problematic.

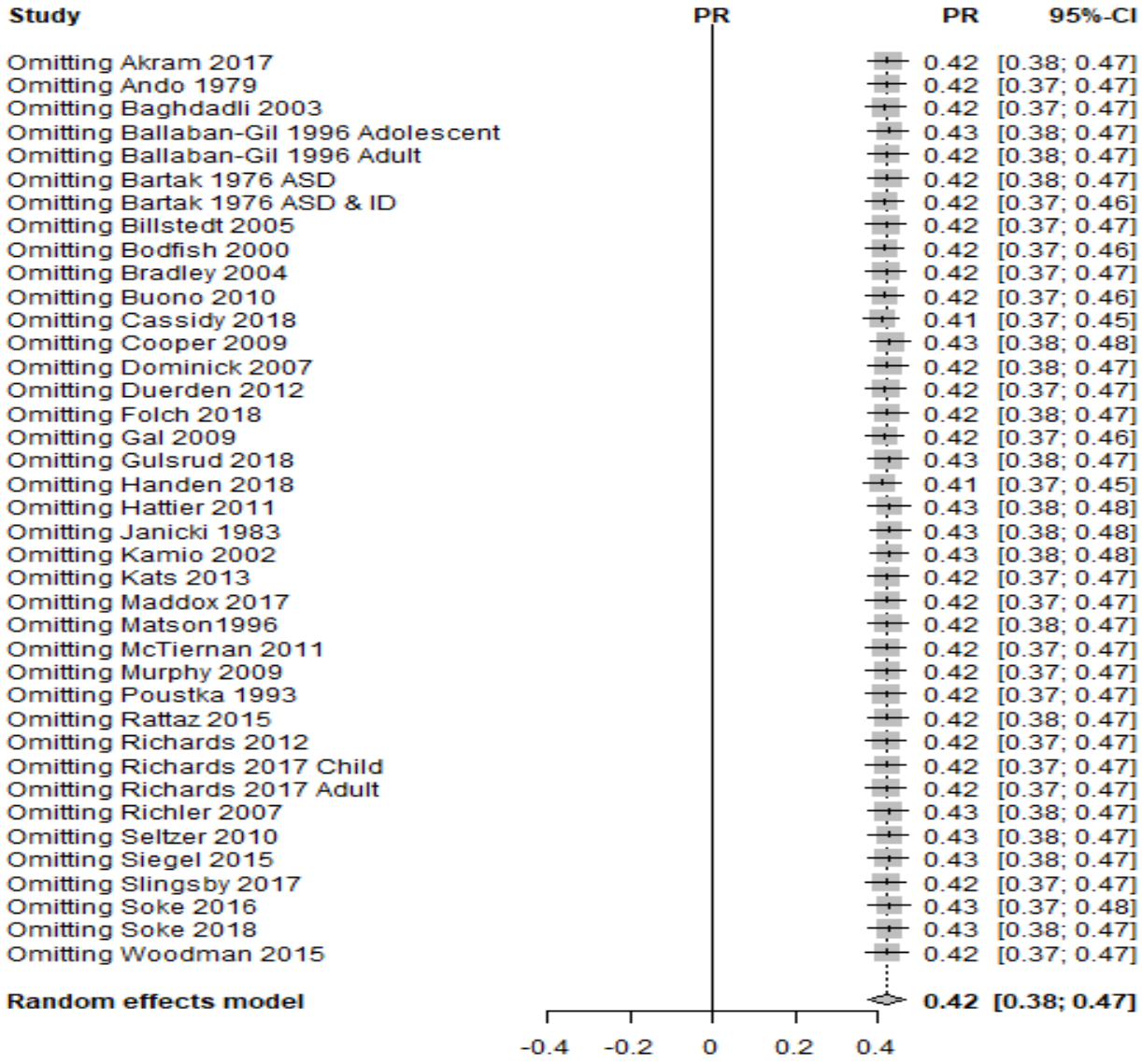

Figure 1.4. Pooled prevalence estimate for self-injurious behaviour in ASD using a random-effects model with each study omitted. Prevalence rates and confidence intervals are reported.

The "leave-one-out" analysis did not alter the prevalence estimate, including the very large sample in the Soke *et al*., (2016) paper, and therefore no studies were considered disproportionally influential on the overall meta-analytic effect.

1.4.4 Publication bias

Due to the substantial heterogeneity between the primary studies it was not possible to estimate and control for publication bias (see Appendix A for a funnel plot of the included studies).

1.4.5 Influence of participant characteristics on prevalence rates

In order to explore the second aim of the meta-analysis, the influence of age, gender and presence of an intellectual disability on prevalence rates was assessed using meta-regression analysis (see Table 1.6). Mean age of participants was extracted from 25 (67.5%) studies, percentage of male participants from 29 (78.3%) studies and percentage of participants with an intellectual disability from 27 (72.9 %) studies.

Table 1.6. Participant characteristic influencing prevalence of SIB in ASD.

Covariate	Estimate	S.E.	Z	p	Lower 95%CI	Upper 95%CI
Age (mean age)	-0.0028	0.0024	-1.1741	.240	-0.0074	0.0019
Gender (% male)	-0.0063	0.0025	-2.4693	.013	-0.0113	-0.0013
Presence of ID	-0.0001	0.0010	-0.0484	.961	-0.0021	0.0020

The meta-regression analysis in Table 1.6 revealed that the association between mean age of participants and prevalence rates of self-injury, and the proportion of participants with an intellectual disability and prevalence rates of self-injury were non-significant (p = .240) and (p = .961), respectively. However, the meta-regression analysis revealed a significant association between the proportion of male participants and prevalence rates (p = .013) suggesting that gender may influence the prevalence of self-injurious behaviour

in ASD. As the percentage of males included in the sample increased, the prevalence of self-injury significantly decreased. Specifically, for every unit increase in the percentage of males in the sample, prevalence of SIB decreased by 0.0063%.

Due to insufficient data it was not possible to meta-analyse the effect of severity of ASD on prevalence of self-injurious behaviour. Only two studies reported prevalence of self-injury across different levels of ASD. In their study, Akram *et al*., (2017) found that 10.3% of participants with mild ASD showed moderate self-injury whilst 6.5% showed severe self-injury. Thirty three percent of those with moderate ASD displayed moderate self-injury and 25.8% showed severe self-injury. Finally, 56.4% of participants with severe ASD showed moderate self-injury and 67% showed severe self-injury. Similarly, Folch *et al*., (2018) reported a higher prevalence of self-injury in those with severe ASD compared to mild-moderate ASD (39.8% and 33.8%, respectively). These results suggest that severity of ASD is associated with higher prevalence of self-injury, and moreover, that severity of self-injurious behaviour is associated with more severe ASD.

It was not possible to meta-analyse the prevalence of self-injurious behaviour across levels of intellectual disability due to insufficient data, however, one study reported prevalence data in this way. Ballaban-Gil *et al*., (1996) reported the prevalence of self-injurious behaviour within those with normal/near normal cognitive ability, mild-moderate intellectual disability and severe intellectual disability in an adult and adolescent sample. Within the adult sample, self-injury was displayed by 31% of participants with normal/near normal cognitive ability, 9% with mild-moderate intellectual disability and 32% with severe intellectual disability. Within the adolescent group, self-injury was displayed by 9% of participants with normal/near normal cognitive

ability, 45% with mild-moderate intellectual disability and 30% with severe intellectual disability. Within both samples, those with severe intellectual disability showed similar prevalence rates of self-injury (32% and 30%, respectively). However, rates varied considerably between those with a mild-moderate intellectual disability. Forty-five percent of participants in the adolescent sample displayed self-injurious behaviour compared to 9% in the adult sample.

1.4.6 Prevalence of different topographies of self-injurious behaviour

A total of 14 studies reported on the prevalence of different topographies of self-injurious behaviour (N=21). In order to address the final aim of the meta-analysis, these prevalence data were extracted and analysed to generate pooled prevalence estimates. Each topography of behaviour was included in the meta-analysis if a minimum of two studies reported data for the behaviour (see Table 1.7)

Table 1.7. Quality criteria, total prevalence of self-injurious behaviour in the total sample and prevalence of topographies of SIB/SH in the ASD population.

	Quality Criteria					**Prevalence of each topography of self-injurious behaviour/self-harm (%)**																				
Author	Sample ASD	ID:IQ	ID:AF	SIB/SH	Total Prevalence of SIB (%)	Hand hitting	Object hitting	Skin picking	Self-scratching	Self-pinching	Object/finger in cavities	Self-biting	Hair pulling	Nail pulling/eating	Teeth grinding	Head banging	Hitting self against object	Self-cutting	Keeping sores open	Throwing self to floor	Induced vomiting	Burning	Ingesting dangerous substances	Rubbing skin against surfaces	Carving	Unspecified
Akram *et al.*, 2017					33.0	15.6	-	-	12.0	10.8	-	10.8	9.6	-	-	-	-	1.2	10.8	-	-		1.2	6.0	-	-
Buono *et al.*, (2010)					70.0	41.0	41.0	-	14.0	6.0	35.0	39.0	6.0	12.0	8.0	-	-	-	-	-	-	-	-	-	-	-
Dominick *et al.*, (2007)					32.7	16.6	-	-	-	-	-	9.2	-	-	-	22.2	-	-	-	-	-	-	-	-	-	-
Duerden *et al.*, (2012)[59]					52.3	34.0	18.0	25.0	23.0	-	18.0	26.0	19.0	-	-	-	30.0	-	-	-	-	-	-	-	-	-
Folch *et al.*, (2018)					37.3	31.6	-	-	14.6	-	1.3	-	6.3	-	-	10.1	-	0.6	8.2	-	-	-	-	-	-	0.6
Gulsrud *et al.*, (2018)					29.1	9 .0[60]	-	2.0	0.7	-	-	3.5	0.7	-	-	10.4	-	-	-	1.4	0.7	-	-	-	-	-
Handen *et al.*, (2018)					74.2	48.3	22.1	33.1	43.3	-	10.5	30.1	24.5	-	-	-	46.7	-	-	-	-	-	-	-	-	-

[59] Total prevalence of SIB based on N=241; prevalence of each topography based on N=171

[60] Reported here as overall prevalence, however, authors separate it by hand hitting head (14.3%), hand hitting face (11.2%) and hand hitting body (5%).

Author	Quality Criteria					Prevalence of each topography of self-injurious behaviour/self-harm (%)																				
	Sample	ASD	ID:IQ	ID:AF	SIB/SH	Total Prevalence of SIB (%)	Hand hitting	Object hitting	Skin picking	Self-scratching	Self-pinching	Object/finger in cavities	Self-biting	Hair pulling	Nail pulling/eating	Teeth grinding	Head banging	Hitting self against object	Self-cutting	Keeping sores open	Throwing self to floor	Induced vomiting	Burning	Ingesting dangerous substances	Rubbing skin against surfaces	Carving	Unspecified
Hattier *et al.*, (2011)						22.9	5.8	-	-	5.8	5.8	2.4	-	-	-	-	-	8.5	-	-	-	-	-	-	-	-	-
Kamio (2002)						23.0	-	-	-	3.6	-	-	1.8	2.4	-	-	-	-	-	-	-	-	-	-	-	-	-
Lecavalier (2006)						Not reported	15.8	-	-	10.3	-	11.5	4.8	-	-	-	-	-	-	-	-	-	-	-	-	-	-
Maddox et al., (2017)						50.0	11.9	14.2	-	35.7	-	-	-	11.9	-	-	-	-	16.6	11.9	-	-	2.3	-	9.5	4.7	-
Richards *et al.*, (2012)						50.0	14.7	5.3	-	8.7	-	3.3	9.3	6.0	-	-	-	8.0	-	-	-	-	-	-	-	-	-
Richards *et al.*,(2017)						45.7 (child) 49.1 (adult)	24.5 child 28.2 adult	6.3 child 2.8 adult	-	11.1 Child 15.3 Adult	-	6.3 Child 5.1 Adult	17.3 Child 15.7 Adult	8.7 Child 10.2 Adult	-	-	-	15.9 Child 16.2 Adult	3.8 Child 3.2 Adult	-	-	-	-	-	-	-	-
Slingsby *et al.*, (2017)						51.0	22.0	17.0	-	-	-	-	17.0	2.0	-	-	-	7.0	-	-	-	-	-	-	-	-	10.0

A total of 13 topographies of self-injurious behaviour were analysed. Topographies that were only reported by one study were excluded from the meta-analysis (N=8). For brevity, the forest plots, prevalence rates, confidence intervals and weighting assigned by the random effects model for each topography of behaviour can be found in Appendix B. For studies that calculated prevalence of topography as a percentage of the number that showed self-injury, this was converted to a percentage of the total sample in order to generate pooled prevalence estimates. Figure 1.5 presents the pooled random effects prevalence with associated confidence intervals for each of the 13 topographies.

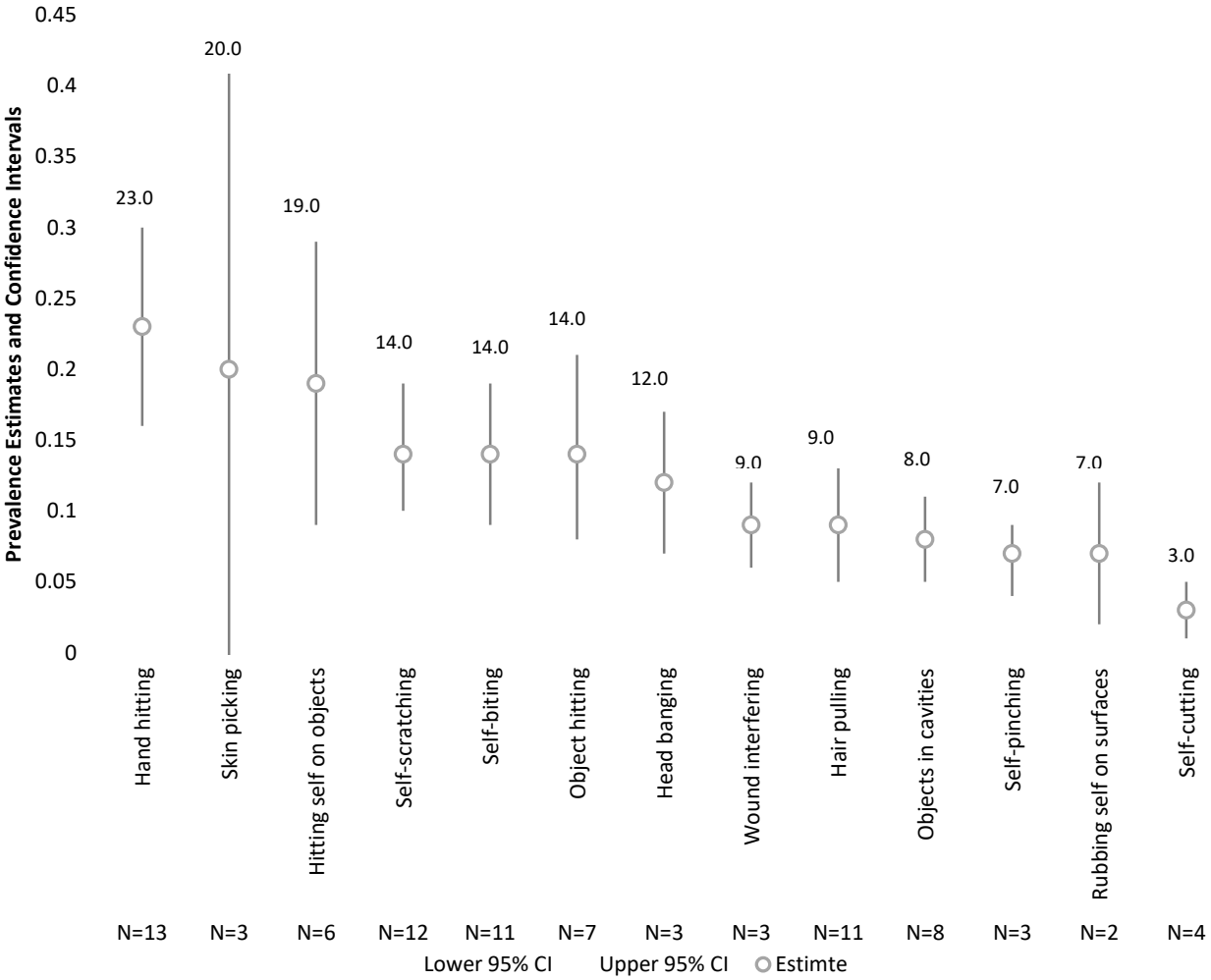

Figure 1.5. Pooled prevalence estimates (%), with 95% confidence intervals for 13 topographies of behaviour using the random-effects model (N refers to number of studies).

1.4.7 Influence of participant characteristics on prevalence rates of different topographies of self-injurious behaviour.

In order to conduct a meta-regression analysis, it is recommended to include at least ten studies which report data on the outcome of interest (Higgins & Green, 2008). As such, meta-regression analyses looking at the influence of participant characteristics on prevalence of behaviour topography were only conducted on four topographies that had sufficient studies reporting data; hair pulling, hand hitting, self-biting and self-scratching. However, not all of the papers that reported data on these topographies included data on intellectual disability, age and gender, therefore, the results of the meta-regression analysis should be interpreted with caution.

Table 1.8. Participant characteristics influencing prevalence of hair pulling, hand biting, self-biting and self-scratching.

	Covariate	Estimate	S.E.	Z	p	Lower 95%CI	Upper 95%CI
Hair pulling	**Age (mean age)**	0.0014	0.0116	0.1235	.901	-0.0212	0.0241
	Gender (% male)	<0.0001	0.0029	0.0081	.993	0.0057	0.0057
	Presence of ID	-0.0013	0.0005	-2.6167	.008	-0.0023	-0.0003
Hand hitting	**Age (mean age)**	0.0245	0.022	1.1111	.266	-0.0187	0.0676
	Gender (% male)	-0.0014	0.0045	-0.3053	.760	-0.0101	0.0074
	Presence of ID	0.0013	0.0013	1.0209	.307	-0.0012	0.0038
Self-biting	**Age (mean age)**	0.0006	0.0138	0.0423	.966	-0.0264	0.0275
	Gender (% male)	-0.0072	0.0043	-1.6956	.09	-0.0155	0.0011
	Presence of ID	-0.0011	0.0016	-0.667	.504	-0.0043	0.0021
Self-scratching	**Age (mean age)**	0	0.0236	0	.585	-0.3902	0.6907
	Gender (% male)	-0.004	0.0036	-1.1305	.269	-0.0111	0.0031
	Presence of ID	-0.0021	0.0007	-3.0253	.002	-0.0035	-0.0008

The association between the proportion of participants with an intellectual disability and prevalence of hair pulling and self-scratching was found to be significant (p=.008 and p=.002, respectively) suggesting that presence of an intellectual disability may increase the prevalence of these topographies of self-injurious behaviour. However, there were no other significant associations between participant characteristics and prevalence of different topographies of self-injurious behaviour.

1.5 Discussion

The prevalence of self-injurious behaviour in ASD was meta-analysed in this study to generate a robust pooled prevalence estimate. Sub-group analyses were completed in order to evaluate how study and participant characteristics influence the overall reported prevalence rates of self-injury, as well as the prevalence of individual topographies of behaviour. This is the first meta-analysis to synthesise prevalence rates of self-injurious behaviour in ASD, and thus is important both clinically and scientifically. The meta-analysis employed a thorough, robust and careful search strategy, adopting key search terms from published literature within the field. Moreover, the use of robust quality criteria further strengthens the findings in this meta-analysis. In summary, prevalence rates of self-injurious behaviour extracted from 37 primary studies were analysed in this meta-analysis, generating a pooled prevalence estimate of 42% for self-injurious behaviour in ASD. There were no significant associations between mean age and proportion of those with an intellectual disability and prevalence of self-injury, however, gender did have an effect on prevalence rates with a higher proportion of male participants associated with a slight decrease in prevalence of self-injurious behaviour. Analysis of the topography data showed hand hitting, skin picking and hitting self against objects to be the most common forms of self-injury, whilst self-cutting was the least common.

