Psychosocial and Behavioural Factors Associated with Self Injurious Behaviour (SIB) in Individuals with Autism Spectrum Disorders (ASD).

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Abstract

Background: Self-injurious behaviour (SIB) is a persistent and distressing difficulty which may be more prevalent and enduring for individuals with an Autism Spectrum Disorder (ASD). SIB has been largely conceptualised in research as a challenging behaviour or a repetitive and restricted behaviour, rather than a unique construct to research. As its own construct, the aetiology of SIB has been conceptualised from a neurobiological perspective, however there remains a need to explore psychosocial and behavioural factors associated with SIB and ASD. A review was conducted to compile evidence and establish current understanding of this behaviour.

Method: 6 databases were systematically searched for research exploring factors relating specifically to SIB limited to ASD populations. Studies were critically appraised using a tool developed for the purpose of this review, adapted from the CASP, AXIS and STROBE quality appraisal tools.

Results: 15 studies met the eligibility criteria. SIB was found to be associated with impairments in adaptive ability, communicative ability, IQ, sleep, atypical sensory processing, and impulsivity/ over-activity. There were mixed findings supporting an association between autism severity and self-injury.

Conclusions: The development of SIB in ASD populations is complex. The range of factors associated with SIB and ASD imply a clinical need for a robust assessment and a multi-disciplinary approach to intervention. Theoretical perspectives regarding the role of impaired behavioural inhibition, communication, and sensory processing difficulties are considered. Limitations and future research are discussed.

Keywords: Autism, ASD, Self-injurious Behaviour, Associations, Review

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Introduction

Self-injurious behaviour (SIB) refers to self-directed behaviours which result in physical harm to the individual without showing apparent intent of harm (Fee & Matson, 1992). Such behaviours include head banging, biting, hitting, and eye gouging. SIB can range from 'mild' to 'severe' and as such are concerning to those who work with the individuals presenting with these behaviours (Durand & Crimmins, 1988). SIB is observed across different groups of individuals at different points in life. Self-directed injurious behaviours and repetitive behaviours, such as head-banging and rocking, can occur in typically developing populations as part of normal development (Berkson & Tupa, 2000; Berkson, Tupa & Sherman, 2001), however these behaviours usually diminish before 3 years old. Self-directed 'proto-injurious' behaviours can also occur from birth in those with developmental difficulties/disorders and differ from SIB as they do not yet cause tissue damage (Roane et al, 2007; Tate & Baroff, 1966). Self-injurious behaviours can also present in individuals with mental health difficulties such as Borderline Personality Disorder (Crowell & Kaufman, 2016), although these behaviours are usually episodic and individuals hold intent to cause themselves harm. The aetiology of SIB is yet to be fully comprehended, although it is understood that self-injury may be underpinned by neurobiological processes (such as in individuals with Lesch-Nyhan Syndrome, Smith-Magenis Syndrome, see Furniss & Biswas, 2020) and is developed and maintained by social and non-social reinforcement and resulting influences in the social and physical environment (Carr, 1977; Guess & Carr, 1991; Iwata et al., 1994; Hastings & Brown, 2000; Kurtz et al 2003). However, in some disorders, such as Autism Spectrum Disorder, some individuals engage with SIB, while others do not. This indicates that other psychosocial and behavioural factors are significant to consider in the emergence of SIB.

Autism Spectrum Disorder (ASD) is a pervasive developmental disorder which is characterised by differences in social interaction, communication, and restricted and repetitive behaviours or interests. ASD is an umbrella term used to describe previous subcategories of autism, including terms such as Asperger's Syndrome and Autistic Disorder (see Diagnostic and Statistical Manual, 5th ed.; DSM-5; American Psychiatric Association, 2013). Individuals with autism can present with a number of challenging behaviours (see Emerson, 2001), including SIB. This is not considered a core symptom of ASD or part of the diagnostic criteria, possibly because it is not endemic to ASD (Minshawi et al, 2014). Indeed, SIB can also be observed in those with Learning Disabilities (LD)/ Intellectual Disabilities (ID) (Cooper et al, 2009). SIB behaviours in individuals with ASD are distinguished by being repetitive and stereotypic in nature, without intent of harm, as opposed to compulsive or episodic self-injury (Yates, 2004); such conceptualisations are considered in this review. Episodic self-injury (where individuals hold intention to cause harm) can also present in individuals with ASD (see Maddox, Trubanova & White, 2016; Hannon & Taylor, 2013), and will be distinguished as 'self-harm' in this paper.