The overall pooled prevalence estimate for self-injury in ASD is 42%, which is significantly higher than prevalence estimates for self-harm in the typically developing population. For example, the prevalence of self-harm in typically developing children and adolescents is approximately 8% and 5.9% in adults (Klonsky, 2011; Morgan, Webb, Carr, Kontopantelis, Green, Chew-Graham *et al*., 2017). The difference between these prevalence rates are striking and suggests that those with ASD are a particularly high risk group for self-injury.

A substantial amount of heterogeneity between studies included in the prevalence estimate was observed (I^2 = 96%), therefore it was necessary to conduct further analyses to explore the factors which may have been contributing to the level of heterogeneity. Sub-group analyses revealed that differences in the methodologies and quality of the primary studies did not influence prevalence rates, however, an overall trend could be observed. Notably, for sample identification and assessment of ASD, prevalence estimates decreased as quality of the study characteristics increased. Additionally, prevalence estimates are seen to increase when more robust methods are used to assess self-injurious behaviour. These observed trends may have important implications for future research. For example, future studies should consider optimising the trade-off between representative sample identification and more robust assessment of self-injurious behaviour. That is, prevalence estimates may be influenced more so by self-injury assessment than sample representativeness. Therefore, studies should ensure that self-injurious behaviour is clearly defined, operationalised and measured using reliable and valid questionnaires in order to produce accurate prevalence rates.

The influence of participant characteristics such as age, gender and presence of an intellectual disability on reported prevalence rates were analysed using meta-regression. Interestingly, gender was found to be significantly associated with prevalence of self-injurious behaviour with greater number of males in the sample associated with a slight decrease in prevalence rates. Previous research has replicated these results demonstrating an association between self-injurious behaviour and female gender (Cohen, Tsiouris, Flory, Kim, Freedland, Heaney *et al.*, 2010). Similarly, in a study exploring characteristics of females and males with ASD, Frazier, Georgiades, Bishop & Hardan (2013) found that females exhibited more externalizing behaviours, such as self-injury, compared to males. However, in a sample of individuals with ASD, Baghdadli *et al.*,

(2003) found no association between self-injury and gender. Therefore, further research may be warranted in order to replicate and confirm the association between self-injury and female gender. Given the heightened prevalence of ASD in males than in females, the association between self-injury and gender is of particular importance (Saracino *et al.*, 2010). Females are commonly underrepresented in autism research due to the inherent difficulties in identifying and diagnosing ASD within this group. Particularly in females with intellectual disability, their presentation of symptoms may be misinterpreted and accurate diagnosis is often delayed (Halladay, Bishop, Constantino, Daniels, Koenig, Palmer *et al.*, 2015). Therefore, in order to explore the association between gender and self-injury further, future research should seek to include more female participants in their studies.

There were no significant associations between age and prevalence of self-injury. Within the literature, the reported associations between age and self-injury in ASD vary. In their study, Esbensen, Greenberg, Seltzer & Aman (2009) reported a correlation between older age and significantly lower levels of self-injury, whilst Baghdadli *et al.*, (2003) found no significant association between younger age and self-injury. In a sample of individuals with an intellectual disability of heterogeneous aetiology, Oliver *et al.*, (1987) demonstrated a curvilinear relationship between age and self-injury, with self-injury peaking between the ages of 15 and 25. The authors suggest that the increase in prevalence of self-injury can be explained by the operant model which suggests that self-injury becomes learnt and is shaped by the environment. Given the high prevalence of self-injury in ASD, there is a need for further research exploring the associations between age and self-injury. The majority of studies included in this meta-analysis recruited participants under the age of 18 (72%), therefore, there is a clear need for future studies to include participant samples that contain individuals with ASD from across the lifespan.

Furthermore, there is a need for longitudinal studies to evaluate changes in self-injury over time and across the lifespan.

There was no significant association between presence of an intellectual disability and prevalence of self-injury. Previous research has identified the presence of an intellectual disability as the most common risk marker for self-injurious behaviour (McClintock *et al.*, 2003), therefore, the lack of association between prevalence of self-injury and intellectual disability was unexpected. It might be that in ASD, the absence of an intellectual disability is not as protective against self-injury or self-harm as might be assumed. Given the emerging evidence of the distinction between self-injury in ASD and self-injury in ASD with co-morbid intellectual disability, it might be that presence of an intellectual disability influences the form and function of self-injury rather than the prevalence of the behaviour. Recent research has suggested that self-injury in those with ASD *without* an intellectual disability may be more similar to behaviours observed in those of typical development with mental health difficulties and may serve a similar function (Maddox *et al.*, 2017). In order to better understand these differences, and to inform more accurate causal models in the future, further exploration of the function of self-injury in this group is clearly needed. Furthermore, these results may suggest a need for services that are tailored towards individuals with ASD *without* intellectual disability who present with self-injurious behaviour, particularly given the link between self-harm and suicide and the high rates of death by suicide in the ASD population (Cassidy *et al*., 2014; Kirby, Bakian, Zhang, Bilder, Brooks, Keeshin & Coon, 2019; Richa, Fahed, Khoury & Mishara, 2014). However, the overall quality of assessment of intellectual disability across studies was poor. It is therefore possible that the lack of association between intellectual disability and prevalence of self-injurious behaviour is related to

poor assessment of intellectual disability, therefore, these findings should be interpreted cautiously.

Due to insufficient data, it was not possible to analyse prevalence of self-injury across different levels of ASD severity. However, those studies that did report data on this suggests that greater severity of ASD is associated with increased prevalence of self-injury. In order to further our understanding of the influence of ASD severity on prevalence of self-injurious behaviour, ASD symptomology should be operationalised more clearly and reported in future studies.

Finally, in order to explore the influence of participant characteristics on prevalence of different topographies of self-injury, meta-regression analyses were conducted on the available data. The results of the analysis highlighted an association between the proportion of participants with an intellectual disability and prevalence of hair pulling and self-scratching, suggesting that intellectual disability may influence these topographies of behaviour. However, due to the limited number of data points available for inclusion in the analysis, these results should be interpreted with caution. Overall, few studies reported on the prevalence of different topographies of behaviour making it difficult to explore any differences between those with ASD only and those with ASD *and* co-morbid intellectual disability. Given the emerging evidence that self-injury in those with ASD may serve a different function to that observed in those with ASD and co-morbid intellectual disability, there is a growing need for research to focus on behavioural topography within these two groups. Specifically, increasing our understanding of the differences in the form and function of self-injury in these two clinical groups is of particular importance for shaping interventions. For example, management of self-injury in individuals with high functioning ASD might be better suited to self-harm interventions typically offered to people with mental health

difficulties, such as dialectical behaviour therapy (DBT), rather than interventions solely derived from an operant model.

The results of this meta-analysis are of significant clinical importance. Following the Winterbourne View scandal in 2011, the Government has been committed to transforming care for people with intellectual disabilities and/or ASD who show behaviours that challenge, such as self-injury. In particular, commissioners have been tasked with re-shaping local services and developing new models of care that reflect the clinical needs of the populations they serve. Policy planning and service provision for those with intellectual disabilities and ASD are reliant on the availability of accurate and reliable data on the prevalence of these conditions, as well as associated co-morbidities, such as mental health difficulties and behaviours that challenge. An estimated forty-two percent of individuals with ASD will show self-injurious behaviour at some point in their lifetime. Furthermore, other studies demonstrated that it is likely that the behaviour will persist over time for the majority of individuals once it has been established (Richards *et al*., 2016). Therefore, it is essential that services are aligned to these behaviours in order to reduce the detrimental impact on the individuals and their families, and to work towards improving outcomes for all involved.

In summary, the meta-analysis has generated a robust estimate of 42% for the prevalence of self-injurious behaviour in ASD. Despite significant heterogeneity in the studies, the prevalence of self-injurious behaviour was not affected. The analysis found no association between intellectual disability, age and self-injury. However, gender was found to be significantly associated with self-injury, with more males in the sample being associated with a slightly lower prevalence of self-injury. Analysis of the topography data showed hand hitting, skin picking and hitting self against objects to be the most common forms of self-injury, whilst self-cutting was the least common.

CHAPTER 2
Risk Markers for Self-Injurious Behaviour in Children with Intellectual Disability

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2.1 Abstract

Background: Previous research has identified a number of child and behavioural characteristics that are associated with self-injurious behaviour (SIB) in those with an intellectual disability and developmental delay. However, to date, few studies have explored the unique contribution of each risk marker to the presence of SIB, nor have any studies translated these risk markers into a clinical algorithm that can distinguish between the presence or absence of SIB. Therefore, the aim of the current study was to develop a model that classifies the presence or absence of SIB in children who have these known risk markers, as identified by the Self-injury, Aggression and Destruction-Screening Questionnaire (SAD-SQ).

Method: The study utilised existing data from previous studies that had recruited individuals with a confirmed or suspected intellectual disability or developmental delay and who had used the SAD-SQ as a measure in their studies. These data formed the training sample (N=1540) which was used to develop the risk model to predict presence or absence of SIB. Eight possible predictor variables were entered into the model. Binary logistic regression was used to identify the most predictive risk markers for SIB. These risk markers formed the final risk model and subsequent risk algorithm. The algorithm was then applied to a new test sample of children (N=320) and receiver operating characteristic (ROC) curve analysis used to assess the algorithm's predictive accuracy.

Results: Diagnosis of autism, presence of health conditions, repetitive behaviour, impulsivity and age were predictive of SIB. Gender, diagnosis of a genetic syndrome and level of ability were the least predictive of SIB, and therefore, excluded from the final risk model. At the optimum cut-off of 0.28, the risk algorithm had a sensitivity of 77%

and a specificity of 58%. When applied to the test sample at the same cut-off, the algorithm had a sensitivity of 87% and a specificity of 34% with positive and negative predictive values of 64.3% and 66.7%, respectively. ROC analysis provided an area under the receiver operating curve (AUC) value of .608 which is considered moderate. Analysis of the most severe cases of SIB did not alter the accuracy of the model.

Conclusion: The SAD-SQ is a sensitive screening tool that offers a simple and reliable way of screening those at risk of developing SIB in individuals with a suspected or confirmed intellectual disability or developmental delay. These results are discussed in relation to clinical and theoretical implications and areas for future research.

2.2 Introduction

Developmental delay and intellectual disability are common paediatric conditions, and accurate recognition of these common subtypes of neurodevelopmental disability is essential to ensure correct identification and appropriate management (Shevell, 2008). Developmental delay is defined as the failure to achieve developmental milestones within the expected age range. Objectively, this refers to a significant delay in two or more areas of development in children under the age of five. These developmental domains include cognition, social and emotional skills, speech and language, fine and gross motor skills and activities of daily living (Vasudevan & Suri, 2017). Children with impairments across two or more areas of development are often referred to as having a global developmental delay and may need further assessment of their needs to guide appropriate intervention. On the other hand, intellectual disability refers to significant impairments of intellectual and adaptive functioning that originates in childhood, typically after the age of five, and persists across the lifespan. Intellectual disability is typically categorized by level of impairment into mild (IQ between 50-70), moderate (IQ between 35-49), severe (IQ between 20-34) and profound (IQ<20). Globally, the prevalence of intellectual disability has been reported to vary between 1% and 3%, (Harris, 2006; Maulik, Mascarenhas, Mathers, Dua & Saxena, 2011). Similarly, global developmental delay affects 1-3% of the population of children under the age of five making it one of the most common conditions presenting in paediatric clinics (Mithyanta, Kneen, McCann & Gladstone, 2017). Typically, in England children with a suspected developmental delay will be referred to Child Development Centres (CDCs) for assessment and subsequent intervention where appropriate. In some cases delays in a child's development will improve over time, however, for others developmental delays may be the first indication of other difficulties, such as an intellectual disability or presence of autism or a genetic

syndrome not previously identified. It is important that children are correctly diagnosed and the appropriate treatment and intervention identified in order to improve outcomes for children and their families.

Accurate and timely diagnosis is critical as the presence of intellectual disability has been associated with a number of negative outcomes. For example, research has shown that those with intellectual disability are at an increased risk of developing mental health, emotional and behavioural difficulties (Cormark, Brown & Hastings, 2000; Dykens, 2000; Einfield & Tonge, 1996a; Emerson, Einfield & Stancliffe, 2010; Emerson & Hatton, 2007). Similarly, those with intellectual disability are at greater risk of poorer physical health outcomes (Emerson & Brigham, 2014; Emerson & Hatton, 2014; Emerson, Hatton, Baines & Robertson, 2016) and are more likely to experience health and social inequalities (Emerson, 2007), compared to their typically developing peers. In addition to the negative outcomes experienced by those with intellectual disability, research highlights a number of negative outcomes for families and carers. For example, it has been shown that caring for somebody with intellectual disability is associated with reduced quality of life as a consequence of restrictions to relationships, leisure activities and employment opportunities (Yoong & Koritsas, 2012). Research suggests that parents of children with intellectual disability experience significantly more psychological distress than parents of typically developing children (e.g. Blacher & Hatton, 2001), and this association is strongly related to the emotional and behavioural difficulties that are often present in those with intellectual disability (Emerson, Robertson & Wood, 2004).

In particular, there is considerable evidence suggesting that those with intellectual disability are at an increased risk of presenting with behaviours that challenge such as self-injury, aggression and destruction of property, than those with typical development (McClintock et al, 2003). In particular, self-injury has been found to have a number of

poor outcomes, including higher rates of psychiatric hospitalisation (Mandell, 2008), more reactive physical interventions (Allen, Lowe, Brophy& Moore, 2009), lower quality of life (Beadle-Brown, Murphy & DiTerlizzi, 2009), exclusion from mainstream services (Knapp, Comas-Herrera, Astin, Beecham & Pendaries, 2005), and in some cases, placement in out-of-area residential care (Allen, Lowe, Moore & Brophy, 2007). Moreover, behaviours that challenge, such as self-injury, have been associated with increased costs of care with higher costs associated with more severe intellectual disabilities (Knapp *et al.*, 2005). The reported prevalence rates of self-injury in the intellectual disability population vary from 4% to 24% (e.g. Cooper, Smiley, Allan, Jackson, Mantry *et al*., 2009; Deb, Thomas, & Bright, 2001). Variability in these estimates relate to methodological variations between studies, such as different definitions of self-injury and different sample characteristics (Cooper *et al.*, 2009). Moreover, research investigating the persistence of self-injury suggests that the behaviour is highly persistent in those with an intellectual disability with Taylor, Oliver and Murphy (2011) reporting approximately 84% persistence over an 18 year period, Emerson, Kiernan, Alborz, Reeves, Mason, Swarbrick *et al.,* (2001) 71% over a 7 year period and Cooper *et al*., (2009) 62% over a 2 year period. Collectively, these data provide strong evidence for the prevalence and persistence of self-injurious behaviour over time, as well as the poor outcomes associated with the behaviour, thus highlighting self-injury as a behaviour of significant clinical importance in the intellectual disability population.

The majority of evidence based interventions for self-injurious behaviour are implemented once the behaviour has already emerged. For example, NICE guidance (2015) for behaviours that challenge suggests the use of functional assessments and behavioural interventions, such as positive behavioural support (PBS) plans, to manage behaviours that challenge, such as self-injury. Both functional assessment and

behavioural interventions rely on the identification of antecedents and consequences to a particular behaviour; therefore, these interventions are typically most useful once the behaviour has already been established. Given the burdensome nature of self-injury and the negative outcomes for the individual, families and the cost to health services, there is a clear rationale for the development of early intervention programmes. Such programmes could arguably target those children who are most at risk of developing self-injurious behaviour, but who are not yet showing it. A first step towards doing this is to develop risk algorithms that predict both the presence and onset of self-injurious behaviour. If successful identification of children with self-injury can be achieved, then longitudinal studies can be utilised to explore the prediction of onset of the behaviour, as well as any changes in severity over time.

In order for early intervention programmes to be successful, those at greatest risk of developing severe self-injurious behaviour must first be identified. It is therefore essential to develop a reliable screening tool that can identify those individuals who are at greatest risk. Previous research has found associations between multiple child characteristics and self-injury in those with intellectual disability. These risk markers include greater severity of intellectual disability (Chadwick, Piroth, Walker, Bernard, & Taylor, 2000; Holden & Gitlesen, 2006; McClintock, Hall, & Oliver, 2003) diagnosis of an autism spectrum disorder (Richards, Oliver, Nelson, & Moss, 2012), pain related physical health difficulties (Symons, 2011), specific genetic syndromes (Christie, Bay, Kaufman, Bakay, Borden & Nyhan, 1982; Clarke & Boer, 1998; Collins & Cornish, 2002; Holland, Whittington, Webb, Boer & Clarke, 2003; Symons, Clark, Hatton, Skinner & Bailey, 2003), and repetitive (Bodfish, Crawford, Powell, Parker, Golden & Lewis, 1995; Powell, Bodfish, Parker, Crawford, & Lewis, 1996; Rojahn, Matson, Naglieri, & Mayville, 2004) and impulsive behaviours (Bradley, Summers, Wood, & Bryson, 2004; Cooper, Smiley,

Allan, Jackson, Mantry *et al*., 2009). A number of these child characteristics, such as presence of a genetic syndrome, presence of a profound or severe intellectual disability, repetitive behaviour and ASD are present very early on in childhood and thus may be identified before the onset of clinically significant self-injury (Oliver & Richards, 2015). Whilst an association between these risk markers and self-injury has been found, these risk markers may overlap making it difficult to determine the extent to which each risk marker contributes independently to risk of self-injury. For example, both intellectual disability and ASD are associated with self-injury; however, individuals with intellectual disability are at an increased risk of having co-morbid ASD, making it difficult to determine the contributions of these individual risk markers to self-injury. Similarly, in order to be diagnosed with intellectual disability, an individual must have expressive communication impairment which may play a role in the development of self-injurious behaviour. Currently, clinicians have a large number of potential risk markers for self-injury but no simple way of quantifying and identifying which risk markers are most important in highlighting children who warrant intervention. In order to make early identification of those at risk of self-injury possible, and thus facilitating early intervention, identifying and quantifying the contribution of these risk markers to the presence of self-injury is necessary.

One way to explore the contribution of individual risk markers to the presence of self-injury is to build and test statistical predictive models of risk. Logistic regression is commonly used to develop risk prediction algorithms in large data sets in relation to an outcomes of interest, typically presence or absence of a disease or condition. Typically, logistic regression is used to build a prediction model using a training sample that is later applied to a test sample in order to validate the model and determine a risk cut-off for the outcome of interest. Training and test models are often used in the medical field to predict

risk of disease based on known risk factors. For example, such techniques have been used to predict risk of cardiovascular disease (Weng, Reps, Kai, Garibaldi & Quereshi, 2017), diabetes and hypertension (Farran, Channanath, Behbehani & Thanaraj, 2013), ischaemic heart disease (Kukar, Kononenko, Groselj, Kralj & Fettich, 1999) and inflammatory bowel disease (Wei, Wang, Bradfield, Li, Cardinale, Frackelton *et al.*, 2013). Additionally, this approach has been used in the early detection of dementia (Weakley, Williams, Cook & Schmitter-Edgecombe, 2015), Alzheimer's disease (Bhagwat, Viviano, Voineskos & Chakravarty, 2018; Rabin, Pare, Saykin, Brown, Wishart, Flashman & Santulli, 2009) and first episode psychosis (Borgwardt, Koutsouleris, Aston, Studerus, Smieskova, Riecher-Rossler *et al.*, 2013) which have informed early intervention models. Therefore, in order to identify those at greatest risk of self-injury, and thus, facilitating early intervention, the use of similar techniques is a crucial next step in the research on risk markers for self-injurious behaviour. By quantifying and modelling the contribution of individual risk markers to the presence of self-injury, it is hoped that identification of those at greatest risk of self-injury will be possible.

Previous research has developed the Self-Injury, Aggression and Destruction Screening Questionnaire (SAD-SQ), which is a short and accessible measure of child characteristics and behavioural risk markers known to be associated with behaviours that challenge, such as self-injury (Davies & Oliver, 2016). The aim of the current empirical study is to develop and evaluate a predictive algorithm, drawn from SAD-SQ data, to distinguish between those with and without self-injurious behaviour based on known risk markers for self-injury.

The study aims to do this by:

1) Identifying which child and behavioural characteristics are most predictive of self-injurious behaviour.
2) Developing a risk algorithm that stratifies children into low and high risk groups for self-injury. The algorithm will be created from a large historical (training) data set and take into account child characteristics and behavioural risk markers, as identified by the SAD-SQ and the presence and severity of existing self-injurious behaviour.
3) Testing the performance of the risk algorithm by applying it to a new (test) dataset to see whether those who are classified as 'high risk' based on their risk markers are those who show self-injurious behaviour.

2.3 Method

2.3.1 Recruitment[61]

Two samples were recruited for this study; one sample comprised the training dataset and the other comprised the test dataset. The training sample comprised historical data on 1540 participants held on a secure database at the Cerebra Centre for Neurodevelopmental Disorders, who had taken part in previous research studies utilising the SAD-SQ. Data from these participants were used to develop the risk algorithm in this study. Participants had taken part in previous research studies conducted by Davies & Oliver (2016), Handley (2014) and Richards, Davies and Oliver (2017). Participants from these studies were recruited via a number of routes. In the study by Davies & Oliver (2016) participants between the ages of 2 and 12 years were recruited from schools catering for children with severe intellectual disability. Similarly, in the study by Handley (2014), participants between the ages of 2 and 11 years were recruited from Child Development Centres and Special Needs schools within Birmingham. Finally, in their study, Richards *et al.*, (2017) collected data from adults and children with ASD attending National Autistic Society (NAS) adult services or NAS schools.[62]

The test sample comprised 320 children aged 2-11 years with a probable (suspected or confirmed) developmental delay. Participants were recruited via Child Development Centres (CDCs) and Special Needs Schools in the West Midlands and from an existing

[61] This research project is part of a larger study which involves another Clinical Psychology Trainee. The dataset used in this empirical study is drawn from the same sample for both projects, although each project is looking at separate forms of behaviours that challenge. The current empirical study is focused on self-injurious behaviour whereas the other study focuses on destruction and aggression.

[62] See original papers for full details on how their participant samples were recruited.

database of children with rare genetic syndromes and/or autism. Data from these participants were used to test the predictive accuracy of the risk algorithm.

2.3.1.1 Test Sample Recruitment: Special Needs Schools

43 Special Needs Schools were contacted and invited to participate in the study with 11 schools consenting to participate (25.6%). A total of 928 parents/carers were contacted via these schools with 86 completing the questionnaire pack (return rate 9.3%).

2.3.1.2 Test Sample Recruitment: Child Development Centres

All five Child Development Centres in Birmingham consented to be involved in the recruitment of participants for this study. Members of the research team attended a total of 155 paediatric clinics across the CDCs and recruited a total of 194 children via this route (1765 packs handed out to parents; return rate 11%).

2.3.1.3 Database

181 parents/carers of children whose data were contained on the database were contacted and invited to take part in the study with 45 completing and returning the questionnaires (return rate 24.9%).

2.3.2 **Procedure for recruitment of the test dataset**

All parents/carers received an information sheet, cover letter, consent form and the SAD-SQ(R) (see Appendix C). To avoid priming, the study was called "Behaviours of children with neurodevelopmental difficulties" and parents were informed that the aim of the study was to describe and assess different behaviours of children with a developmental delay. For parents/carers of children who were recruited via their child's Special Needs School, recruitment occurred via one of two routes. The first route involved contacting the head teachers of the Special Needs Schools and asking them to distribute the questionnaire packs by placing them inside the children's school bags. Parents/carers who wished to

participate completed the consent form and questionnaire and returned them via a prepaid envelope that was provided to them. Parents/carers were informed they could contact a member of the research team if they had any queries. A reminder letter was distributed via the same method two weeks after the questionnaire packs were distributed.

The second route involved members of the research team attending school clinics run by health professionals such as community paediatricians, nurses, physiotherapists, occupational therapists and speech and language therapists. Parent's had received an initial contact letter along with their child's appointment letter informing them that there would be a member of the research team present at the school while they attended their appointment and who might approach them to discuss the research with them. Parents/carers who wished to take part in the research study provided their written consent and either completed the questionnaire at the clinic with the assistance of the researcher, or completed it at home at a convenient time returning the questionnaire via a prepaid envelope.

Similarly to the school clinic route, parents/carers who's children were recruited via their child's CDC were approached by members of the research team and invited to participate in the study whilst attending an appointment with their child's paediatrician. Parents had received an initial contact letter along with their child's appointment letter informing them that there would be a member of the research team in the waiting room while they attended their appointment who might approach them to discuss the research with them. Parents/carers who wished to take part in the research study provided their written consent and either completed the questionnaire at the clinic with the assistance of the researcher, or completed it at home at a convenient time returning the questionnaire via a prepaid envelope.

Finally, recruitment via the database route involved contacting parents/carers of children who had taken part in previous research, and who had consented to be contacted for future research studies. Members of the research team identified children on the database who met the inclusion criteria for the study. Parents/carers whose children met the inclusion criteria were sent an information pack containing an initial contact letter, consent form, questionnaire and a pre-paid envelope.

Ethical approval for this study was obtained from NHS ethics (see Appendix D).

2.3.3 Participants

Participants were excluded from the study if data relating to self-injurious behaviour were missing. These exclusions resulted in a total of 1540 participants being included in the training sample and 320 participants in the test sample.[63]

Table 2.1 presents the demographic characteristics of the sample. The mean age of the training sample was 11.34 years (SD=10.84; Range = 1.00-61.00) and 1063 (69%) were male. The mean age of the test sample was 6.75 (SD=2.64; Range = 1.00-13.00) and 230 (71.4%) were male.

A series of Mann Whitney U and Chi-Square analyses were conducted to detect possible significant differences between participants in the two samples. The results of these analyses revealed that there were no significant differences between the two samples with respect to gender, level of ability and presence of ASD. However, the participants in the test sample were significantly younger than the participants in the training sample (*U*=184503.00, p = <.001) and showed significantly more impulsive (*U* = 137619.500, p

[63] The database comprised 1552 participants in total but 12 were excluded due to missing self-injury data. 325 participants were recruited in the second sample but five were excluded based on missing self-injury data.

= <.001), repetitive (χ^2 (1, N= 1791) = 26.804, p = <.001) and self-injurious behaviour (χ^2 (1, N= 1185) = 86.687, p = <.001). Similarly, there were a significantly higher number of participants in the test sample that were diagnosed with a genetic syndrome (χ^2 (1, N= 1005), p = .004) and significantly more health conditions present in the test sample than in the training sample (*U*=165828.00, p = <.001).

Table 2.1. Mean age (standard deviation) and range, percentage of males, percentage of participants with ASD, genetic syndrome, low and high ability and mean (standard deviation) health problems, frequency of repetitive behaviour and impulsivity score.

		Training Sample	Test Sample	χ^2 / U	p
N		1540	320		
Age[64]	Mean (SD)	11.34 (10.84)	6.74 (2.64)	*U*=184503.00	**<.001**
	Range	*1.00-61.00*	*1.00-13.00*		
Gender[65]	Male	1063	230	χ^2 = .293	.588
	(%)	(69.0)	(71.8)		
ASD[66]	Present	861	201	χ^2 = .000	.988
	(%)	(55.9)	(62.8)		
Genetic Syndrome[67]	Present	158	70	χ^2 = 8.357	**.004**
	(%)	(10.3)	(21.8)		
Level of Ability[68]	Low Ability	748	196	χ^2 = 3.292	.070
	(%)	(48.6)	(61.2)		
	High Ability	578	119		
	(%)	(37.5)	(37.2)		
Health Problems[69]	None	697	66	*U*= 165828.00	**<.001**
	(%)	(45.3)	(20.6)		
	>1	811	255		
	(%)	(52.7)	(79.6)		
Repetitive Behaviour[70]	Mean (SD)	0.97 (1.18)	1.86 (1.56)	χ^2 = 26.804	**<.001**
	Present	1127	254		
	(%)	(73.2)	(79.3)		
Impulsive Behaviour[72]	Mean (SD)[71]	1.84 (1.58)	2.35 (1.49)	*U* = 137619.500	**<.001**
	Present	1137	301		
	(%)	(73.8)	(94.1)		
Self-injurious behaviour	Mean (SD)[73]	3.44 (2.84)	5.61 (2.54)	χ^2 = 86.687	**<.001**
	Present	486	189		
	(%)	(31.6)	(59.1)		

[64] Age data reported for N=1473 (training sample)
[65] Gender data reported for N=1506 (training sample), N=319 (test sample)
[66] ASD data reported for N=1346 (training sample), N=313 (test sample)
[67] Genetic syndrome data reported for N=1006 (training sample), N=305 (test sample)
[68] Ability data reported for N= 1326 (training sample), N=315 (test sample)
[69] Health problems data reported for N=1508, N=321 (test sample)
[70] Repetitive behaviour data reported for N=1476, N=315 (test sample)
[71] Recorded on a frequency scale of 0-4
[72] Impulsive behaviour data reported for N=1485 (training sample), N=319 (test sample)
[73] Scores recorded between 0-8

2.3.4 Measures

The SAD-SQ has been developed as a screening measure to assess putative risk markers for behaviours that challenge, namely self-injurious behaviour, aggression and destruction of property. The measure assesses risk markers for self-injurious behaviour which includes level of ability, diagnosis of ASD, pain related health conditions, diagnosis of a genetic syndrome, and repetitive and impulsive behaviour. The SAD-SQ was developed by examining all current published questionnaires that target these three behaviours and condensing them to conclude the minimum number of questions required to effectively measure the behavioural construct (Davies & Oliver, 2016). Specific components of the questionnaire assess ability, health problems, level of activity, repetitive behaviour and level of behaviours that challenge. Responses to items on the SAD-SQ are either binary yes/no, Likert or short answer to minimise time pressure. Davies and Oliver (2016) report good reliability of the SAD-SQ with inter-rater reliability ranging from .21 to .47, as well as good concurrent and convergent validity of the SAD-SQ. For example, participants classed as 'high risk' on the SAD-SQ, scored significantly higher on standardised measures of over activity ($U = 33$, $p = 0.001$), impulsiveness ($Z = -2.727$, $p < 0.008$), repetitive ($U = 0.49$, $p = .003$) and restricted ($U = 61.5$, $p = 0.017$) behaviours than low risk participants (Davies & Oliver, 2016).

There are two versions of the SAD-SQ, one for children under the age of six which comprises 54 items (see Appendix E) and one for children over the age of six which comprises 64 items (see Appendix F). The difference between the two measures is due to variation in the measurement of severity of developmental delay. The Denver Developmental Screening Test II (Frankenburg, Dodds, Archer, Shapiro & Bresnick, 1992) is used for children under 6, whilst the Wessex Behaviour Scale (Kushlick, Blunden

& Cox, 1973) is used for children over 6 years of age. This study utilised the SAD-SQ(R) which is a revised version of the SAD-SQ. The SAD-SQ(R) was revised to include an assessment of the child's current communication skills. It was considered important to assess communication, as these skills significantly relate to socially reinforced functions of behaviours that challenge, such as escape from demand or gaining attention. Therefore, assessment of these skills is necessary to guide future interventions; however, these communication data were not analysed in the present study.

2.4 Data Analysis

2.4.1 Developing a clinical risk algorithm for self-injurious behaviour

In order to develop an algorithm that identifies those with self-injurious behaviour based on presence of risk markers, participants from the historical database were used to create a training sample. The training sample data were used to identify the optimal predictive model specification. This was achieved using backward step-wise logistic regression in which all explanatory variables were entered into the model and removed in a step-wise manner to find a reduced model that best explained the data. The optimised model was then applied to a new data set, known as the test sample, to test predictive accuracy and generalisation of the model using receiver operating characteristic (ROC) curve analysis and the area under the curve (AUC). AUC provides a measure of how well the model distinguishes between the presence and absence of self-injury. The AUC value achieved from ROC can range between 0 and 1, with 0 representing poor classification and 1 suggesting excellent classification. An AUC value of 0.7 is generally considered good for a screening tool (Rice & Harris, 2005). Sensitivity and specificity as well as positive and negative predictive values were calculated.

The SAD-SQ and SAD-SQ(R) require respondents to record presence of self-injurious behaviour on a frequency scale of 0-4. In order to conduct logistic regression, these data were converted into binary responses of present or absent. In order to maximise the dataset and include as many participants as possible, all responses indicating presence of self-injury, regardless of frequency, were included.

Finally, as well as recording the presence or absence of self-injurious behaviour, the SAD-SQ and SAD-SQ(R) require respondents to rate how difficult the behaviour is to manage and how concerned they are about the behaviour on a scale of 0-4, where 0 represents "not difficult" and "not at all concerned" and 4 represents "extremely difficult" and "extremely concerned." Therefore, severity of self-injury might be assumed by higher scores on these two indices. In order to test this hypothesis, Kruskal-Wallis analysis was used to compare risk prediction values across different severity levels of self-injury using the management and concern indices. Post-hoc analyses were used to explore any associations between different group pairings. Significance values were adjusted by the Bonferroni correction to reduce the likelihood of a Type 1 error occurring.

2.5 Results

2.5.1 Model Identification

In order to test the first aim of the study, a backward step-wise binary logistic regression (Hosmer, Lemeshow & Sturdivant, 2013) was used to identify the risk markers that were most associated with self-injurious behaviour, and therefore, the optimal predictive model specification. At the first iteration of the logistic regression, the six risk markers included in the SAD-SQ and known to be associated with self-injurious behaviour, as well as age and gender, were entered as potential predictor variables into the model. Presence or absence of self-injurious behaviour was entered as the dependant variable into the model. Age, presence of ASD, presence of a health condition, repetitive behaviour and impulsive behaviour were significantly associated with self-injury (see Table 2.2 for model outputs). This resulted in a model χ^2 = 138.332 (df = 8, N=678[74]; p = <0.001). This equation afforded an overall percentage correct prediction of 72%.

At the second iteration of the logistic regression, diagnosis of a genetic syndrome was removed from the equation as it was found to be the third least predictive of self-injurious behaviour and made no significant contribution to the presence of self-injury. This resulted in a model χ^2 = 138.183 (df = 7, N=678; p = < 0.001). This equation afforded an overall percentage correct prediction of 72.1%.

At the third iteration of the logistic regression, gender was removed from the equation as it was found to be the second least predictive of self-injurious behaviour and made no significant contribution to the presence of self-injury. This resulted in a model χ^2 =

[74] Due to missing data for key variables entered into the logistic regression model, 873 participants were excluded from the analysis

137.675 (df = 6, N=678; p = < 0.001). This equation afforded an overall percentage correct prediction of 72.1%.

At the final iteration of the logistic regression, level of ability was removed from the equation as it was found to be the least predictive of self-injurious behaviour and made no significant contribution to the presence of self-injury. This resulted in a model χ^2 = 135.203 (df = 5, N=678; p = <0.001). This equation afforded an overall percentage correct prediction of 72.6%. A summary of the stepwise model selection is provided in Table 2.2 and the omnibus test of model coefficients in Table 2.3.

Table 2.2. Backward step-wise logistic regression identifying the optimal predictive model.

Step	Predictor variable	B	S.E	Wald	df	p	Exp(B)	95% CI for Exp(B)	
								Lower	Upper
1	Age	.020	.009	5.074	1	.024	1.020	1.003	1.038
	Gender	-.147	.212	.481	1	.488	.863	.570	1.308
	Level Of Ability	-.321	.192	2.800	1	.094	.725	.498	1.057
	ASD	.751	.218	11.876	1	.001	2.119	1.382	3.248
	Genetic Syndrome	-.113	.293	.148	1	.700	.893	.503	1.587
	Health Problems	.229	.074	9.563	1	.002	1.258	1.088	1.454
	Repetitive behaviour	.215	.061	12.435	1	<.001	1.240	1.100	1.397
	Impulsive behaviour	.203	.035	33.362	1	<.001	1.225	1.144	1.313
	Constant	-2.730	.290	88.715	1	<.001	.065		
2	Age	.020	.009	5.189	1	.023	1.020	1.003	1.038
	Gender	-.151	.212	.505	1	.477	.860	.568	1.303
	Level Of Ability	-.312	.191	2.679	1	.102	.732	.504	1.063
	ASD	.761	.217	12.348	1	<.001	2.140	1.400	3.271
	Health Problems	.226	.074	9.435	1	.002	1.254	1.085	1.449
	Repetitive behaviour	.215	.061	12.394	1	<.001	1.239	1.100	1.397
	Impulsive behaviour	.204	.035	33.647	1	<.001	1.226	1.144	1.313
	Constant	-2.754	.284	94.151	1	<.001	.064		

Step	Predictor variable	B	S.E	Wald	df	p	Exp(B)	95% CI for Exp(B)	
								Lower	Upper
3	Age	.020	.009	4.961	1	.026	1.020	1.002	1.038
	Level Of Ability	-.297	.189	2.464	1	.116	.743	.512	1.077
	ASD	.790	.213	13.800	1	<.001	2.204	1.452	3.344
	Health Problems	.224	.073	9.254	1	.002	1.251	1.083	1.444
	Repetitive behaviour	.216	.061	12.528	1	<.001	1.241	1.101	1.398
	Impulsive behaviour	.204	.035	33.766	1	<.001	1.226	1.145	1.314
	Constant	-2.814	.272	106.754	1	<.001	.060		
4	Age	.019	.009	4.831	1	.028	1.020	1.002	1.037
	ASD	.779	.212	13.467	1	<.001	2.179	1.437	3.302
	Health Problems	.230	.073	9.837	1	.002	1.258	1.090	1.453
	Repetitive behaviour	.238	.059	16.139	1	<.001	1.269	1.130	1.425
	Impulsive behaviour	.200	.035	32.755	1	<.001	1.222	1.141	1.309
	Constant	-2.964	.257	133.046	1	<.001	.052		

Table 2.2 shows each step of the logistic regression model with variables removed at each step. Step 1 is the first iteration of the model with all eight variables included and step 4 is the final regression model with the least predictive variables removed.

Table 2.3. Omnibus tests of model coefficients.

		Chi-square	df	p
1	Step	138.332	8	<.001
	Block	138.332	8	<.001
	Model	138.332	8	<.001
2	Step	-.149	1	.699
	Block	138.183	7	<.001
	Model	138.183	7	<.001
3	Step	-.508	1	.476
	Block	137.675	6	<.001
	Model	137.675	6	<.001
4	Step	-2.472	1	.116
	Block	135.203	5	<.001
	Model	135.203	5	<.001

Gender, diagnosis of a genetic syndrome and level of ability were found to be the least predictive of self-injurious behaviour and therefore were not included in the final logistic regression model. Due to a large proportion of missing data accounted for by these three variables, excluding them from the model resulted in an increased number of participants being included in the final model (N=1195). The beta coefficients and associated significance tests for the variables within the final predictive model are described in Table 2.4.

Table 2.4. Variables included in the final logistic regression model including beta coefficients and significance values.

	B	S.E.	Wald	df	Sig.	Exp(B)	95% C.I.for EXP(B)	
							Lower	Upper
Age	.026	.006	17.061	1	<.001	1.027	1.014	1.039
ASD	.556	.165	11.330	1	.001	1.744	1.262	2.411
Health Problems	.257	.057	19.922	1	<.001	1.293	1.155	1.447
Repetitive behaviour	.161	.042	14.424	1	<.001	1.175	1.081	1.277
Impulsive behaviour	.209	.025	67.662	1	<.001	1.233	1.173	1.296
Constant	-2.806	.191	215.842	1	<.001	.060		

2.5.2 Identifying the optimum cut-off

The five variables in the predictive model can be expressed in the following form to achieve a prediction variable that is never less than 0 and never greater than 1.

$$prediction = \frac{1}{1 + e^{-t}}$$

$t = -2.806 + (0.026\ Age) + (\ 0.556\ ASD) + (0.257\ Health\ Problems) + (0.161\ Repetitive\ Behaviour) + (0.209\ Impulsive\ Behaviour)$

Typically, logistic regression selects a cut-off of 0.5 to identify risk categories. This value is often selected as it results in a balance between false positive and false negative predictions. However, for the purpose of screening for self-injurious behaviour, it may be preferable to select a cut-off value that maximises sensitivity at the expense of specificity to minimise the risk of too many false negatives. Table 2.5 presents false and true negative and positive rates for a range of different cut-off values.

Table 2.5. Sensitivity and specificity at different cut-off values in the training sample.