SIB has been extensively researched across different fields. Neurobiological factors have contributed significantly to an understanding about the aetiology of SIB, where a number of factors have been suggested to associate with SIB, including pain reactivity and alterations in the somatosensory system, among other things. Exploration of this research base is beyond the scope of the present review; for further discussion see Deurden et al (2014), Tordjman et al (2018), Shirley et al (2016), Christenson et al (2009), Kolevzon et al (2014), Devine et al, (2014), Summers et al (2017) and Wolff et al (2013). Additionally, SIB has been conceptualised as part of different constructs of behaviour. First it is conceptualised as a challenging behaviour, and second, as a repetitive and restricted behaviour (RRB). Factors associated with challenging behaviours and RRB have also been explored in previous research (Matson et al, 2010; Cohen et al, 2018; Rattaz et al, 2018; McTiernan et al, 2011; Stratis & Lecavalier, 2013; Antezana et al, 2019).

However there has been a recent emphasis on conducting research into SIB as an individual difficulty. A primary reason for this emphasis centres around the prevalence of SIB both for individuals with ID and autism (Oliver, Licence & Richards, 2017; McClintock, Hall and Oliver, 2003). Accurate

prevalence estimates are difficult to determine due to methodological differences across studies, definitions of SIB, and participant characteristics (Summers et al, 2017). However, findings from a 37-paper meta-analysis has indicated that current pooled prevalence estimates of self-injury in individuals with autism is 42% (Steenfeldt-Kristensen, Jones & Richards, 2020). It has been long established that people with autism may be at particular risk of developing SIB (see Ando & Yoshimura, 1979). Research has indicated that individuals with higher rates of autistic behaviours displayed significantly more SIB across those with ASD, Fragile X Syndrome and Downs Syndrome (Richards et al, 2012). Researchers have been cautioned not to assume that SIB in ASD and ID populations arise from the same motivations (according to Weiss, 2002), and that research into SIB in ASD specific populations is warranted.

SIB has also been shown to be a persistent difficulty. Longitudinal and follow up studies have evidenced that SIB can be enduring for those with ASD (Richards et al, 2016; Baghdadli et al, 2008; Rattaz et al, 2015; Taylor et al, 2011). This is not to say that SIB is always life-long and untreatable, however, interventions such as medication and behavioural interventions are mixed in terms of efficacy (Schroeder et al, 1978; Eurtuk, Machalicek & Drew, 2018; Baghdadli et al, 2008).

Besides the high prevalence and persistence of SIB in ASD populations, it is an important area to research due to the associated outcomes. Individuals with ASD displaying SIB may cause long term damage or injury to themselves, including concussions, contusions, bleeding, lacerations, fractures, loss of sensory function and infections which cumulatively present as one of the primary reasons for adolescents with ASD accessing hospital emergency departments (Ianuzzi et al, 2012; Soke et al, 2018; Minshawi et al, 2014). The chance of placement in residential facilities or inpatient hospital settings increases for ASD populations with the presentation of SIB (Siegal et al 2012; Mandell, 2008). SIB also impacts significantly on carers such as teachers and parents, where SIB is considered to relate to increased caregiver stress and lower parental reported quality of life (Konstantareas & Homatidis, 1989; Lecavaller, Leone & Wiltz, 2006; Rattaz, Michelon & Baghdadli, 2015; South, Ozonoff, & McMahon, 2005).

Risk markers and factors associated with SIB have been explored in previous review. McClintock, Hall and Oliver (2003) conducted a meta-analysis and demonstrated that SIB was associated with autism, more profound LD/ID, and deficits in receptive and expressive communication. A systematic review conducted by Furniss and Biswas (2012) also indicated that SIB is associated with increased aggression, impulsivity and repetitive behaviour, although this research was limited to individuals with an LD/ID (also see Symons, Devine & Oliver, 2012; Richman, 2008). There are currently no systematic reviews which explore SIB in an ASD focused population.

Rationale

The association between SIB and ASD is an important area of research due to individuals with ASD seemingly being at higher risk of SIB. Prevalence estimates and persistence of SIB are higher in this population, which not only impacts on the individual and places them at risk of harm, but impacts more widely on parents, carers and teachers. This demonstrates the importance of developing targeting interventions to reduce SIB in this population, where it has been proposed that interventions should be based on hypotheses about the cause of a problem (Repp & Karsh, 1994). Furthermore, behavioural outcomes are improved by early identification and treatment of emerging SIB (Lance et al, 2014; Richman, 2008). Thus, exploration of risk markers and factors associated with SIB is key to better understanding and intervening for SIB, yet there remains limited understanding of the role of psychosocial and behavioural factors associated with SIB.

Research Question

What are the associated psychosocial and behavioural factors and predictors for self-injurious behaviour in individuals with autism spectrum disorders?

Terminology

Self-Injurious Behaviour (SIB)

Throughout this review, SIB is the term used to describe repetitive and stereotypic self-directed behaviours which results in physical harm to the individual without showing apparent intent of harm (Fee & Matson, 1992; Yates, 2004).

Autism

Since the release of the Diagnostic and Statistical Manual 5th Edition (DSM-5; American Psychiatric Association, 2013) diagnostic terminology reflects the conceptualisation of autism as a spectrum, thus the diagnostic term is 'Autism Spectrum Disorder'. Previously this would have included terms such as 'autism spectrum condition', 'high functioning autism' and pervasive developmental disorders such as Asperger's syndrome. For the purpose of this review, the term 'autism' is used to incorporate these previous diagnostic terminologies.

Psychosocial

For the purpose of this review, psychosocial factors refer to the combined influence of two categories of variables. This includes psychological factors which exist at an individual level, and second, social factors which are situated within surrounding environmental structures. (Singh-Manoux, MacLeod & Smith, 2003). Examples of psychosocial factors may be mood, intelligence, social communication.

Behavioural

The Australian Institute of Health and Welfare define behavioural risk factors as behaviours which individuals have the most ability to modify, which holds associations to a health disease (AIHW, 2016). Examples of this may be sleep, levels of activity or adaptive ability. This description was utilised in this review.

Associated Factors

This terminology is inclusive of both risk markers, which are factors which are associated with the occurrence of a behaviour, and of risk factors, which are causal to the emergence of a behaviour. Factors associated with SIB are therefore not protective factors which may prevent or reduce the likelihood of a behavioural occurrence.

Methodology

Scoping Searches

An initial unlimited search of Google Scholar, the Cochrane Library and Staffordshire University Library collection (Summon) was conducted, where existing reviews on this topic by Minshawi et al (2014) and Weiss (2002) were identified. These studies were narrative in nature, considering a broad range of topics. As these narrative overviews were not systematic reviews focusing uniquely on psychosocial and behavioural factors associated with SIB, the current review proceeded.

Search Strategy

Systematic searches of online databases were carried out during April 2019. Studies were identified through searches of the following databases: PsycINFO, PsycArticles, Scopus, Medline, CINAHL, and Research Autism. The search terms used were as follows: (Autis* OR “autism spectrum disorder” OR “autism spectrum condition” OR Asperger* OR PDD-NOS OR ASD OR ASC) AND (“self-injurious behaviours” OR “self-injurious behaviour” OR “self-injury”) AND (Predictors OR “risk factors” OR “associated factors” OR associat* OR predisposition OR correlation). Each database was searched separately. Limiters of English language and peer reviewed studies were set to ensure quality of the review. A limiter of publication after 1987 was set as this was the release date of the Diagnostic and Statistical Manual III-R, when people with autism were considered to potentially present with self-injury, such as head banging, as a form of RRB. The eligibility criteria are outlined in Table 1. Citations from eligible studies identified in the main search were then reviewed to identify additional relevant studies. No additional studies were identified through this citation review. The search strategy followed the Preferred Reporting Items for Systematic Review and Meta-Analyses (PRISMA, Moher et al, 2009) and is illustrated in Figure 1.

Table 1. *Eligibility criteria for inclusion in review*

Criteria	Inclusion	Exclusion
Participants	Research with a primary focus on people with an Autism Spectrum Diagnosis	Research focusing on people with learning disabilities or genetic disorders, due to the clinical distinctions between these populations
Study Design	Peer reviewed, empirical research which reports qualitative, quantitative or mixed method results	Book chapters Overviews Summaries Discussion papers
Topic	Research exploring psychosocial and behavioural factors associated with self-injury	Research on interventions, Genetic, physical, neurobiological research, research on self-harm, research into general challenging or repetitive behaviour.
Publication Year	1987 onwards	Pre-1987, before the release of the DSM-III-R which references repetitive and restricted behaviours (e.g. head banging)
Language	Written in the English Language Research conducted in any country.	