Cut-off	TP	TN	FP	FN	Sensitivity	Specificity	Proportion Correct %	PPV %	NPV %
0.2	361	310	481	43	0.89	0.39	56.2	42.8	87.8
0.22	349	351	440	55	0.86	0.44	58.6	44.2	86.5
0.24	338	399	392	66	0.83	0.50	61.7	46.3	85.8
0.26	327	431	360	77	0.80	0.54	63.4	47.6	84.8
0.28	315	459	332	89	0.77	0.58	64.8	48.7	83.8
0.3	302	483	308	102	0.74	0.61	65.7	49.5	82.6
0.32	291	511	280	113	0.72	0.64	67.1	51.0	81.9
0.34	275	539	252	129	0.68	0.68	68.1	52.2	80.7
0.36	256	561	230	148	0.63	0.70	68.4	52.7	79.1
0.38	246	583	208	158	0.60	0.73	69.4	54.2	78.7
0.4	234	606	185	170	0.57	0.76	70.3	55.8	78.1
0.5	159	688	103	245	0.39	0.86	70.9	60.7	73.7
0.6	87	751	20	317	0.21	0.97	70.10	81.3	70.3
0.7	37	782	9	367	0.09	0.98	68.5	80.4	68.0
0.8	13	787	4	391	0.03	0.99	66.9	59.1	66.8

TP = true positive, TN = true negative, FP = false positive, FN = false negative
PPV = positive predictive value
NPV = negative predictive value

2.5.3 Sensitivity and specificity in the training sample

As can be seen in Table 2.5, a cut-off in the range of 0.2 to 0.32 provides that best compromise of sensitivity to specificity required of a screening tool. A cut-off of 0.2 provides the most robust level of sensitivity by accurately predicting 89% (PPV 42.8%) of true positive cases in the training sample. However, at this level of sensitivity, the level of specificity is poor with only 39% of true negative cases identified and an overall accuracy of 56.2% (NPV 87.8%). Whilst it is important to maximise sensitivity in order for the screening tool to be useful, a level of specificity which is too low could affect the utility of the screening tool by including too many cases where self-injury is not present.

Therefore, a cut-off of 0.28 was selected as the optimal cut-off for the data in this training sample. At this level, the model correctly predicted 315 cases from a total of 404, resulting in a sensitivity of 77% and specificity of 58% (PPV 48.7%; NPV 83.8%).

2.5.4 Cross validation in the test dataset

The performance of the predictive equation derived from the training sample was then assessed using the test sample for the cut-off value 0.28 (see Table 2.6)[75]. The negative and positive predictive values and the area under the receiver operating characteristic curve (AUC) were calculated.

Table 2.6. Sensitivity and specificity in the test sample

Cut-off	TP	TN	FP	FN	Sensitivity	Specificity	Proportion Correct %	PPV %	NPV %	AUC
0.28	153	44	85	22	0.87	0.34	64.8	64.3	66.7	.608

As identified in the training sample, a cut-off of 0.28 provided the optimal cut-off for sensitivity and specificity. When applied to the test dataset, a cut-off of 0.28 yielded a sensitivity value of 87% and a specificity value of 34%, correctly predicting 153 cases of self-injury from a total of 175. At this cut-off, the positive predictive value is 64.3% and the negative predictive value is 66.7%.

[75] Due to missing data for 14 participants, 175 participants were included in the ROC analysis

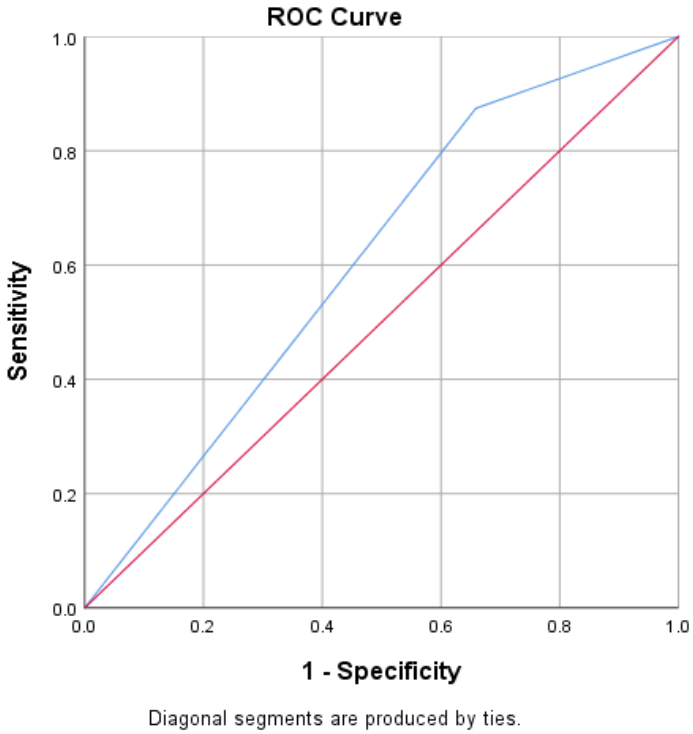

Figure 2.1. Sensitivity and specificity of the risk model at the optimal cut-off of 0.28.

At a cut-off of 0.28 the predictive value of the risk algorithm is considered moderate (AUC = .608).

2.5.5 Association between severity of self-injurious behaviour and level of risk in the test sample

In the test sample, a total of 189 children showed self-injurious behaviour. In 181 cases, parents reported having difficulty managing the behaviour, as indicated by endorsing a response between one and four on the behaviour management index. Similarly, in 177 cases, parents reported being concerned about their child's behaviour, as indicated by endorsing a response between one and four on the behaviour concern index. In order to assess the predictive accuracy of the cut-off in predicting severe cases of self-injurious behaviour, scores on each of these respective behaviour indices were summed to give a maximum possible score of eight. Participants were only included if they reached a

threshold of four or above as this indicated more severe cases of self-injury, which resulted in a total of 132 participants being included. At a cut-off of 0.28 and a severity threshold of four, the algorithm correctly predicted 88% of self-injury cases (N=116). This is comparable to the sensitivity value obtained at this cut-off (87%) suggesting that the algorithm does not perform any better or worse when looking only at the most severe cases of self-injury.

Finally, in order to further explore the association between severity of self-injury (as measured by the behaviour management and concern indices) and level of risk, a Kruskal Wallis test was carried out between 0, 1, 2, 3 and 4. The results of these analyses are presented in Figure 2.2 and 2.3.

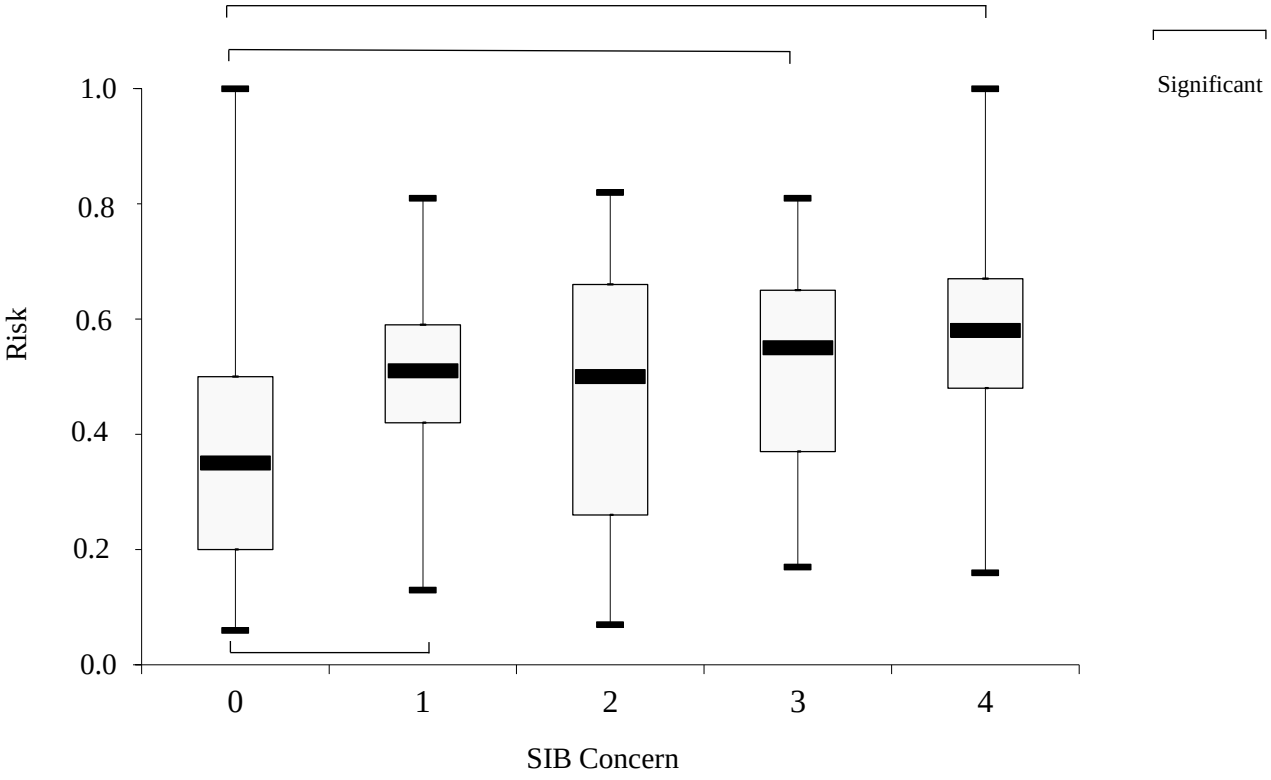

Figure 2.2. Independent-Samples Kruskal-Wallis Test for risk prediction and behaviour concern.

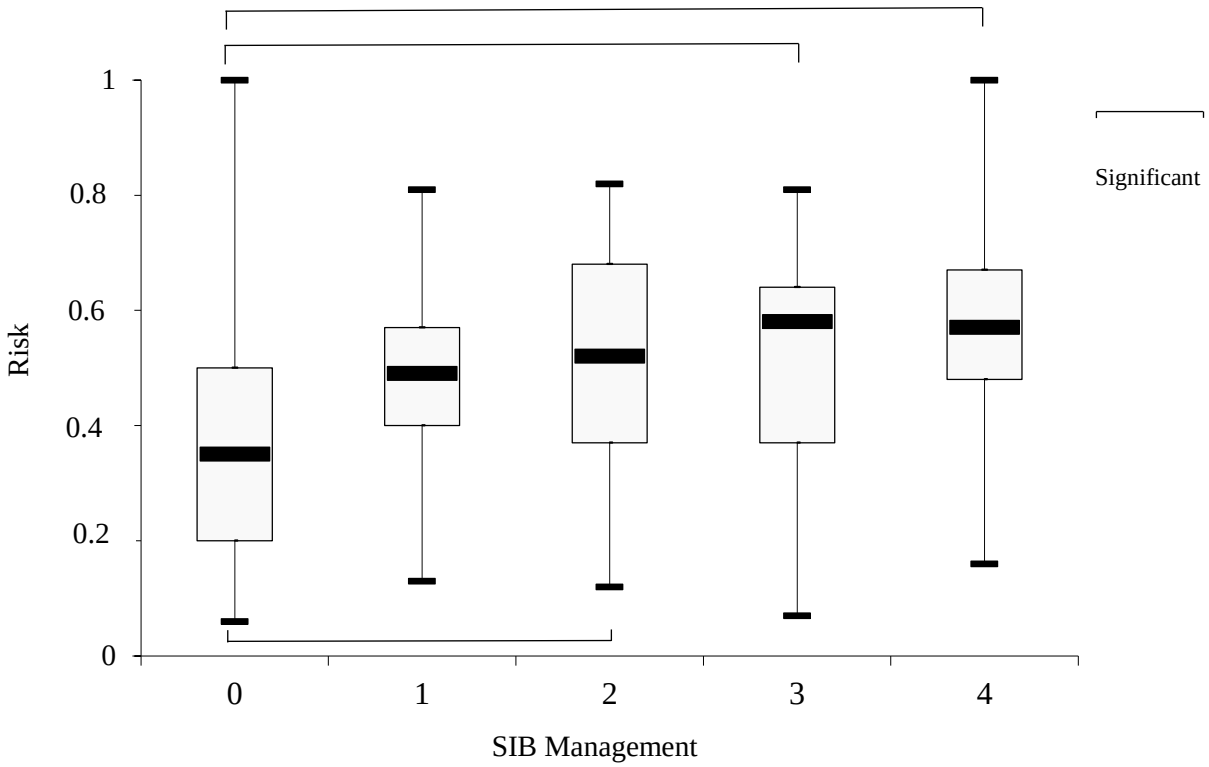

Figure 2.3. Independent-Samples Kruskal-Wallis Test for risk prediction and behaviour management.

The analysis revealed a significant association between behaviour management and level of risk (H (4) = 56.764, p = <.001) and behaviour concern and level of risk (H (4) = 59.575, p = <.001). Post-hoc analyses revealed significant differences between absence of behaviour (score of 0) and three levels of behaviour management difficulty (scores of 2, 3 and 4) and absence of behaviour (score of 0) and three levels of behaviour concern (scores of 1, 3 and 4). However, no significant differences were found between each level of severity (scores of 1, 2, 3 and 4) and level of risk, suggesting that the algorithm does not discriminate between levels of behaviour concern and behaviour management (see Table 2.7). Despite no significant differences between severity levels, an overall trend can be seen for both indices. That is, scores on the behaviour concern and management indices increase slightly as level of risk increases.

Table 2.7. Post-hoc analyses to evaluate differences between level behaviour management difficulty and behaviour concern and level of risk.

	Behaviour Concern		Behaviour Management	
Pair	Test Statistic	p	Test Statistic	p
0-1	-63.076	.009	-49.615	0.25
0-2	-46.199	.078	-73.449	<.001
0-3	-70.384	<.001	-68.782	<.001
0-4	-90.229	<.001	-90.326	<.001
2-1	16.877	1.000	-23.833	1.000
2-3	-24.185	1.000	4.667	1.000
2-4	-44.030	.182	-16.877	1.000
1-3	-7.308	1.000	-19.167	1.000
1-4	-27.153	1.000	-40.711	.285
3-4	-19.845	1.000	-21.544	1.000

Significance values have been adjusted by the Bonferroni correction for multiple tests.

2.6 Discussion

The aim of this study was to develop and evaluate a predictive algorithm drawn from SAD-SQ data to distinguish between those with and without self-injurious behaviour based on known risk markers for self-injury. The study sought to achieve this by first identifying which child and behavioural characteristics are most predictive of self-injurious behaviour, and therefore, those that may be considered the most important risk markers for self-injury. Specifically, this study evaluated how various variables were associated with an increase in the probability of the presence of self-injurious behaviour. There are a number of studies that have identified and described the risk markers that are associated with self-injurious behaviour (e.g. Baghdadli, Pascal, Grisi & Aussilloux, 2003; Davies & Oliver, 2016; Holden & Gitlesen, 2006; Richards, Moss, Nelson & Oliver, 2016; Richards, Davies & Oliver, 2017), however, no studies to date have translated this into a clinical tool that can predict risk of self-injury. Therefore, the second aim of this study was to develop a risk algorithm that predicts presence of self-injurious behaviour based on the most predictive risk markers. This study is the first to explore the predictive value of a screening tool for self-injurious behaviour and furthermore, the utility of the SAD-SQ as screening tool for self-injurious behaviour. Thus, the results of this study are of significant clinical importance. A major strength of this study relates to the large and representative training and test samples which meant the samples were well powered for predictive modelling. Participants in the test sample were recruited from child development centres and schools across the West Midlands and therefore are representative of the children in this region who have development delays or intellectual disability. Furthermore, the success of the model in correctly classifying participants with self-injury in the test sample recruited via these routes is indicative of the validity and generalisability of the model. This is particularly important in highlighting the utility of

the SAD-SQ as a screening measure which can be used in clinical and educational settings.

In order to develop the risk algorithm, six risk markers known to be associated with self-injurious behaviour (ASD, genetic syndrome, level of ability, health problems, repetitive behaviour, and impulsivity; McClintock *et al.*, 2003; Bamford, Richards, Jones & Oliver, in preparation) were entered into the model as possible predictors of self-injury. Although not previously identified as risk markers for self-injury, age and gender were also entered into the model in order to explore whether there was an association between these characteristics and self-injury. Out of a possible eight predictor variables, the risk model identified five as providing a unique contribution to risk of self-injurious behaviour. Consistent with previous literature, the results of this study found that a diagnosis of ASD, presence of one or more health conditions, repetitive behaviour and impulsivity were highly associated with self-injurious behaviour (Bodfish *et al*., 1995; Bradley, Summers, Wood, & Bryson, 2004; Cooper *et al*., 2009; Oliver, Petty, Ruddick & Bacarese-Hamilton, 2012; Powell, Bodfish, Parker, Crawford, & Lewis, 1996; Richards, Oliver, Nelson, & Moss, 2012; Rojahn, Matson, Naglieri, & Mayville, 2004; Symons, 2011). Age was also found to be associated with self-injury and was included in the risk model. This finding is of particular importance as previous studies have found that the risk of self-injury increases significantly after ten years of age until approximately 50 years of age (Kebbon & Windahl, 1986; Rojahn, Borthwick-Duffy & Jacobson, 1993). Moreover, in a recent study Petty, Bacarese-Hamilton, Davies & Oliver (2014) found a surprisingly high prevalence of self-injury in children under the age of five (50.9%), although self-injury was found not to be influenced by increasing age. The findings of these studies, as well as the significant association between age and self-injury in the present study, support the rationale for early intervention for those who are at risk of developing self-

injury but who may not yet be showing the behaviour. In particular, consideration needs to be given to very young children with other known risk markers who can be followed up in longitudinal studies to explore the onset and trajectory of self-injurious behaviour over time. As well as confirming the significant associations between a number of risk markers and self-injury, the present study replicated previous studies investigating the influence of gender and self-injury (e.g. Davies & Oliver, 2016). Consistent with these studies, gender was not significantly associated with self-injurious behaviour and was therefore excluded from the risk model.

The replication of significant associations between a number of child and behavioural characteristics and self-injury in the current study have important clinical and theoretical implications. Having a better understanding of the risk markers that contribute most significantly to self-injury will help to inform future early interventions for behaviours that challenge. For example, a number of studies have suggested a causal relationship between pain and self-injury (Breau, Camfield, McGrath & Finley 2003; Luzzani, Macchini, Valade, Milani & Selicorni, 2003; Symons, 2011) which has been alluded to by the predictive value of the presence of health conditions in the current study, suggesting that this may be a key area that future interventions could address. Moreover, a high number of participants in the training and test sample had one or more health conditions (52.7% and 79.6%, respectively), which is likely to be representative of the prevalence of health conditions in individuals with intellectual disability or developmental delay more generally (e.g. Berg, Arron, Burbidge, Moss & Oliver, 2007; Jansen, Krol, Groothoff, & Post, 2004). Therefore, consideration of the influence of painful health conditions on the development and maintenance of self-injury is important and highlights an important role for physical health care in early intervention for self-injury.

Similarly, the association between repetitive behaviour, impulsivity and self-injury was replicated in this study which lends support to previous studies that have found an association between these behavioural characteristics and self-injury (e.g. Arron *et al*., 2011; Bradley *et al.*, 2004; Cooper *et al.*, 2009; Oliver *et al.*, 2012; Richards *et al.*, 2012). Importantly, the replication of these associations suggest these child and behavioural characteristics play a causal role in the onset and maintenance of self-injury. For example, the theoretical explanations of Turner (1999) of repetitive behaviour and of Nigg (2005) of impulsivity suggest that the onset, maintenance and persistence of self-injury may relate to deficits in executive functioning (Oliver & Richards, 2015), whilst the association between painful health conditions and self-injury is suggestive of a possible underlying function of self-injury in "gating" pain. Furthermore, operant models suggest that once the behaviour has entered a child's repertoire, self-injury is mediated by the external environment through negative and positive reinforcement. As these child and behavioural characteristics are likely to be present early on in childhood, and before self-injury has emerged, assessment of these characteristics provides a unique opportunity to intervene early before self-injury becomes an established behaviour in the child's repertoire. This is of paramount importance if early intervention strategies are to be successful.