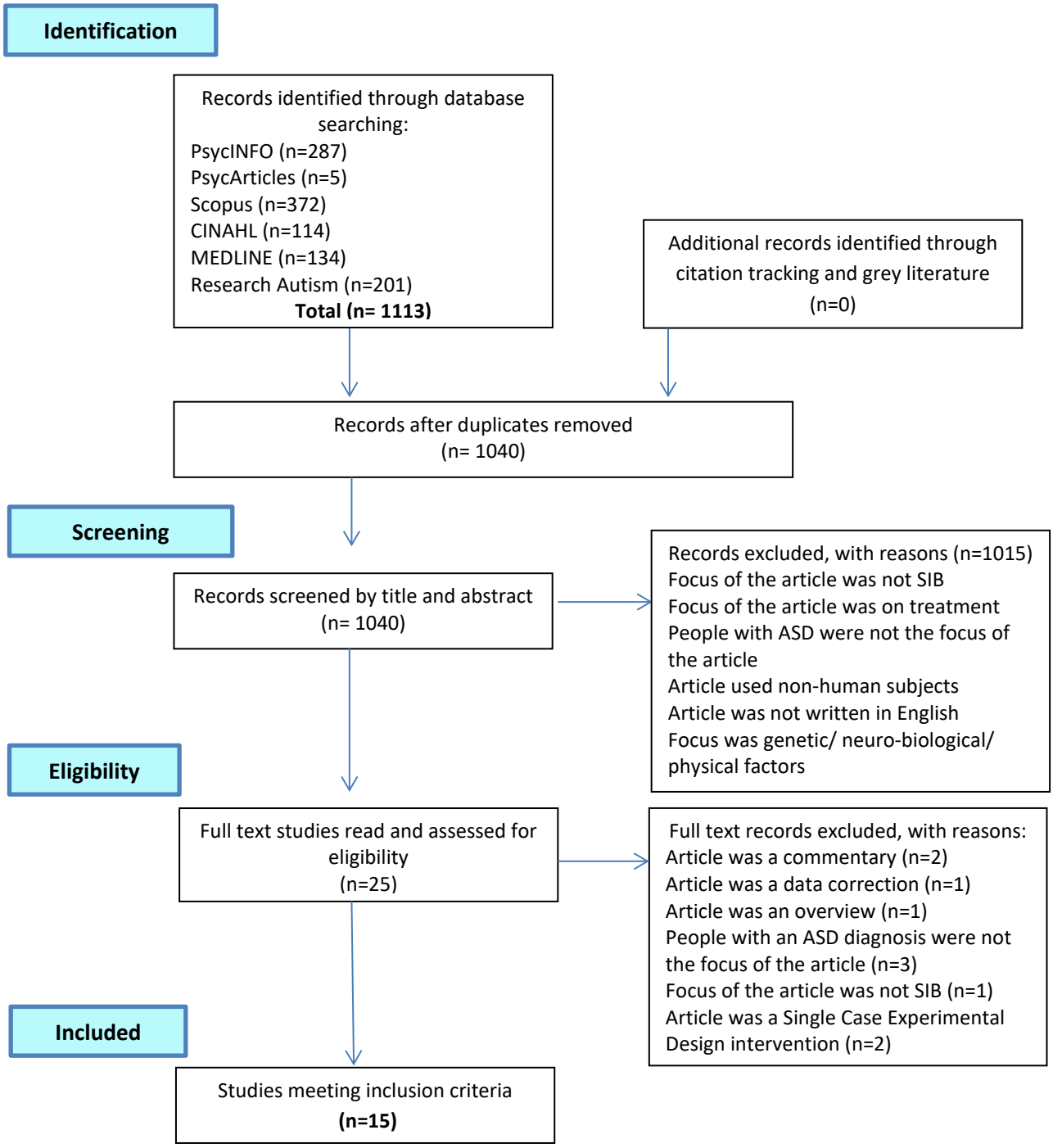

Figure 1. PRISMA Flowchart demonstrating search strategy for article inclusion (Moher et al, 2009)

Publication Bias

A search of the grey literature was conducted to minimise publication bias (see Dickersin, 1990). This search included Google Scholar, the Ethos Database for unpublished dissertations, and searches of charitable organisations including the National Autistic Society and the Interactive Autism Network. No additional empirical studies were identified, and no relevant unpublished theses were identified.

Quality Assessment

Eligible studies were all quantitative observational studies using cross sectional and cohort designs. Due to the mixed designs of the research, no single appraisal tool was identified from existing literature which was deemed sufficiently fit for purpose. An 18-item critical appraisal checklist was developed from evaluation tools for cohort studies and cross sectional studies, and from quality guidance for observational studies (Appendix 1). The use of a single tool rather multiple tools was also necessary to operationalise the quality of studies and allow for direct comparisons between studies. This informed the development of a data extraction tool which was applied to eligible studies. Specifically, the tools used to develop the critical appraisal checklist were:

- The Critical Appraisal Skills Programme checklist for cohort studies (CASP, 2018). 8 items from the CASP tool were used in the development of the current appraisal tool.
- The Appraisal of Cross-Sectional Studies tool (AXIS; Downes et al, 2016). 9 items from the AXIS tool were used in the development of the current tool.
- The Strengthening the Reporting of Observational Studies in Epidemiology statement (STROBE; Von Elm et al, 2007). 1 item in the current tool was taken from the STROBE statement.

Items were selected through a process of exhaustive comparison across the three tools; each item on the CASP was systematically considered against the items on the AXIS and STROBE, and items which were unaccounted for by the CASP on the AXIS tool were considered against the STROBE to

produce a comprehensive list of items. Where appropriate certain items were then collapsed to ensure that questions were binary rather than qualitative, which allowed for operationalised scoring. All items were scored in the same way and given equal weighting. Items which were answered 'yes' received 1 point, items which were answered 'no' or 'can't tell/comment' received 0 points. Points were totalled and a score was calculated as a percentage of the total score possible.

Results

Search Results

The initial search produced 1113 results from the combined database searches. Citations were transferred to RefWorks ProQuest. Duplicates were removed, limiting the results to 1040. Studies were then screened by title and abstract which resulted in 25 studies. These were read in full to assess for relevance, where 10 studies were removed. Of these 10, two studies were excluded only after discussion with a supervisor and independent reviewer. Overall a total of 15 studies were retained for inclusion.

Study Characteristics

The main characteristics of the eligible studies are outlined in Table 2. Of the design of studies included, five were cross sectional observational studies (Gulsrud et al, 2018; Handen et al, 2018; Richard, Davies & Oliver, 2017; Duerden et al, 2012; Poustka & Lisch, 1993), and three were cross sectional observational studies which utilised existing data (Baghdadli et al, 2003; Soke et al 2018; Lance et al, 2014). Four studies were observational studies based on information obtained from databases and data repositories (Richman et al, 2013; Soke et al, 2017; Dempsey et al, 2016; Soke et al, 2019). One was a prospective cohort study (Richards et al, 2016), two were a longitudinal follow up of data reported in previously existing studies (Bagdadli et al, 2008; Rattaz, Michelon & Baghdadli, 2015). 8 studies originated from the USA, 1 from Canada, 1 from Germany, 2 from the UK and 3 from France.

Table 2. *Summary of studies included in the review.*

Author, Year of Publication, Aims	Sample	Method	Analysis	Findings	Limitations	Critical Appraisal Tool Rating (%)
Poustka & Lisch, 1993 Aims to find out if self-injury in ASD is significantly correlated with autistic phenomena and/or degree of intellectual functioning.	Sample originated from Germany N=61 individuals diagnosed with ASD The median age of the sample was 15.3years, mean age not reported. 80% were males	Cross sectional observation study	Statistical methods not outlined, but reference given to chi-square correlations and multivariate analyses	Association between lower IQ and increased SIB was visibly observed, but statistical analyses did not reveal significant correlation. No correlation between SIB and severity of communication difficulties, social interaction difficulties, and repetitive stereotyped behaviours.	Limitations are not discussed, although there are potential limitations around sample size, recruitment and sampling strategy.	41

Baghdadli et al, 2003 Aims to identify risk factors for SIB among children with ASD with respect to age, ID, medical condition, degree of autism and parental social class	Sample originated from France N=222 children with ASD Mean age of the sample was 5 years. 80% were males A subset of participants from a cohort study identifying prognosis factors in children with autistic disorders (Ausilloux et al, 2001)	Cross sectional observation study. Data already existing from previous study. A subset of data collected during a cohort study.	Mann-Whitney test and Chi-Square tests used to compare groups (no SIB, SIB) Logistic regression	Significant relationships between SIB and presence of perinatal condition ($p<0.05$), higher speech delay ($p<0.01$), higher adaptive delays in communication, socialisation and daily living skills ($p<0.05$), degree of autism ($p<0.001$). SIB was more severe in children with higher autism severity, lower speech level, and lower adaptive skills ($p<0.01$). Risk factors (by adjusted odds ratio [OR]): higher degree of autism ($OR=1.1$), daily living skills delay	Limitations are not discussed, although there are limitations around recruitment and sampling.	47

				(OR=0.98), perinatal condition (OR=5.5), low chronological age (OR=0.69).		
Baghdadli et al, 2008 **A follow up study of Baghdadli et al, 2003.** Aims to describe the changes in children's SIB and determine whether childhood risk factors are related to a negative outcome of SIB	Sample originated from France N=185 children with ASD Mean age of the sample was 8 years. 80% were males.	Observation study A longitudinal follow up of a subset of data previously reported in an existing study.	Mann-Whitney test and Chi-Square tests used to compare groups (no SIB/disappearance of SIB, persistent SIB or emergence of SIB) Logistic regression	Significant relationship between SIB persistence or emergence and adaptive delay (p<0.001), worse cognitive deficits (p<0.001), speech impairment (p<0.001) autism severity (p<0.05) use of psychoactive drugs (p<0.007). Risk factors for persistent or new SIB: greater autism severity (OR=1.1), lower speech level (OR=3.5).	Sample may not be representative: Psychiatric clinics- may be lower functioning SIB rating obtained by caregivers so maybe not reliable. SIB questionnaire had not been validated SIB between time 1 and 2 not analysed.	61