Interestingly, despite being identified as the most important risk marker for self-injurious behavior (McClintock *et al.*, 2003), level of ability was not significantly associated with self-injurious behavior in the training sample and was therefore excluded from the final model. Studies that have shown ability to be a risk marker for self-injurious behaviour have typically recruited participants with a range of ability levels (e.g. Arron, Oliver, Moss, Berg & Burbidge, 2011), however, in the present study there were very few children with high levels of ability, as participants were recruited because they had either

a confirmed or suspected intellectual disability or development delay. Therefore, it is likely that the limited number of children with high ability in these samples meant that ability was less strongly predictive of self-injurious behaviour. This is an important finding as early intervention for behaviours that challenge would likely be aimed at children identified through CDCs and/or special schools, who are identified as a result of delays in their ability/development.

Finally, genetic syndrome was found to be least predictive of self-injury and was excluded from the final risk model despite being identified as a risk marker for self-injurious behaviour in previous studies (e.g. Arron, *et al.,* 2011; Christie *et al.,* 1982). However, only 10.3% of participants in the training sample had a genetic syndrome, therefore, it is possible that there were insufficient data pertaining to genetic syndrome for any meaningful association to be found. Furthermore, previous research has identified that only some genetic syndromes are associated with self-injury, including Lesch-Nyhan, Fragile-X, Cornelia de Lange, Cri du Chat and Smith-Magenis Syndromes (Arron *et al.*, 2011; Christie *et al.,* 1982; Colley, Leversha, Voullaire & Rogers, 1990). It was not possible to stratify risk by type of syndrome, and so the inclusion of high risk syndromes (e.g. Cornelia de Lange and Fragile X) with low risk syndromes (e.g. Downs Syndrome) may have flattened effects across the group. Further research in these rare, but arguably high risk syndromes is therefore required.

When evaluated in the test sample, the predictive accuracy of the risk algorithm was promising. At a cut-off of 0.28, the algorithm had a level of sensitivity of 87%. These results suggest that the algorithm can identify those at "high" risk of having self-injurious behaviour and correctly classifies the presence of self-injury 87% of the time. However, at this cut-off, specificity was low suggesting that the algorithm correctly classified absence of self-injury 34% of the time. The ROC analysis produced an AUC value of

.608 suggesting the predictive value of the risk algorithm was moderate. Typically, an AUC of .7 or greater is desirable; however, for the purposes of developing a screening tool, it was felt that high sensitivity over specificity was preferable. Therefore, the results of this study are still encouraging. Furthermore, the level of sensitivity achieved in this study is comparable to or better than those observed in other disease prediction models (e.g. Rabin *et al.*, 2008; Weng *et al.*, 2017).

When assessing the desirability of different levels of sensitivity and specificity, there is a need to consider the costs and benefits of each. A significant consideration is the immediate and long-term burdens on the healthcare system, the treatability of the condition and the psychological effects on the individual and their families (Trevethan, 2017). For the purposes of developing a screening tool for self-injury, it was preferable to maximise sensitivity over specificity to ensure that those at risk of self-injury are not missed. Given the number of negative outcomes associated with self-injury in people with intellectual disabilities, it is of particular importance that individuals who have a number of risk markers for self-injury are able to be identified for targeted early intervention. The low levels of specificity of the SAD-SQ mean that a significant number of children (N = 85 in the test sample) may be identified as being at risk for self-injury but who are not currently showing any behaviour. The consequence of this is that parents and carers of these children may be offered an intervention when their child does not show self-injurious behaviour or has not yet developed the behaviour. The type of early interventions that would be offered to families typically involve parent/carer groups that provide psychoeducation about intellectual disability and behaviours that challenge. These interventions are unlikely to be invasive, unethical or burdensome to parents or their children, and may in fact have value even in the absence of behaviours that challenge. Therefore, there is no anticipated cost to parents of “high” risk children who

may complete an intervention in the absence of their child developing self-injurious behaviour. However, the financial cost of offering interventions to such a large number of parents is something that needs to be considered. Particular consideration should be given to the modes of delivery of early intervention programmes which minimise resources and cost while maximising the number of families the interventions can reach, for example, by developing e-learning packages. Taking into consideration the impact of missing children who have self-injury versus being over-inclusive of children who do not develop self-injury, it was deemed acceptable to select a cut-off that maximised sensitivity over specificity.

The classification of self-injurious behaviour from the data is considered a strength of the study. The SAD-SQ measures presence of self-injury on a Likert-scale of 0-4, where 0 represents no behaviour and 4 represents frequent self-injury. In order to conduct logistic regression on the data, self-injury was converted into a binary "present" or "absent" response. Self-injury was classed as being "present" if parents endorsed any number between 1 and 4, which ensured that all frequencies of self-injury were being captured by the model. As self-injury usually emerges gradually and has the potential to become more severe over time (Berkson *et al*., 2001; Berkson 2002; Richman & Lindauer, 2005) it was felt that in order for the risk model to be as useful as possible, it should include all instances of self-injury, so that it was not limited to predicting only the most frequent and/or severe cases. Furthermore, analysis of the level of risk and severity of behaviour management and behaviour concern revealed significant associations between absence of behaviour and each level of severity, but not between levels of severity. Furthermore, when looking only at the most severe cases of self-injury, the algorithm performed the same. That is, the algorithm correctly classified the presence of self-injury for 87% of cases in the whole sample and 88% in the most severe cases. This might suggest that the

algorithm does not discriminate between levels of severity, as measured by the behaviour management and concern indices. However, as the majority of self-injury cases were classed as severe (N=132), it is possible that the algorithm would be able to discriminate between severity levels if severity of self-injury varied more within the sample.

One of the limitations of the current study was the difference in prevalence of self-injurious behaviour between the two samples. Typically, when a condition is rare it is difficult to achieve good levels of sensitivity and specificity. However, in cases where a condition, such as self-injury, is more common, it becomes easier to achieve good sensitivity and specificity on a measure (Glaros & Kline, 1988). Therefore, in this study it is possible that sensitivity and specificity are slightly inflated due to an increased prevalence of self-injury. However, the study utilised community samples and thus the prevalence of self-injury obtained in these samples may be representative of the base rate of self-injury in community samples more generally.

The current study evaluated the *presence of existing* self-injurious behaviour and the predictive accuracy of a risk algorithm in discriminating between presence and absence of self-injury. However, further work is required in order to demonstrate the accuracy of the model in predicting self-injury in a sample of children who are not yet showing any self-injurious behaviour. Future studies should seek to address this need by using longitudinal designs to explore whether the model, using the algorithm and cut-off derived here, can predict the *emergence* of self-injury in "high" risk children as well as exploring changes in severity over time.

In summary, this study has shown that the SAD-SQ offers a simple and reliable way of screening self-injurious behaviour in individuals with a suspected or confirmed developmental delay or intellectual disability. A cut-off of 0.28 maximises sensitivity at

the expense of specificity, which is deemed acceptable for use as a screening tool. Moreover, the speed and ease of administration make the SAD-SQ more likely to be successfully implemented in busy clinical settings, such as in paediatric clinics.

CHAPTER 3

Executive Summary

3.1. Literature Review

3.1.1. Background

Autism Spectrum Disorder (ASD) is a neurodevelopmental disorder characterised by persistent deficits in social interaction and communication, as well as restricted or repetitive patterns of thoughts, behaviours and interests. ASD is very common, with recent estimates suggesting that 1 in 68 individuals have a diagnosis of ASD. Prevalence rates of ASD have increased significantly in a short period of time as a result of better diagnostic and screening methods, increased community awareness and increased knowledge of the risk factors associated with ASD.

Some individuals with ASD are able to lead independent lives, however, for others the impact of ASD symptoms and co-morbid conditions can severely affect their quality of life. Individuals with ASD are at greater risk of having co-morbid conditions such as sleep disorders, psychiatric diagnoses such as anxiety and depression and intellectual disability, compared to their peers without ASD. In addition, they are more likely to develop behaviours that challenge, such as self-injurious behaviour.

Self-injury refers to aggressive behaviours in which the individual physically harms themselves. Common forms of self-injury in ASD include head banging, hair pulling and self-biting. Self-injury is associated with a number of poor outcomes, including higher rates of psychiatric hospitalisation, more reactive physical interventions, lower quality of

life and exclusion from mainstream services. Furthermore, research shows that self-injury is highly persistent in individuals with ASD.

There has been a lot of published scientific research investigating the prevalence of self-injury in ASD, however, studies vary widely in the prevalence rates that they report. These differences in prevalence rates may be due to differences in the individuals studied and differences in the way that studies were carried out. Additionally, there is emerging evidence that self-injury in those with ASD *without* an intellectual disability may differ from self-injury observed in those with ASD and co-morbid intellectual disability.

In order to ensure that the needs of this population are met, it is important that we accurately estimate the prevalence of self-injury in ASD in order to inform service planning and delivery. Additionally, to ensure that individuals are offered the most appropriate interventions, it is important that we further our understanding of the influence of certain characteristics, such as presence of an intellectual disability.

3.1.2. What did the review do?

A large literature search was conducted to find all the research papers that detailed the prevalence of self-injury in ASD. A system for reviewing the quality of the research papers was devised with each individual paper rated on quality, and an overall prevalence estimate for self-injurious behaviour in ASD was generated. The overall prevalence estimate was influenced more heavily by the highest quality research papers, and least heavily by the poorest quality papers.

3.1.3. What did the review find?

The estimated prevalence of self-injurious behaviour in ASD is 42%. Furthermore, increased prevalence of self-injury was found in studies in which there were more female

participants suggesting an association between self-injury and female gender. Hand-hitting was found to be the most prevalent form of self-injurious behaviour, whereas self-cutting was found to be the least prevalent. The presence of an intellectual disability was found to influence prevalence of some forms of self-injury, but not prevalence of self-injury overall, whereas gender and age were found to have no effect, specifically hair pulling and self-scratching topographies were more common when intellectual disability was present.

3.1.4 What do these findings really mean?

These findings have given us a robust prevalence estimate of self-injury in ASD and shows that the prevalence is high. Moreover, these findings have furthered our understanding of the influence of certain person characteristics on prevalence of self-injury, as well as prevalence of different forms of self-injury, which can help us to plan clinical services more appropriately for individuals with ASD. These findings can also be used to focus future research in this area.

3.2. Empirical Paper

3.2.1. Background

Developmental delay is defined as the failure to achieve developmental milestones within the expected age range. A developmental delay is typically diagnosed when there is a significant delay in two or more areas of development in children under the age of five. These developmental domains include cognition, social and emotional skills, speech and language, fine and gross motor skills and activities of daily living (e.g. washing and dressing). On the other hand, intellectual disability refers to significant impairments of

intellectual and adaptive functioning that originates in childhood, typically after the age of five, and persists across the lifespan.

In some cases delays in a child's development will improve over time, however, for others developmental delays may be the first indication of other difficulties, such as an intellectual disability or presence of autism or a genetic syndrome not previously identified. It is important that children are correctly diagnosed and the appropriate treatment and intervention identified in order to improve outcomes for children and their families.

Accurate and timely diagnosis is critical as the presence of intellectual disability has been associated with a number of negative outcomes. For example, research has shown that those with intellectual disability are at an increased risk of developing mental health, emotional and behavioural difficulties, such as self-injury. Within this population, the presence of self-injury has been associated with a number of negative outcomes for individuals, their families and services, such as increased rates of hospitalizations, out-of-area placements and significant financial costs to health and welfare providers. Furthermore, self-injury has been found to be highly persistent over time, and is therefore a behaviour of significant clinical importance in this population.

The majority of interventions for self-injury are implemented once the behaviour has already emerged, however, given the burdensome nature of self-injury there is a clear rationale for the development of early intervention programmes. Such programmes could arguably target those children who are most at risk of developing self-injurious behaviour but who are not yet showing it.

The first step towards achieving this is to be able to identify those at the highest risk of developing self-injury.

Previous research has identified a number of child and behavioural characteristics that are associated with self-injurious behaviour in those with an intellectual disability and developmental delay. These child and behavioural characteristics are known as risk markers for self-injury. However to date, no studies have explored the unique contribution of each risk marker to the presence of self-injury, nor translated these risk markers into a model that can distinguish between presence or absence of self-injury. Therefore, the aim of the current study was to build upon existing research by developing a model that can accurately classify the presence or absence of self-injury in children who have these known risk markers as identified by the Self-injury, Aggression and Destruction-Screening Questionnaire (SAD-SQ). The SAD-SQ is a brief measure of child and behavioural characteristics.

3.2.2 What did the study do?

The study used the data from an existing database of 1540 children and adults who had taken part in previous research studies to build a risk model for self-injurious behaviour. Analysis was carried out to determine which risk markers were most strongly associated with self-injurious behaviour and these risk markers were included in the model. The risk model was then tested in a new sample of 320 children in order to test how well it could identify those with self-injurious behaviour and those without based on the presence of these risk markers.

3.2.3. What did the study find?

The study identified five risk markers that were most predictive of self-injurious behaviour; age, presence of ASD, presence of one or more health conditions, repetitive behaviour and impulsivity. Presence of a genetic syndrome, gender and level of ability were the least associated with self-injury and were not included in the final risk model.

When tested in a new sample, the model was able to correctly identify children with self-injurious behaviour 87% of the time. The model was less accurate in correctly identifying cases where self-injury was not present with the model correctly identifying non-cases only 34% of the time.

3.2.4. What do these findings really mean?

The findings show that the SAD-SQ is a highly sensitive screening tool that offers a simple and reliable way of screening those at risk of developing self-injurious behaviour in individuals with a developmental delay or intellectual disability. Future work should seek to evaluate the ability of the model to predict the onset of self-injurious behaviour before it emerges as well as any changes in severity over time.

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APPENDICES

Appendix A

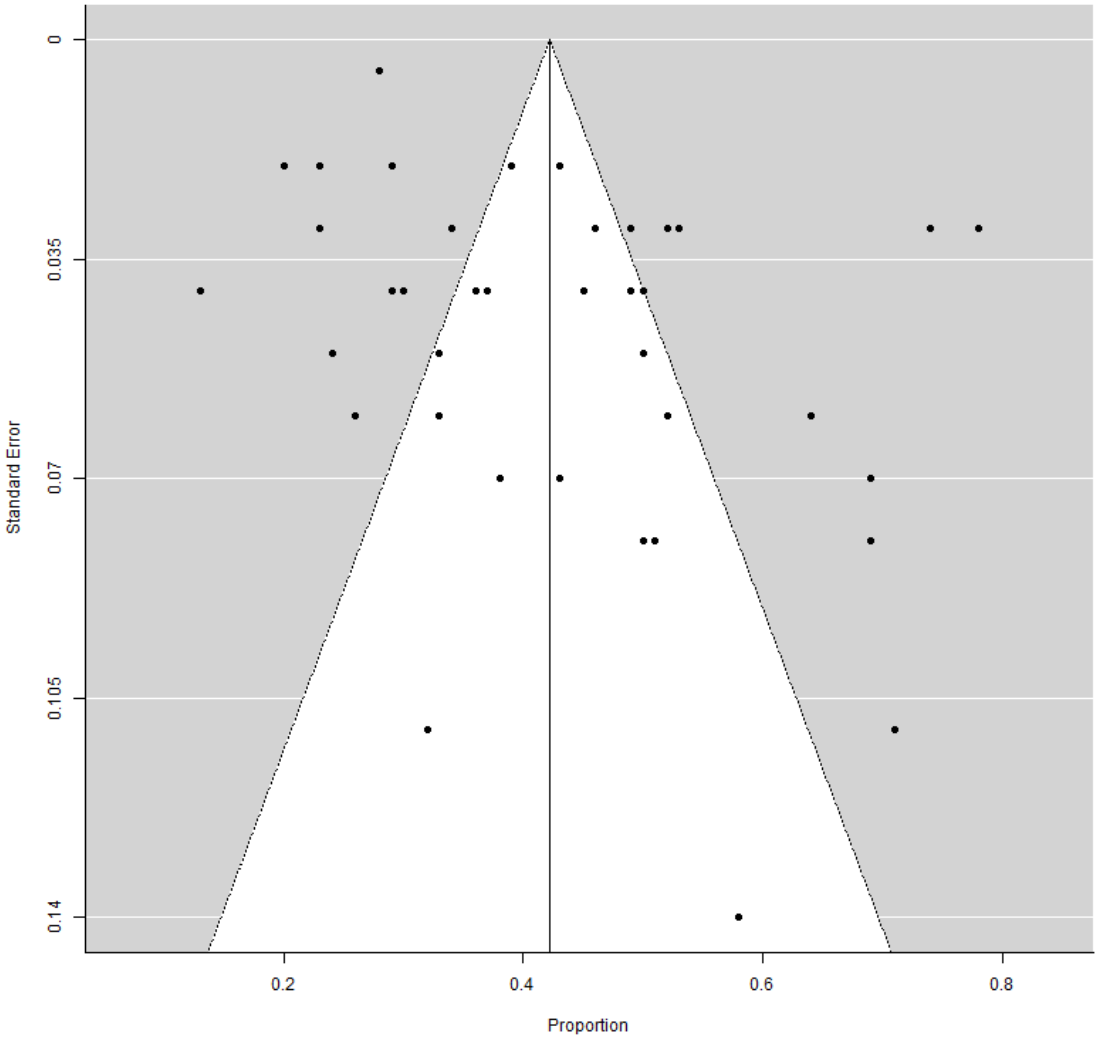

Figure 1.6. A funnel plot depicting publication bias

Appendix B.

Table 1.10 Reporting of the prevalence rates and confidence intervals for hair pulling**.**

Study Name	PR	95%-CI	Weighting in Random Effects Model
Akram 2017	0.1	[0.0412; 0.1588]	7.9
Buono 2010	0.06	[0.0012; 0.1188]	7.9
Duerden 2012	0.19	[0.1312; 0.2488]	7.9
Folch 2018	0.06	[0.0208; 0.0992]	8.6
Gulsrud 2018	0.01	[-0.0096; 0.0296]	9.2
Handen 2018	0.25	[0.2108; 0.2892]	8.6
Kamio 2002	0.02	[0.0004; 0.0396]	9.2
Maddox 2017	0.12	[0.0220; 0.2180]	6.1
Richards 2012	0.06	[0.0208; 0.0992]	8.6
Richards 2017 Child	0.09	[0.0508; 0.1292]	8.6
Richards 2017 Adult	0.1	[0.0608; 0.1392]	8.6
Slingsby 2017	0.02	[-0.0192; 0.0592]	8.6

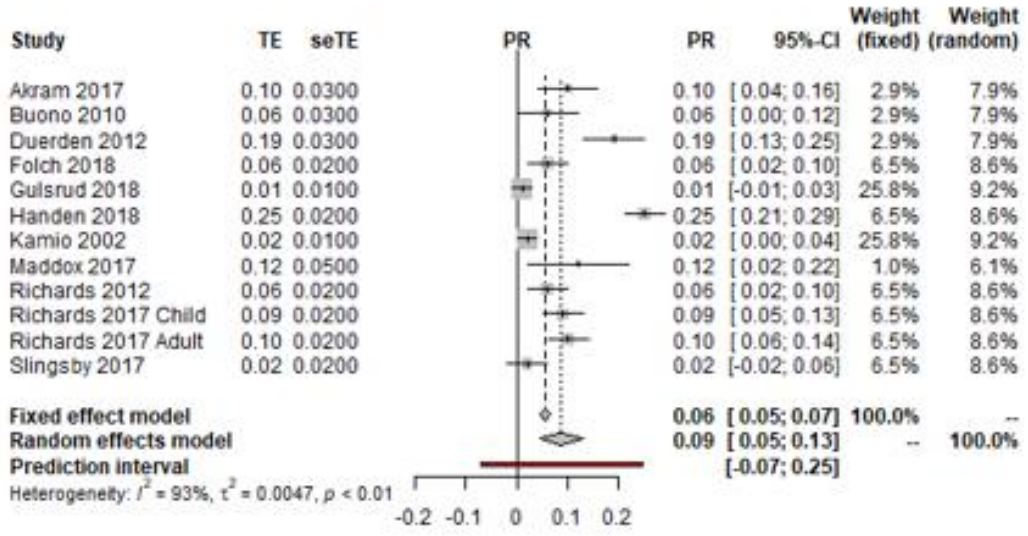

Figure 1.7. The pooled prevalence estimates for hair pulling in ASD using the random-effects model.