Duerden et al 2012 Aims to assess incidents of SIB in ASD in a large sample of children and adolescents with ASD with previously defined risk factors.	Sample originated from Canada. N=250 children and adolescents with ASD. Mean age of the sample was 7.4 years old. 85% were males	Cross sectional (cohort) observation study	Hierarchical regression analysis Multivariate linear model	Factors predicting SIB: atypical sensory processing (explaining 12% of variance, p<0.0001), IQ (explaining 4% of variance, p<.01), social ability, (explaining 3% of variance, p<0.55) and sameness/resistance to change (explaining further 10% of variance, p<0.001). Severity of autism and rituals and compulsions did not predict self-injury.	Significant predictors did not account for much overall variance Participants had a high rate of autism severity: may not be representative	78
Richman et al, 2013 Aims to replicate and extend previous research	Sample originated from the USA N=617 individuals with ASD.	Cross sectional observation study	Structural equation modelling	SIB predicted by impulsivity (p<0.01), stereotypy (p<0.01) and low IQ (p<0.05). Unanticipated positive correlation between IQ and	Use of database – more up to date information may have been available Error through measurement and use of	65

on risk factors associated with SIB using items from the Aberrant Behaviour Checklist	Average age of the sample was 11.2 years old. 83% were male.	Analysis of information from a database.		Autism Severity ($p<0.0001$) but no relation between Autism severity and SIB.	indirect measures (use of secondary data).	
Lance et al, 2014 Aims to examine the associations between types of SIBs and a history of regression in a group of hospitalised patients with neuro-behavioural disorders.	Sample originated from the USA N=125adolescent inpatients with ASD The mean age of the sample was 10.9 years old. 75% were male	Observational retrospective review	Logistic regression	No significant differences in SIB observed between individuals with or without social, language, and behavioural regression.	Selection bias Small samples Limited generalisability Non-standardised definitions	71

Rattaz, Michelon & Baghdadli, 2015 Aims to identify the risk factors for SIB among adolescents with ASD, to describe the prevalence of SIB and the relationship between SIB and clinical or environmental factors.	Sample originated from France N= 152 adolescents with ASD, recruited from 46 autism-specialist clinics. Average age of the sample was 15 years old. 82% were male. A subset of participants from the French 'EpiTED' cohort, which follows the development of children with ASD over	Cross sectional observation study A longitudinal follow up of data reported previously.	Kruskal-Wallis Bonferroni post hoc Two polytomic logistic regressions	Factors associated with SIB: increased aberrant behaviours, autism symptom severity (p<0.001), drug use (p<0.006), lower adaptive skills (p<0.001), person related cognition (including theory of mind, attention, imitation and symbolic play) and object related cognition (spatial reasoning), (p<0.001) functional language (p<0.001), and developmental trajectory (p<0.001). Risk factors: autism symptom severity (p<0.04).	Subset of data – bias as the observations were not random	67

	a 10 year period (see Baghdadli et al, 2012).			Protective factors: IQ, communicative ability		
Dempsey et al, 2016 Aims to update the model of Deurden et al (2012) by re-running in a large sample, including anxiety as a factor, exploring the impact of IQ, and using a dichotomous and clinically relevant definition of SIB.	Sample originated from USA N=2341 children with ASD Mean age of the sample was 9 years. 85% were male.	Cross sectional observation study. Analysis of information from databases	Multivariate linear regression Multivariate logistic regression	Factors associated with SIB: lower non-verbal IQ (p<0.01) and social communication (p<0.05), increased anxiety (p<0.001), insistence on sameness (p<0.001), atypical sensory seeking (p<0.001)	Significant predictors did not account for much overall variance Function of SIB not analysed	71

Richards et al, 2016 Aims to compare SIB over time and establish persistence, to investigate variables associated with SIB at Time 2, to evaluate variables at Time 1 to assess presence of SIB at Time 2.	Sample originated from the UK N=67 carers of individuals with ASD Median age of sample was 13.5 years old 85% were males	Prospective cohort Follow up time was 36.4 months	McNemar and Wilcoxen signed ranks tests Chi-square, relative risks statistics and Mann-Whitney U tests. Kruskall Wallis tests	SIB is persistent and stable over time. SIB associated with non-verbal communication (p=0.005), lower ability (p=0.008), mood (p=0.032), social interactions (p<0.001), higher levels of stereotyped behaviour (p<0.013), compulsive behaviour (p=0.005), over-activity (p=0.004), sameness (p=0.043), repetitive behaviour (p<0.001) SIB risk markers: lower social interaction (p=0.026) and higher impulsivity (p=0.021)	Relatively small sample prevented some data analysis Under-representation of individuals with self-injury at T2 may limit external validity. Did not collect data on pharmaceutical and behavioural treatments for SIB.	83

Richards, Davies & Oliver, 2017 Aims to describe the prevalence, topography and severity of SIB and self-restraint within and between children and adults with ASD.	Sample originated from the UK N=424 individuals attending National Autistic Society adult services or schools. Mean age of the sample was 24.10 years. 78% were male.	Cross sectional observation study	Chi Square tests Relative Risk statistics Binary logistic regressions	Associations with SIB in child sample: lower ability, increased self-restraint, overactive/ impulsive behaviours, health problems (skin and digestive problems). Associations with SIB in adult sample: self-restraint repetitive behaviour, and overactivity/impulsivity SIB predicted by: overactivity/ impulsivity for child and adult samples. SIB in child sample also predicted by increased repetitive/restricted behaviour, health problems,	Possible sampling bias Use of screening tool rather than in depth instrument Studied limited number of factors previously identified in literature	82

				lower ability. (All statistics reported as relative risk statistics with 99% confidence intervals, small-medium effect sizes across all significant factors)		
Soke et al, 2017 Aims to assess factors associated with SIB in two large and distinct national samples, and to determine if any associations found are moderated by gender, IQ, or	Sample originated from the USA N=13,167 children with ASD The mean age of the ADDM database was 8 years old. 82% were males. The mean age of the AS-ATN database was 5.7 years old.	Cross sectional observation study. Analysis of information from the ADDM and AS-ATN databases	Non-linear mixed method model, multiple imputation	Across datasets, SIB associated with: impaired adaptive behaviour (p=0.006), developmental regression (p=0.003), maladaptive behaviours (aggression, p=<0.001. hyperactivity, p=0.05), problems with sleep (p=0.004) and sensory processing (p=0.004).	Retrospective data – not all desired data available. Selection bias Different methods of data collection Possible type II errors	71

maternal education.	83% were males					
Gulsrud et al, 2018 Aims to utilise a sample of individuals with ASD across a wide range of variables to provide characteristics of markers associated with SIB	Sample originated from the USA N=144 individuals with ASD The mean age of the sample was 9.3 years old 81% were males	Cross sectional observation study	ANOVA Likelihood Ratio Chi-square	Small-medium effect sizes (between 0.18-0.64) found for differences between individuals with/without SIB. Variables associated with current functioning: Impairments in verbal (p=0.019) and non-verbal IQ (p=0.036), cognition (p=0.012), awareness (p=0.014), social communication (p=0.005) and communication (p=0.037). Early markers associated with SIB: lower birth weight, premature birth, delayed	Sample size No direct observation of SIB Unable to collect desired data e.g. SIB persistence, onset. Large number of statistical tests may have obscured findings.	53

				crawling and bladder and bowel control		
Handen et al 2018 Aims to explore whether individuals who present with SIB at home and in hospital show more irritability and hyperactivity, and to explore predictors of SIB for an inpatient population	Sample originated from the USA N=302 children and adolescents with ASD in hospital inpatient units Mean age of the sample was 12.9 years old 79% were males	Cross sectional naturalistic observation	ANOVA tests Chi-square and Fisher's exact tests Tree structure classification	SIB is associated with lower non-verbal IQ (p<0.0001), higher externalising behaviours (irritability, p<0.0001), hyperactivity, p<0.0001, and stereotypy (p<0.0001). ASD severity and age not associated with SIB.	Naturalistic study – differences between recruitment sites e.g. length of stay, level of observation. Inpatient setting -onset of interventions and medications.	82
Soke et al, 2018 Aims to enhance our knowledge of	Sample originated from the USA	Cross sectional observation study	Log-binominal regression	Multivariable analysis of Current/Ever SIB factors: lower adaptive skills, sleep, and behavioural difficulties,	Large sample but SEED network only included 6 sites – not generalizable.	59

factors influencing SIB, and to evaluate the concordance between parental report of SIB and clinical observations of SIB.	N=692 children with ASD The mean age of the sample was 4.7 years old. 82% were males	Using data from the Study to Explore Early Development (SEED)		gastrointestinal problems, younger maternal age. Additional factors for Current SIB: genetic conditions, higher IQ, caesarean delivery, sensory problems. All statistically significant to minimum p<0.05 level.	Parent reports – possible over reporting due to stress or 'proto' SIB.	
Soke et al, 2019 Aims to explore associations between SIB and perinatal, prenatal and neonatal factors, and to validate associations	Sample originated from the USA N=4343 children from the Autism and Developmental Disabilities Monitoring Network surveillance.	Cross sectional observation study	Non-linear mixed models.	SIB associated with: developmental regression (OR = 1.35) IQ (OR=1.34), sleep (OR=1.61) and sensory problems (OR=1.35), aggression (OR=2.15) and argumentative behaviours (OR=1.24), temper tantrums (OR=1.24), co-occurring	Did not consider severity of SIB Missing data 'Ever' SIB may include 'proto' SIB. Information may have been under/over reported.	65

between SIB and developmental, medical and behavioural factors.	Average age of the sample was 8 years old. 83% were males		developmental (OR=1.21) and psychiatric diagnoses (OR=1.77) (95% confidence intervals) . SIB associated with maternal smoking and education, and electronic fetal monitoring during labour.	Sample not representative Possible type II errors

Quality Appraisal

Studies were appraised based on the information reported in individual studies. All studies reported clear aims and objectives of the research and provided a sufficient rationale for the study, and designed a study appropriate to the aims.