Table 1.11 Reporting of the prevalence rates and confidence intervals for hand hitting.

Study Name	PR	95%-CI	Weighting in Random Effects Model
Akram 2017	0.16	[0.0816; 0.2384]	7.1
Buono 2010	0.41	[0.2728; 0.5472]	6
Dominick 2007	0.17	[0.0720; 0.2680]	6.8
Duerden 2012	0.34	[0.2616; 0.4184]	7.1
Folch 2018	0.32	[0.2416; 0.3984]	7.1
Gulsrud 2018	0.09	[0.0508; 0.1292]	7.6
Handen 2018	0.48	[0.4212; 0.5388]	7.4
Hattier 2011	0.06	[0.0404; 0.0796]	7.8
Lecavalier 2006	0.16	[0.1208; 0.1992]	7.6
Maddox 2017	0.12	[0.0220; 0.2180]	6.8
Richards 2012	0.15	[0.0912; 0.2088]	7.4
Richards 2017 Child	0.25	[0.1912; 0.3088]	7.4
Richards 2017 Adult	0.28	[0.2212; 0.3388]	7.4
Slingsby 2017	0.22	[0.1024; 0.3376]	6.4

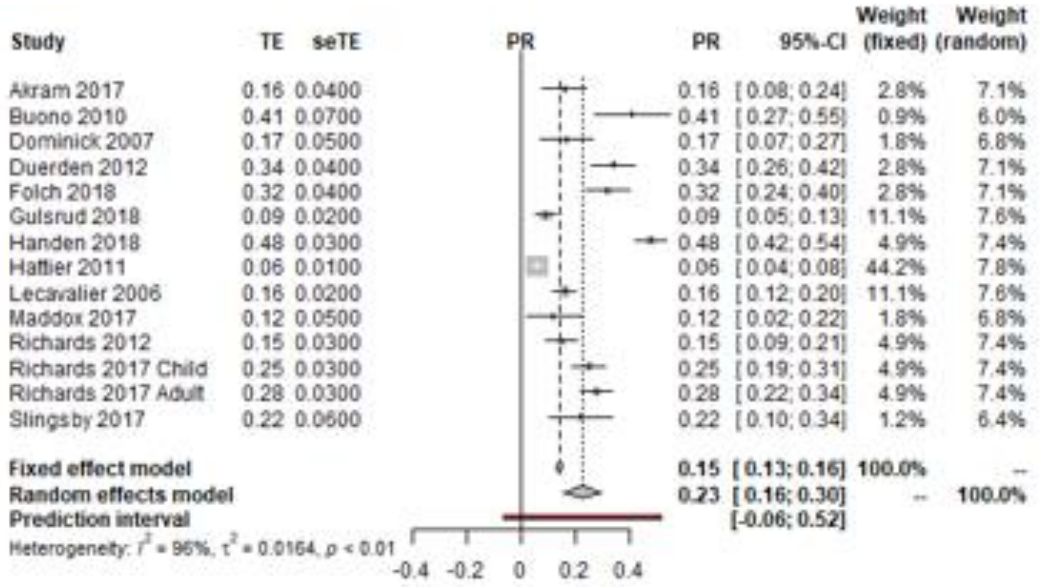

Figure 1.8. The pooled prevalence estimates for hand hitting in ASD using the random-effects model.

Table 1.12. Reporting of the prevalence rates and confidence intervals for head banging.

Study Name	PR	95%-CI	Weighting in Random Effects Model
Dominick 2007	0.2200	[0.1024; 0.3376]	14.3
Folch 2018	0.1000	[0.0608; 0.1392]	49.9
Gulsrud 2018	0.1000	[0.0412; 0.1588]	35.9

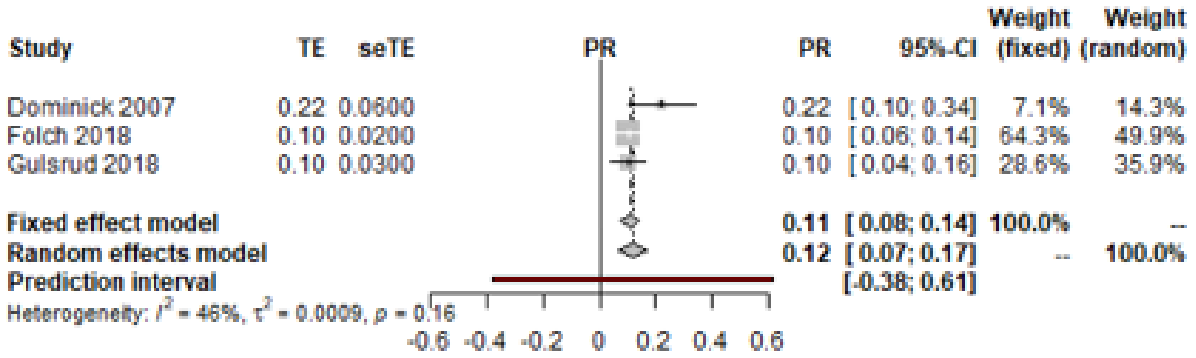

Figure 1.9. The pooled prevalence estimates for head banging in ASD using the random-effects model**.**

Table 1.13. Reporting of the prevalence rates and confidence intervals for hitting self against objects.

Study Name	PR	95%-CI %	Weighting in Random Effects Model
Duerden 2012	0.3	[0.2412; 0.3588]	14.2
Handen 2018	0.47	[0.4112; 0.5288]	14.2
Hattier 2011	0.09	[0.0704; 0.1096]	14.9
Richards 2012	0.08	[0.0408; 0.1192]	14.6
Richards 2017 Child	0.16	[0.1012; 0.2188]	14.2
Richards 2017 Adult	0.16	[0.1012; 0.2188]	14.2
Slingsby 2017	0.07	[-0.0084; 0.1484]	13.6

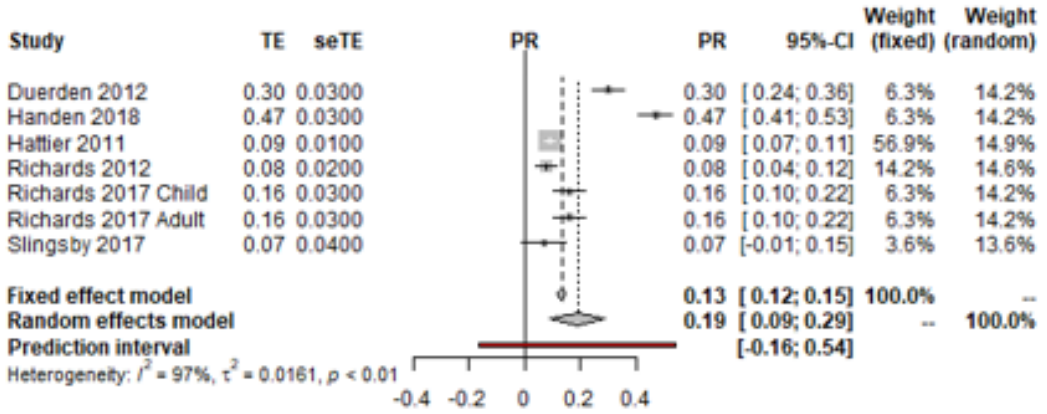

Figure 1.10. The pooled prevalence estimates for hitting self against objects in ASD using the random-effects model.

Table 1.14. Reporting of the prevalence rates and confidence intervals for object hitting.

Study Name	PR	95%-CI %W	Weighting in Random Effects Model
Buono 2010	0.41	[0.2728; 0.5472]	9.1
Duerden 2012	0.18	[0.1212; 0.2388]	13.2
Handen 2018	0.22	[0.1808; 0.2592]	14
Maddox 2017	0.14	[0.0420; 0.2380]	11.2
Richards 2012	0.05	[0.0108; 0.0892]	14
Richards 2017 Child	0.06	[0.0208; 0.0992]	14
Richards 2017 Adult	0.03	[0.0104; 0.0496]	14.5
Slingsby 2017	0.17	[0.0524; 0.2876]	10.1

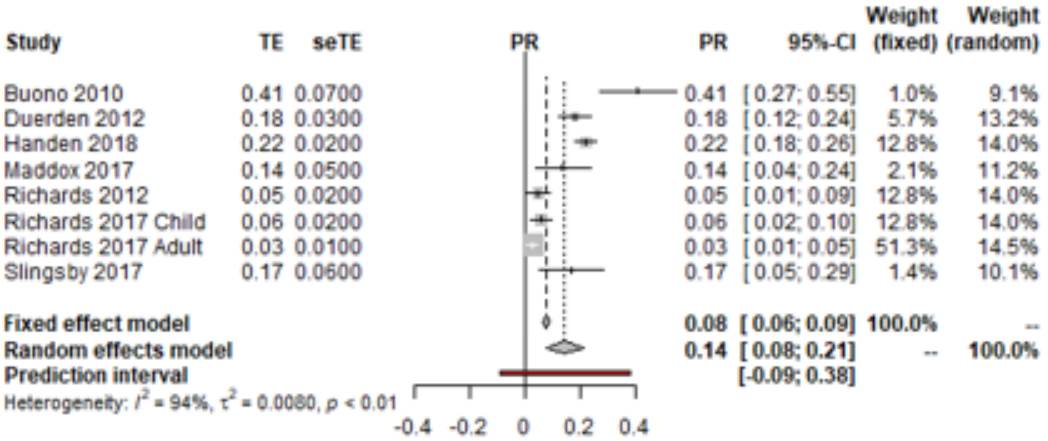

Figure 1.11. The pooled prevalence estimates for object hitting in ASD using the random-effects model.

Table 1.15. Reporting of the prevalence rates and confidence intervals for inserting objects in cavities.

Study Name	PR	95%-CI	Weighting in Random Effects Model
Buono 2010	0.35	[0.2128; 0.4872]	3.8
Duerden 2012	0.18	[0.1212; 0.2388]	9.3
Folch 2018	0.01	[-0.0096; 0.0296]	13.2
Handen 2018	0.11	[0.0708; 0.1492]	11.4
Hattier 2011	0.02	[0.0004; 0.0396]	13.2
Lecavalier 2006	0.11	[0.0708; 0.1492]	11.4
Richards 2012	0.03	[0.0104; 0.0496]	13.2
Richards 2017 Child	0.06	[0.0208; 0.0992]	11.4
Richards 2017 Adult	0.05	[0.0304; 0.0696]	13.2

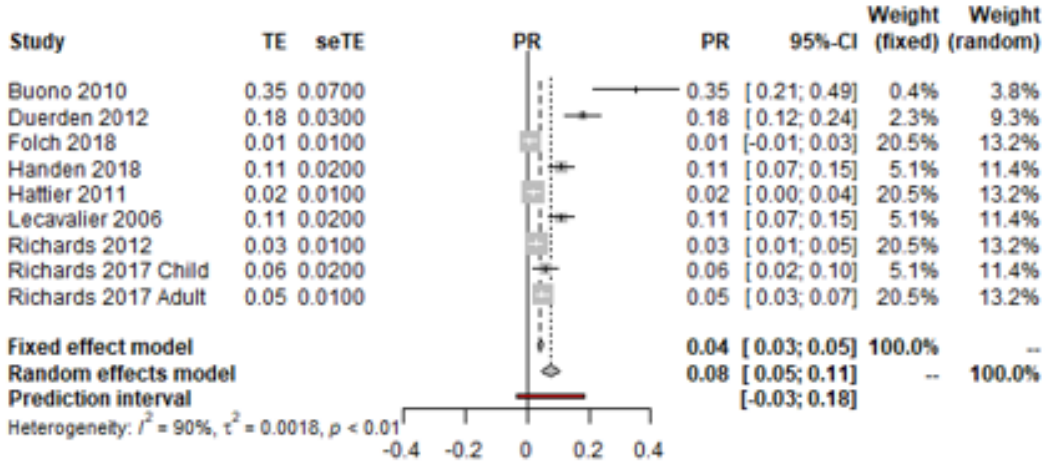

Figure 1.12. The pooled prevalence estimates for inserting objects in cavities in ASD using the random-effects model.

Table 1.16. Reporting of the prevalence rates and confidence intervals for rubbing self on surfaces.

Study Name	PR	95%-CI	Weighting in Random Effects Model
Akram 2017	0.0600	[0.0012; 0.1188]	73.5
Maddox 2017	0.01000	[0.0020; 0.1980]	26.5

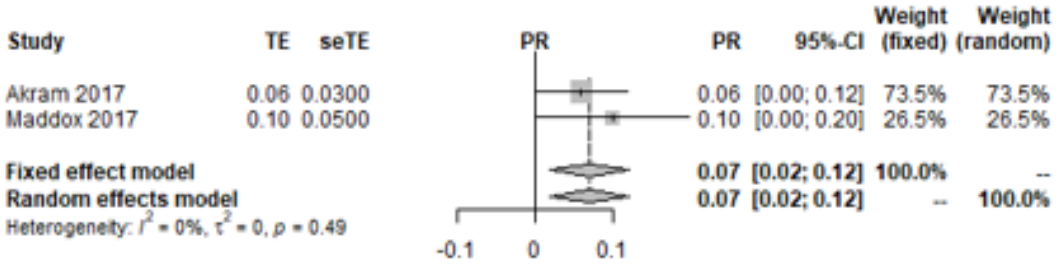

Figure 1.13. The pooled prevalence estimates for rubbing self on surfaces in ASD using the random-effects model.

Table 1.17. Reporting of the prevalence rates and confidence intervals for self-biting.

Study Name	PR	95%-CI	Weighting in Random Effects Model
Akram 2017	0.11	[0.0512; 0.1688]	8.5
Buono 2010	0.39	[0.2528; 0.5272]	5.6
Dominick 2007	0.09	[0.0116; 0.1684]	7.8
Duerden 2012	0.26	[0.2012; 0.3188]	8.5
Gulsrud 2018	0.03	[-0.0092; 0.0692]	9.1
Handen 2018	0.3	[0.2412; 0.3588]	8.5
Kamio 2002	0.02	[0.0004; 0.0396]	9.5
Lecavalier 2006	0.05	[0.0304; 0.0696]	9.5
Richards 2012	0.09	[0.0508; 0.1292]	9.1
Richards 2017 Child	0.17	[0.1112; 0.2288]	8.5
Richards 2017 Adult	0.16	[0.1208; 0.1992]	9.1
Slingsby 2017	0.17	[0.0524; 0.2876]	6.3

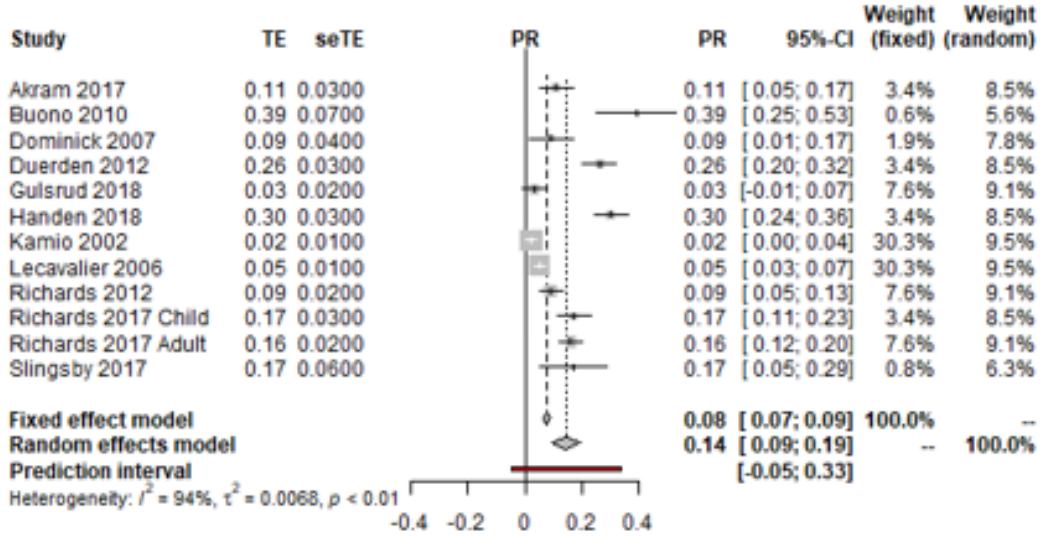

Figure 1.14. The pooled prevalence estimates for self-biting in ASD using the random-effects model.

Table 1.18. Reporting of the prevalence rates and confidence intervals for self-pinching.

Study Name	PR	95%-CI	Weighting in Random Effects Model
Akram 2017	0.1100	[0.0512; 0.1688]	15.7
Buono 2010	0.0600	[0.0012; 0.1188]	15.7
Hattier 2011	0.0600	[0.0404; 0.0796]	68.6

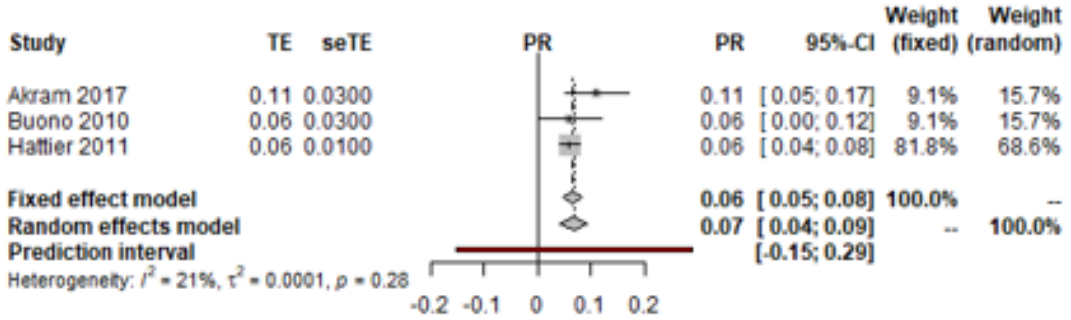

Figure 1.15. The pooled prevalence estimates for self-pinching in ASD using the random-effects model.

Table 1.19. Reporting of the prevalence rates and confidence intervals for self-scratching.

Study Name	PR	95%-CI %W	Weighting in Random Effects Model
Akram 2017	0.12	[0.0416; 0.1984]	6.9
Buono 2010	0.14	[0.0420; 0.2380]	6.2
Duerden 2012	0.23	[0.1712; 0.2888]	7.6
Folch 2018	0.15	[0.0912; 0.2088]	7.6
Gulsrud 2018	0.01	[-0.0096; 0.0296]	8.6
Handen 2018	0.43	[0.3712; 0.4888]	7.6
Hattier 2011	0.06	[0.0404; 0.0796]	8.6
Kamio 2002	0.04	[0.0204; 0.0596]	8.6
Lecavalier 2006	0.1	[0.0804; 0.1196]	8.6
Maddox 2017	0.36	[0.2228; 0.4972]	4.8
Richards 2012	0.09	[0.0508; 0.1292]	8.2
Richards 2017 Child	0.12	[0.0808; 0.1592]	8.2
Richards 2017 Adult	0.15	[0.1108; 0.1892]	8.2

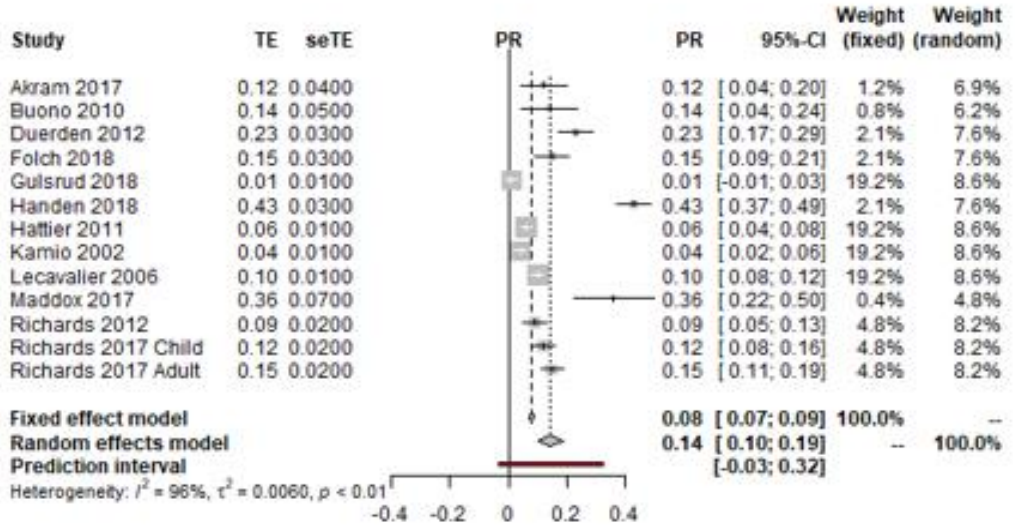

Figure 1.16. The pooled prevalence estimates for self-scratching in ASD using the random-effects model.

Table 1.20. Reporting of the prevalence rates and confidence intervals for wound interfering.

Study Name	PR	95%-CI %	Weighting in Random Effects Model
Akram 2017	0.11	[0.0512; 0.1688]	27.7
Folch 2018	0.08	[0.0408; 0.1192]	62.3
Maddox 2017	0.12	[0.0220; 0.2180]	10

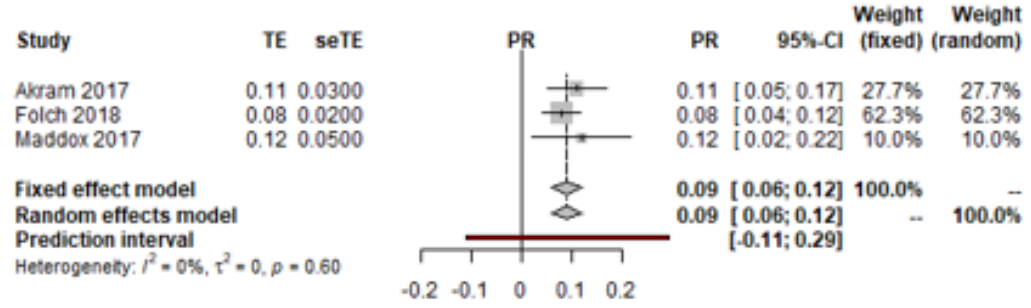

Figure 1.17. The pooled prevalence estimates for wound interfering in ASD using the random-effects model.

Table 1.21. Reporting of the prevalence rates and confidence intervals for self-cutting.

Study Name	PR	95%-CI %	Weighting in Random Effects Model
Akram 2017	0.01	[-0.0096; 0.0296]	24.4
Folch 2018	0.01	[-0.0096; 0.0296]	24.4
Maddox 2017	0.17	[0.0524; 0.2876]	2.4
Richards 2017 Child	0.04	[0.0204; 0.0596]	24.4
Richards 2017 Adult	0.03	[0.0104; 0.0496]	24.4

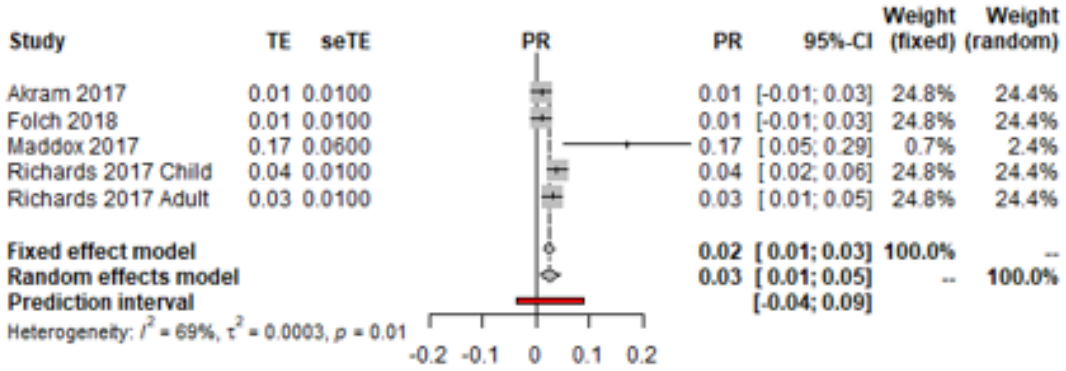

Figure 1.18. The pooled prevalence estimates for self-cutting in ASD using the random-effects model.

Table 1.22. Reporting of the prevalence rates and confidence intervals for skin picking.

Study Name	PR	95%-CI	Weighting in Random Effects Model
Duerden 2012	**0.25**	**[0.1912; 0.3088]**	**33.1**
Gulsrud 2018	**0.02**	**[0.0004; 0.0396]**	**33.8**
Handen 2018	**0.33**	**[0.2712; 0.3888]**	**33.1**

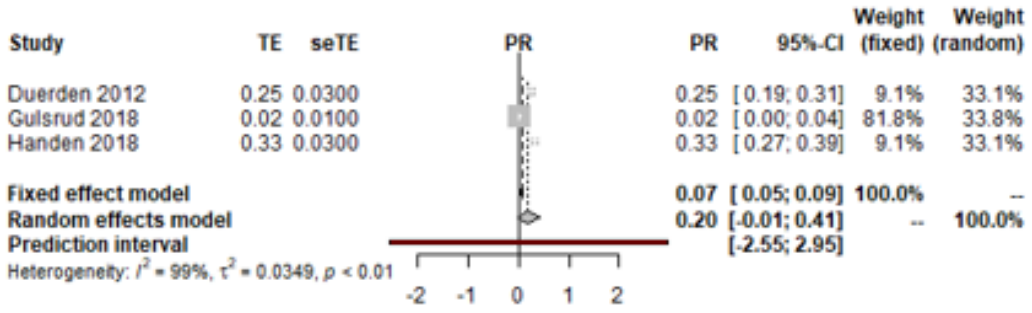

Figure 1.19. The pooled prevalence estimates for skin picking in ASD using the random-effects model.

APPENDIX C

Dr Caroline Richards: c.r.richards@bham.ac.uk, 0121 415 8098

Laura Groves: LXG502@student.bham.ac.uk, 0121 414 9775

Dear Parent/Carer,

We are writing to let you know about a new research project that will be taking place at the Cerebra Centre for Neurodevelopmental disorders at the University of Birmingham.

We have contacted you because your child attends a Special Needs School in the West Midlands**.** Your child's Head Teacher has sent out this information on our behalf. Your personal details will not be known to us unless you decide to take part in the research study and contact us directly.

The project is called 'Behaviours of children with neurodevelopmental difficulties' and aims to describe and assess different behaviours of children who are a delayed in some aspect of their development such as communication, social interaction or physical skills.

The study involves the completion of a brief 10-15 minute questionnaire, focussing on a wide spectrum of behaviours that your child may or may not demonstrate.

We have enclosed an information sheet that gives you more details about what participation in this research will involve, what will happen to the information you provide and the benefits of participating. We have also enclosed a consent form, the questionnaire and two pre-paid envelopes. If you would like to participate, we would very much appreciate it if you could complete the consent form and questionnaire and return them to the research team separately in the prepaid envelopes. This is to ensure your personal identifying information, cannot be linked to your questionnaire responses, without your personal identification number.

If you have any questions / concerns about the research, please do not hesitate to contact Laura Groves.

Due to our methods of recruitment, there is a small possibility you may have already been contacted by the Cerebra Centre to take part. If this is the case, please ignore the latest invitation to participate which has been sent to you.

This research project has been approved by the North East Tyne & Wear South Research Ethics Committee and has all the necessary approvals (Health Research Authority reference number: 235418; Protocol number: RG_17 182; REC number: 18/NE/0249).

Thank you very much for your time,

Kind regards,

Dr Caroline Richards, Dr Debbie Allen and Professor Chris Oliver

Behaviours of children with neurodevelopmental difficulties

Participant Information Sheet

We would like to invite you to take part in a research study being conducted at the Centre for Neurodevelopmental Disorders (CNDD) in the School of Psychology at the University of Birmingham.

Please read this information carefully before deciding whether you wish to take part in the research study. If you would like any more information about the study or you have any health and / or language difficulties which make it difficult for you to read this information please contact the research team or you can ask someone to contact the research team on your behalf (see details at the end of the information sheet).

This research project has been approved by the North East Tyne & Wear South Research Ethics Committee and has all the necessary approvals (Health Research Authority reference number: 235418; Protocol number: RG_17 182; REC number: 18/NE/0249).

What is the research study about and why is it important?

This research project, led by Professor Chris Oliver, Laura Groves and Dr Caroline Richards, is based in the Cerebra Centre for Neurodevelopmental Disorders (CNDD) in the School of Psychology at the University of Birmingham. The study aims to better understand the various behaviours of children with neurodevelopmental difficulties. We will use the information provided by parents to understand whether particular characteristics, such as age or gender, make certain behaviours more or less likely. We are also seeking to test whether the questionnaire used in this study could be useful in a clinical setting, as a screening tool.

In the future, the information obtained during this study could help us to identify ways in which we may be able to prevent the onset of particular behaviours that have a negative impact on the quality of life of children with neurodevelopmental difficulties.

Why have I been invited to take part in the study?

You have been invited to take part in this study because you are the parent / carer of a child aged between 2 and 11 years old who attend one of a range of educational and health facilities / services in the West Midlands specifically for children with neurodevelopmental difficulties. You may also have received this invitation if your child is undergoing an assessment of their development or you have previously provided consent for your details to be held on the CNDD research participant database and to be contacted about future research projects. Unless your details are already held on the CNDD participant database, we do not have your contact details and we do not know your name or your child's name. The health / education facility that your child attends has sent out this information on behalf of the research team so please be assured that we do not have access to any of your personal information.

If I decide to participate in the research study, what will I be asked to do?

If you would like to participate in the research, you will be asked to sign a consent form, and provide us with a few personal details (your name, postal address, telephone number, email address, and your child's gender and age) you will then be asked to complete a short questionnaire about your child's current behaviour. This questionnaire will take between 10-15 minutes to complete.

You will be asked to complete the questionnaire and return it to the research team in a pre-paid envelope along with a completed consent form. Alternatively, you can contact a member of the research team and arrange to have the questionnaire administered over the telephone or, if you prefer, they may be able to visit you at home if you live within a reasonable travelling distance.

You will be contacted by the research team again 12 months later (Time 2) to complete the same questionnaire that you did at Time 1. This is to help the study team understand how behaviour changes over time. The research team will post the questionnaire to your home address with a pre-paid envelope for you to return the completed questionnaire and / or telephone you to administer the questionnaire on the telephone. Once again, a member of the research team may also be able to visit you at home

Who will be involved in collecting the information?

Members of the research team at the Cerebra Centre for Neurodevelopmental Disorders based at the University of Birmingham including experienced academic research staff and supervised

undergraduate and postgraduate students. The research team will also include NHS Research Nurses.

What are the possible benefits of taking part in this study?

After you have completed the questionnaire, you will receive a summary report specific to your child based on your responses to the questionnaire and the opportunity to ask any questions. You will receive a similar report 12 month later, when you have completed the questionnaire for the second time.

These individualised feedback reports may be useful for you and the health and education professionals involved with your child / the child you care for to highlight particular difficulties that your child might face and identify resources that might be useful for them. We happily share a copy of this report with any health / education professionals if you request this and provide us with written consent.

What are the possible disadvantages of taking part in this study?

While the questionnaire is very brief, it includes questions about challenging behaviour which you might find upsetting. If this does happen and you feel that you do not want to continue with participation in the research, you can withdraw at any time. You will also be given the opportunity to contact the research team and discuss any concerns you have about the project at any stage during the 12-month research period who may be able to signpost you to helpful resources or professionals.

If I decide to participate, what will happen to the information I provide?

Personal identifying information, such as names, ages, addresses, telephone numbers and email addresses will be treated as strictly confidential and handled in accordance with the provisions of the Data Protection Act 1998 & 2018. Personal identifying information will be stored for the duration of the study on a password protected portable hard drive that is kept in a locked filing cabinet at the University of Birmingham.

Your completed questionnaires will be stored separately from the personal identifying information described above in a locked filing cabinet at the University of Birmingham. Your name will not appear on the completed questionnaires. Instead, each participant will be allocated a participant number and this will appear on the questionnaires instead of names. An electronic file will be created which links the participant number to the participant name which will be stored on a password protected portable hard drive kept in a locked filing cabinet at the University of Birmingham. Only the research team have access to the research project filing cabinets.

In the unlikely event that the research team have concerns about the welfare of a participant, information will be disclosed as necessary.

What would happen to the information after the project ends?

If you are not already on the existing participant database, your contact details will be destroyed at the end of the study (within 6 months of receipt of your Time 2 questionnaire or 6 months after the end of the Time data collection cut-off date) and you will not be contacted further unless your details are already stored on the CNDD database. Your anonymous completed questionnaires will be stored for up to 10 years following the end of the project.

The research team will publish the findings from the study in scientific journals and will present the results at relevant conferences and in newsletters. Any published reports which use the information you have provided us, will be completely confidential, and will never use your child's name.

If I would like to participate in the project, what should I do now?

Please remember that participation in the project is purely optional and the decision not to participate will not restrict access or affect the right to any education / health services. When you are satisfied that you have all of the information you need to be able to decide whether or not you / the person you care for would like to take part in the study and if you decide that you would like to participate, please complete the enclosed consent form and the questionnaire and return them to us separately in the two prepaid envelope provided. This is to ensure your responses and personal details cannot be matched except by the identified number on the envelopes.

What if I change my mind about participating after I have provided consent?

Even after you have provided consent to participate in the study, you can request to be withdrawn from the study and for your research data to be destroyed without giving a reason. This will not restrict access or affect the right to any education / health services. You will have up to 6 months after completion of the Time 2 questionnaire to indicate that you would like to withdraw from the study. This is purely for practical reasons as after your personal identifying information has been destroyed, your personal details will no longer be linked to the information collected as part of this study. This means that we would no longer be able to trace the results of your assessments back to you and withdraw you from the study.

What can I do if I have any concerns about the research or there is a problem?

In the unlikely event that you have any cause for concern about any aspect of the research, in the first instance, please contact Professor Chris Oliver (Chief Investigator) on 0121 414 4909, c.oliver@bham.ac.uk, Laura Groves (Research Assistant) on 0121 414 9775, LXG502@student.bham.ac.uk, or Dr Caroline Richards (Principal Investigator) on 0121 415 8098, c.r.richards@bham.ac.uk at the University of Birmingham. If you wish to speak to someone who is independent to the research, or you have any concerns related to the research after

contacting the Chief Investigator, Research Assistant or Principal Investigator, you can contact Birmingham Community Healthcare Foundation Trust team on:
Tel - 0800 917 2855
E-mail - complaints.bchc@nhs.net
Write to - Complaints Team, 3 Priestley Wharf, Holt Street, Aston, Birmingham, B7 4BN.

How do I contact the research team to find out more about the research study?
Please contact Laura Groves on:
Tel - 0121 414 9775
E- mail - LXG502@student.bham.ac.uk
Write to - Laura Groves, School of Psychology, 52 Pritchatts Road, University of Birmingham, Edgbaston, Birmingham, B15 2TT

Or Dr Caroline Richards on:
Tel - 0121 415 8098
E- mail - c.r.richards@bham.ac.uk
Write to - Caroline Richards, School of Psychology, 52 Pritchatts Road, University of Birmingham, Edgbaston, Birmingham, B15 2TT

Alternatively, you can contact Birmingham Community Healthcare Foundation Trust Customers Services Team for independent advice about the study on:
Tel - 0800 917 2855
E- mail - contact.bchc@nhs.net
Write to - Customer Services Team, The Lodge, Moseley Hall Hospital, Alcester Road, Moseley, Birmingham, B13 8JL.

The University of Birmingham is the sponsor for this study based in the United Kingdom. We will be using information from you in order to undertake this study and will act as the data controller for this study. This means that we are responsible for looking after your information and using it properly. The University of Birmingham will keep identifiable information about you for 6 months following receipt of your Time 2 questionnaire to enable you to receive your Time 2 feedback report. If the Time 2 questionnaire isn't completed, any identifiable data will be destroyed 6 months after the data collection window.

Your rights to access, change or move your information are limited, as we need to manage information in specific ways in order for research to be reliable and accurate. If you withdraw from the study, we will keep the information we have already obtained. To safeguard your rights, we will use minimum personally-identifiable information possible.

You can find out more information about how we use your information by contacting the Data Protection Officer at the University, Mrs Carolyn Pike (OBE)
legalservices@contacts.bham.ac.uk

The University of Birmingham will use your name, and contact details (and your child's age and gender) to contact you about the research study and to oversee the quality of the study. Individuals from the University of Birmingham and regulatory organisations may look at your research records check the accuracy of the research study. The only people in the University of Birmingham who will have access to the information that identifies you will be the people who need to contact you about your participation in the study or audit the data collection process.

Health and care research should serve the public interest, which means that we have to demonstrate that our research serves the interests of society as a whole. We do this by following the UK Policy Framework for Health and Social Care Research.

If you wish to raise a complaint on how we have handled your personal data, you can contact our Data Protection Officer who will investigate the matter. Our Data Protection Officer is Mrs. Carolyn Pike OBE and you can contact them at legalservices@contacts.bham.ac.uk If you are not satisfied with our response or believe we are processing your personal data in a way that is not lawful you can complain to the Information Commissioner's Office (ICO).

Thank you for your time.

Behaviours of Children with Neurodevelopmental Difficulties

Parental Consent Form

If you are reading this information on behalf of someone you care for who is a child ages 2-11, then we would like to ask you to decide whether or not you think that it is in your child's best interests for them to participate in the study and whether you would like to provide your consent to participation on their behalf. If you would like your child/person you care for to participate in this study, please complete the consent form and return it to the research team in the prepaid envelope provided.

PART 1 **Please initial box…**

1. I confirm that I have read and understood the information sheet dated V5 18.09.18 for the above study. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily. ☐

2. I understand that participation involves completion of 1 short questionnaire now (Time 1) and that I will be contacted again in 12 months (Time 2) to complete the same questionnaire again, using the contact details I provide. ☐

3. I understand that my participation and that of my child/person I care for is voluntary and that I am free to withdraw at any time without giving any reason, without my or that of my child's/person I care for's medical care or legal rights being affected. ☐

4. I understand that all information collected during the study will be confidential. Only members of the research team at the Cerebra Centre for Neurodevelopmental disorders will know who has participated in the study. All information collected during the study will be stored in locked cabinets ☐

and / or on password protected portable hard drives that only members of the research team will have access to. No names will be published in any reports. Information will be treated as strictly confidential and handled in accordance with the provisions of the Data Protection Act 2018.

5. I understand that my contact details will only be used by the research team for the purpose of this study only. ☐

6. I agree, on behalf of myself and the child I care for, to take part in the study 'Behaviours of Children with a Neurodevelopmental Difficulties'. ☐

Please complete the details below

Name of Parent / Carer: _____

Your relationship to the participant: _____

Signed: _____ Date: _____

Researcher name (if present)_____ Countersigned:_____

PART 2

Child's gender: Male Female
Age of Child: _____years _____months.

Address: _____

Landline Telephone Number: _____

Mobile Telephone Number: _____

Email Address: _____

APPENDIX D

Professor Chris Oliver
Professor of Neurodevelopmental Disorders / Director of the Cerebra Centre for Neurodevelopmental Disorders

University of Birmingham

School of Psychology

University of Birmingham

B15 2TT

Email: hra.approval@nhs.net
Research-permissions@wales.nhs.uk

15 August 2018

Dear Professor Oliver

HRA and Health and Care

Study title:	**The Identification of young children at highest RIsk for developing Severe Challenging behaviour (i-RISC): Proof of principle and appraisal of feasibility.**
IRAS project ID:	**235418**
Protocol number:	**RG_17-182**
REC reference:	**18/NE/0249**
Sponsor	**University of Birmingham**

I am pleased to confirm that **HRA and Health and Care Research Wales (HCRW) Approval** has been given for the above referenced study, on the basis described in the application form, protocol, supporting documentation and any clarifications received. You should not expect to receive anything further relating to this application.

How should I continue to work with participating NHS organisations in England and Wales? You should now provide a copy of this letter to all participating NHS organisations in England and Wales, as well as any documentation that has been updated as a result of the assessment.

Following the arranging of capacity and capability, participating NHS organisations in England and Wales that are hosting all site activities should **formally confirm** their capacity and capability to undertake the study. How this will be confirmed is detailed in the "*summary of assessment*" section towards the end of this letter. You should then work with each organisation that has confirmed capacity and capability and provide clear instructions when research activities can commence.

Participating NHS organisations in England and Wales that are acting as participant identification centres **will not** be required to formally confirm capacity and capability before you may commence research activity at site. As such, you may commence the research at each organisation 35 days following sponsor provision to the site of the local information pack, so long as:

- You have contacted participating NHS organisations (see below for details)
- The NHS organisation has not provided a reason as to why they cannot participate
- The NHS organisation has not requested additional time to confirm.

You may start the research prior to the above deadline if the site positively confirms that the research may proceed.

If not already done so, you should now provide the local information pack for your study to your participating NHS organisations. A current list of R&D contacts is accessible at the NHS RD Forum website and these contacts MUST be used for this purpose. After entering your IRAS ID you will be able to access a password protected document (password: **White22**). The password is updated on a monthly basis so please obtain the relevant contact information as soon as possible; please do not hesitate to contact me should you encounter any issues.

Commencing research activities at any NHS organisation before providing them with the full local information pack and allowing them the agreed duration to opt-out, or to request additional time (unless you have received from their R&D department notification that you may commence), is a breach of the terms of HRA and HCRW Approval. Further information is provided in the "*summary of assessment*" section towards the end of this document.

It is important that you involve both the research management function (e.g. R&D office) supporting each organisation and the local research team (where there is one) in setting up your study. Contact details of the research management function for each organisation can be accessed here.

How should I work with participating NHS/HSC organisations in Northern Ireland and Scotland?

HRA and HCRW Approval does not apply to NHS/HSC organisations within the devolved administrations of Northern Ireland and Scotland.

If you indicated in your IRAS form that you do have participating organisations in either of these devolved administrations, the final document set and the study wide governance report (including this letter) has been sent to the coordinating centre of each participating nation. You should work with the relevant national coordinating functions to ensure any nation specific checks are complete, and with each site so that they are able to give management permission for the study to begin.

Please see IRAS Help for information on working with NHS/HSC organisations in Northern Ireland and Scotland.

How should I work with participating non-NHS organisations?

HRA and HCRW Approval does not apply to non-NHS organisations. You should work with your nonNHS organisations to obtain local agreement in accordance with their procedures.

What are my notification responsibilities during the study?

The document "*After Ethical Review – guidance for sponsors and investigators*", issued with your REC favourable opinion, gives detailed guidance on reporting expectations for studies, including: Registration of research

- Notifying amendments
- Notifying the end of the study

The HRA website also provides guidance on these topics, and is updated in the light of changes in reporting expectations or procedures.

I am a participating NHS organisation in England or Wales. What should I do once I receive this letter?

You should work with the applicant and sponsor to complete any outstanding arrangements so you are able to confirm capacity and capability in line with the information provided in this letter.

The sponsor contact for this application is as follows:

Name: Dr Sean Jennings

Tel: 01214158011

Email: researchgovernance@contacts.bham.ac.uk

Who should I contact for further information?

Please do not hesitate to contact me for assistance with this application. My contact details are below.

Your IRAS project ID is **235418**. Please quote this on all correspondence.

Yours sincerely

Helen Penistone
Assessor

Email: hra.approval@nhs.net

Copy to: *Dr Sean Jennings (sponsor contact)*
Mrs Priti Parmar, Research and Innovation, Birmingham Community Healthcare NHS Foundation Trust, (lead NHS R&D contact)

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Summary of assessment

The following information provides assurance to you, the sponsor and the NHS in England and Wales that the study, as assessed for HRA and HCRW Approval, is compliant with relevant standards. It also provides information and clarification, where appropriate, to participating NHS organisations in England and Wales to assist in assessing, arranging and confirming capacity and capability.

Assessment criteria

Section	Assessment Criteria	Compliant with Standards	Comments
1.1	IRAS application completed correctly	Yes	No comments
2.1	Participant information/consent documents and consent process	Yes	Non-substantial amendments have been made to the Participant Information Sheet and Consent Form following REC favourable opinion. This was primarily to ensure compliance with the GDPR.
3.1	Protocol assessment	Yes	No comments

4.1	Allocation of responsibilities and rights are agreed and documented	Yes	The sponsor intends to use the Statement of Activities to form an agreement with the participating NHS organisation hosting all site activities. No additional agreement is expected. Although formal confirmation of capacity and capability is not expected of all or some organisations participating in this study, and such organisations would therefore be assumed to have confirmed their capacity and capability should they not respond to the contrary, we would ask that these organisations pro-actively engage with the sponsor in order to confirm at as early a date as possible. Confirmation in such cases should be by email to the CI and Sponsor confirming participation based on the relevant Statement of Activities and
Section	**Assessment Criteria**	**Compliant with Standards**	**Comments**
			information within this letter.
4.2	Insurance/indemnity arrangements assessed	Yes	No comments
4.3	Financial arrangements assessed	Yes	Funding to support the study has been granted by the British Academy of Childhood Disability - Castang Foundation. The funder has granted a no-cost extension. As per the Statement of Activities, there will be no funding available to either site type. The research team will provide paper copies of all study documents and prepaid addressed envelopes.

5.1	Compliance with the Data Protection Act and data security issues assessed	Yes	No comments
5.2	CTIMPS – Arrangements for compliance with the Clinical Trials Regulations assessed	Not Applicable	No comments
5.3	Compliance with any applicable laws or regulations	Yes	No comments
6.1	NHS Research Ethics Committee favourable opinion received for applicable studies	Yes	No comments
6.2	CTIMPS – Clinical Trials Authorisation (CTA) letter received	Not Applicable	No comments
6.3	Devices – MHRA notice of no objection received	Not Applicable	No comments
6.4	Other regulatory approvals and authorisations received	Not Applicable	No comments

Participating NHS Organisations in England and Wales

This provides detail on the types of participating NHS organisations in the study and a statement as to whether the activities at all organisations are the same or different.

There will be two NHS study site types:

1. The site acting as a participant identification centre.
2. The site where participants will be recruited. Participants may also complete a questionnaire at site if they choose to.

The Chief Investigator or sponsor should share relevant study documents with participating NHS organisations in England and Wales in order to put arrangements in place to deliver the study. The documents should be sent to both the local study team, where applicable, and the office providing the research management function at the participating organisation. Where applicable, the local LCRN contact should also be copied into this correspondence.

If chief investigators, sponsors or principal investigators are asked to complete site level forms for participating NHS organisations in England and Wales which are not provided in IRAS, the HRA or HCRW websites, the chief investigator, sponsor or principal investigator should notify the HRA immediately at hra.approval@nhs.net or HCRW at Research-permissions@wales.nhs.uk. We will work with these organisations to achieve a consistent approach to information provision.

Principal Investigator Suitability

This confirms whether the sponsor position on whether a PI, LC or neither should be in place is correct for each type of participating NHS organisation in England and Wales, and the minimum expectations for education, training and experience that PIs should meet (where applicable).
The chief investigator for the study will be responsible for the research at the participant identification centre. At sites hosting all site activities it is expected that there will be a local collaborator to facilitate the access of externally employed researchers. All local staff at sites involved in the recruitment process will be trained on the protocol specific recruitment and consenting procedures. The sponsor expects that all staff at sites are familiar with the standard procedures for recruiting patients from the NHS. GCP training is not a generic training expectation, in line with the HRA/HCRW/MHRA statement on training expectations.

HR Good Practice Resource Pack Expectations

This confirms the HR Good Practice Resource Pack expectations for the study and the pre-engagement checks that should and should not be undertaken

It is not expected that externally employed researchers will need access to the NHS organisation acting as a PIC.

No Honorary Research Contracts, Letters of Access or pre-engagement checks are expected for local staff employed by the participating NHS organisation. Where arrangements are not already in

place, research staff not employed by the NHS host organisation undertaking any of the research activities listed in the research application would be expected to obtain a Letter of Access based on an enhanced DBS checks and occupational health clearance. No barred list check will be necessary.

Other Information to Aid Study Set-up

This details any other information that may be helpful to sponsors and participating NHS organisations in England and Wales to aid study set-up.
The applicant has indicated that they intend to apply for inclusion on the NIHR CRN Portfolio.

APPENDIX E

Child Health & Behaviour Screen

Behaviours of Children with Neurodevelopmental Difficulties

Dr Caroline Richards: c.r.richards@bham.ac.uk, 0121 415 8098
Laura Groves: LXG502@student.bham.ac.uk, 0121 414 977

Today's date:..... **Participant ID Number**:……..……..

Child's gender: **Male** □ **Female** □..... Child's Age:_____(Years)_____(Months)

Name and address of child's G.P.:

.....

1. **Has any professional (e.g. doctor, clinical geneticist, paediatrician etc.) said that the child:**
   Is autistic, has an autistic spectrum disorder, autistic like traits and/or features of autism
   .....
   Yes/No .....
   Has cerebral palsy or muscular dystrophy
   ..... …….....Yes/No
   Has a genetic syndrome (e.g. Downs Syndrome, Fragile X, Cri du Chat etc.)
   …….....Yes/No
   If you answered **yes** to genetic syndrome, please state which:
   ……...…….
   If you answered **no** to genetic syndrome, please state the cause of the child's learning disability (if known):
   ……
   ……

2. **Please name any prescribed medications the child is currently taking whilst at school:**
   .....

   .....

3. **Please give details of any contact/visits the child has had with any health professionals (e.g. dentists, schools nurses, psychologists etc.) in the last month regarding the child's:**

Health:.....
……
Behaviour:
……..
……..

4. Have you ever thought about seeking help regarding the child's behaviour?

Yes and have made contact □ Yes but not made contact □ No □

5. Please indicate the number of days that the child did not attend school in the last full term:

0-4 □ 5-9 □ 10-14 □ 15+ □

6. Please circle the appropriate response regarding the child's general development

Smile spontaneously	Yes/No	Turn to a voice	Yes/No
Feed self	Yes/No	Imitate speech sounds	Yes/No
Wave bye-bye	Yes/No	Say 2 words	Yes/No
Use spoon/fork	Yes/No	Point to pictures	Yes/No
Put on clothing	Yes/No	Name 1 colour	Yes/No
Grasp rattle	Yes/No	Roll over	Yes/No
Thumb and finger grasp	Yes/No	Stand holding on	Yes/No
Scribbles	Yes/No	Stand alone	Yes/No
Build a tower of two cubes	Yes/No	Run	Yes/No
Imitate drawing a vertical line	Yes/No	Jump up	Yes/No

7. To what extent have the following health problems affected the child in the last month?

	Never	Mild	Mod	Sev
Eye problems (e.g. infections)	0	1	2	3
Ear problems (e.g. infections)	0	1	2	3
Dental problems (e.g. cavities/gum problems)	0	1	2	3
Digestive problems (e.g. reflux/stomach problems)	0	1	2	3
Skin problems (e.g. eczema/dry skin)	0	1	2	3
Respiratory problems (e.g. asthma/infections)	0	1	2	3
Epilepsy	0	1	2	3
Any other health or painful condition, (please specify)	0	1	2	3

Q8. Please circle the appropriate response to indicate whether your child is able to carry out each skill.

Is your child able to request Food? Yes / No
Toys? Yes / No
Activities? Yes / No
Is your child able to request help/ assistance? Yes / No
Is your child able to request to take a break? Yes / No
Is your child able to reject items/ activities? Yes / No
Is your child able to communicate Yes? Yes / No
No? Yes / No
Is your child able to respond appropriately to being asked to "wait"? Yes / No
Is your child able to respond to the following **Visual** Directions (gestures/ signs)?
Respond to their name being signalled….. Yes / No
"Come here"…….. Yes / No
"Stop"……. Yes / No
"Sit down"……. Yes / No
"Give it to me"……..…… Yes / No
"Go get…" (Familiar item)……..…… Yes / No
"Go to…" (Familiar location)…….. Yes / No
"Put it back/down" …… Yes / No
"Let's go/ come with me"……...Yes / No
Is your child able to respond to the following **Verbal** Directions (spoken)?

To their name being called…….. Yes / No
"Come here"……..… Yes / No
"Stop"……..... Yes / No
"Sit down"…….. Yes / No
"Give it to me"…….. Yes / No
"Go get…" (Familiar item)…… Yes / No
"Go to…" (Familiar location)…….. Yes / No
"Put it back/ down"……... Yes / No
"Let's go/ come with me"……. Yes / No

Is your child able to react appropriately to changing activities?..... Yes / No

Is your child able to follow a visual schedule?..... . Yes / No

Does the child you care for	How frequently does this problem occur from never (0) to very often (4)?					How difficult is it to manage this problem from not difficult (0) to extremely difficult (4)?					How concerned are you about this problem from not at all concerned (0) to extremely concerned (4)?				
Show repetitive movements?	0	1	2	3	4	0	1	2	3	4	0	1	2	3	4
Show obsessions and rituals?	0	1	2	3	4	0	1	2	3	4	0	1	2	3	4
Show aggression (e.g. punching, pushing, kicking, pulling hair, grabbing other's clothing etc.?)	0	1	2	3	4	0	1	2	3	4	0	1	2	3	4
Show destruction of property (e.g. tearing or chewing own clothing, tearing newspapers, breaking windows or furniture, slamming doors, spoiling a meal etc.?)	0	1	2	3	4	0	1	2	3	4	0	1	2	3	4
Show self injury (e.g. head banging, head-punching or slapping, removing hair, self-scratching, body hitting, eye poking or pressing etc.?)	0	1	2	3	4	0	1	2	3	4	0	1	2	3	4
Show emotional outbursts (e.g. excessive emotions including anger or distress etc.?)	0	1	2	3	4	0	1	2	3	4	0	1	2	3	4
Find it difficult to wait?	0	1	2	3	4	0	1	2	3	4	0	1	2	3	4
Act as if driven by a motor?	0	1	2	3	4	0	1	2	3	4	0	1	2	3	4
Want things immediately?	0	1	2	3	4	0	1	2	3	4	0	1	2	3	4
Find it difficult holding still?	0	1	2	3	4	0	1	2	3	4	0	1	2	3	4

APPENDIX F

Child Health & Behaviour Screen

Behaviours of Children with Neurodevelopmental Difficulties

Dr Caroline Richards: c.r.richards@bham.ac.uk, 0121 415 8098
Laura Groves: LXG502@student.bham.ac.uk, 0121 414 977

Today's date:..... **Participant ID Number**:……..

Child's gender: **Male** □ **Female** □..... Child's Age: _____(Years)_____(Months)

Q1. Has any professional (e.g. doctor, geneticist, paediatrician etc.) said that the child you care for: (Please circle your response)

Is autistic, has an autistic spectrum disorder, autistic like traits and/or features of autism?

.....

Yes / No .....

Has cerebral palsy or muscular dystrophy? …….....

Yes / No

Has a genetic syndrome (e.g. Downs Syndrome, Fragile X, Cri du Chat etc.)?…….....Yes / No

If you answered **Yes** to genetic syndrome, please state which:

……..…….

If you answered **No** to genetic syndrome, please state the cause of the child's learning disability (if known):

……

Q2. Please name any prescribed medications the child is currently taking whilst at school:

.....

.....

Q3. Please give details of any contact/visits the child has had with any health professionals (e.g. dentists, schools nurses, psychologists etc.) in the last month regarding the child's:

Health:.....

……

Behaviour:

……..

……

Q4. Have you ever thought about seeking help with the child's behaviour? (Please circle your response)

Yes and have made contact □ Yes but not made contact □ No □

Q5. Please indicate the number of days that the child you care for did not attend school in the last full term. (Please circle your response)

0-4 □ 5-9 □ 10-14 □ 15+ □

Q6. Please circle the appropriate response regarding the child's general development

Walk without help elsewhere	1…Not at all	2…Not upstairs	3…Upstairs and
Feed self	1…Not at all	2…With help	3…Without help
Wash self	1…Not at all	2…With help	3…Without help
Dress self	1…Not at all	2…With help	3…Without help
Wetting (days)	1…Frequently	2…Occasionally	3…Never
Soiling (days)	1…Frequently	2…Occasionally	3…Never
Reads books	1…Nothing	2…A little	3…Newspapers and/or
Writes correspondence	1…Nothing	2…A little	3…Own
Counts values	1…Nothing	2…A little	3…Understands money
Speech normal	1…Never a word	2…Odd words only	3…Sentences and
Vision	1…Blind or almost	2…Poor	3…Normal
Hearing	1…Deaf or almost	2…Poor	3…Normal

Q7. To what extent have the following health problems affected the child in the last month? (Please circle your response)

	Never	Mild	Mod	Sev
Eye problems (e.g. infections)	0	1	2	3
Ear problems (e.g. infections).....	0	1	2	3
Dental problems (e.g. cavities/gum problems)	0	1	2	3
Digestive problems (e.g. reflux/stomach problems)	0	1	2	3
Skin problems (e.g. eczema/dry skin).....	0	1	2	3
Respiratory problems (e.g. asthma/infections)	0	1	2	3
Epilepsy	0	1	2	3
Any other health or painful condition, (please specify)	0	1	2	3

Q8. Please circle the appropriate response to indicate whether your child is able to carry out each skill.

Is your child able to request Food?..... Yes / No
Toys?..... Yes / No
Activities?..... Yes / No
Is your child able to request help/ assistance?..... Yes / No
Is your child able to request to take a break?..... Yes / No
Is your child able to reject items/ activities?..... Yes / No
Is your child able to communicate Yes?..... Yes / No
No?... ..... Yes / No
Is your child able to respond appropriately to being asked to "wait"?..... Yes / No
Is your child able to respond to the following **Visual** Directions (gestures/ signs)?
Respond to their name being signalled…... Yes / No
"Come here"…….. Yes / No
"Stop"……. Yes / No

"Sit down"……. Yes / No
"Give it to me"……...…… Yes / No
"Go get…" (Familiar item)……...…… Yes / No
"Go to…" (Familiar location)……... Yes / No
"Put it back/down" …… Yes / No
"Let's go/ come with me"……... Yes /No

Is your child able to respond to the following **Verbal** Directions (spoken)?

To their name being called…….. Yes / No
"Come here"……..… Yes / No
"Stop"……..... Yes / No
"Sit down"…….. Yes / No
"Give it to me"…….. Yes / No
"Go get…" (Familiar item)…… Yes / No
"Go to…" (Familiar location)…….. Yes / No
"Put it back/ down"……... Yes / No
"Let's go/ come with me"……. Yes / No

Is your child able to react appropriately to changing activities?..... Yes / No

Is your child able to follow a visual schedule?..... . Yes / No

oes the child you care for:	How frequently does this problem occur from never (0) to very often (4)?					How difficult is it to manage this problem from not difficult (0) to extremely difficult (4)?					How concerned are you about this problem from not at all concerned (0) to extremely concerned (4)?				
Show repetitive movements?	0	1	2	3	4	0	1	2	3	4	0	1	2	3	4
Show obsessions and rituals?	0	1	2	3	4	0	1	2	3	4	0	1	2	3	4
Show aggression (e.g. punching, pushing, kicking, pulling hair, grabbing other's clothing etc.?)	0	1	2	3	4	0	1	2	3	4	0	1	2	3	4
Show destruction of property (e.g. tearing or chewing own clothing, tearing newspapers, breaking windows or furniture, slamming doors, spoiling a meal etc.?)	0	1	2	3	4	0	1	2	3	4	0	1	2	3	4
Show self injury (e.g. head banging, head-punching or slapping, removing hair, self-scratching, body hitting, eye poking or pressing etc.?)	0	1	2	3	4	0	1	2	3	4	0	1	2	3	4
Show emotional outbursts (e.g. excessive emotions including anger or distress etc.?)	0	1	2	3	4	0	1	2	3	4	0	1	2	3	4
Find it difficult to wait?	0	1	2	3	4	0	1	2	3	4	0	1	2	3	4
Act as if driven by a motor?	0	1	2	3	4	0	1	2	3	4	0	1	2	3	4
Want things immediately?	0	1	2	3	4	0	1	2	3	4	0	1	2	3	4
Find it difficult holding still?	0	1	2	3	4	0	1	2	3	4	0	1	2	3	4