The majority of studies sought ethics approval or consent from participants or parents, although this was not explicitly achieved by Lance et al (2014), Richman et al (2013), or Deurden et al (2012), however there was no evidence of ethical misconduct. Limitations and theoretical or practical implications were discussed in all studies except two which were Poustka and Lisch (1993) and Baghdadli et al (2003). This may not be reported in the latter case because the study was continued and findings published in a separate article (Baghdadli et al, 2008) which did report such details.

There were a number of issues with study methodology. Richards et al (2017), Richards, Davies & Oliver (2016) and Handen et al (2018) appeared to recruit a representative sample and collected data in a way that would minimise bias. However, for the majority of studies there was generally a lack of explanation or transparency about methodology, particularly regarding sampling, recruitment and data collection methods. A number of studies utilised secondary data and stated that sampling and data collection methods were reported in a primary data source. The means of approaching participants were generally not reported and purposive sampling was generally used rather than random sampling which has implications for selection bias and representativeness. A wide range of subjective and standardised measures were used, some of which were not accurate measures of variables.

Sample size was justified only in one study (Deurden e al, 2012). A number of samples were limited by small or modest sample sizes (Lance et al, 2014; Richards et al, 2016; Gulsrud et al, 2018; and Rattaz et al, 2015; Poustka & Lisch, 1993) and studies reported various comorbidities. With the exception of Poustka and Lisch (1993), all studies reported on statistical analysis methods, although four studies lacked clear justification for the data analysis method carried out (Soke et al, 2019; Lance et al, 2014; Soke et al, 2018; Gulsrud et al, 2018). Four studies did not report confidence intervals to support precision of results (Deurden et al, 2012; Poustka & Lisch, 1993; Richman et al, 2013; and

Gulsrud et al, 2018), although Gulsrud et al (2018) did report high effect sizes. All studies except Poustka and Lisch (1993) reported statistical significance levels. Six studies reported methods sufficiently to allow for replication (Handen et al, 2018; Richard, Davies & Oliver, 2017; Richman et al, 2013; Deurden et al, 2012; Richards et al, 2016; Soke et al, 2017). See table 2 for information about the strength of associations, such as significance levels.

Three studies reported conflicts of interest which could affect the interpretation of results, either due to the support that authors' received or due to funding arrangements and participant recruitment (Handen et al, 2018; Soke et al, 2018; Dempsey et al, 2016). Five studies reported no conflicts of interest (Soke et al, 2019; Richards et al, 2016; Gulsrud et al, 2018; Deurden et al, 2012, Richards, Davies & Oliver, 2016). The remaining seven studies did not report this information.

Using the critical appraisal tool the lowest scoring study was appraised at 41% (Poustka & Lisch, 1993) and the highest scoring article was appraised at 83% (Richards et al, 2016). Studies by Baghdadli and colleagues (Baghdadli et al, 2003; Baghdadli et al, 2008; Rattaz et al, 2015) received similar critiques largely focused around their methodology and lack of transparency. Upon reading the full text it becomes apparent to the reader that each study uses a subset of data from primary publications (Ausilloux et al, 2001; Baghdadli et al, 2007). Whilst authors direct readers to primary sources for full explanations of their sampling and participants, readers are left without clarity as to information regarding population, sampling method, sample size calculation, justification for eligibility criteria, definitions of variables, and use of standardised measures. Similar concerns can be reported regarding the study by Poustka and Lisch (1993), which lacks clarity and transparency both around the methodology and the statistical analyses undertaken and reported on. For these reasons, results and conclusions from these studies might be viewed with particular caution.

Overview of Methodological Quality of Studies

Sample

Sample sizes ranged from 61 (Poustka & Lisch, 1993) to 13, 167 participants (Soke et al, 2017). The age of participants ranged between 2 years (Baghdadli et al, 2003; Soke et al, 2017; Soke et al, 2018) to 61 years (Richards, Davis & Oliver, 2017). Twelve studies focused exclusively on child and adolescent populations (Baghdadli et al, 2003; Soke et al, 2017; Soke et al, 2018; Duerden et al, 2012; Rattaz et al, 2013; Handen et al, 2018; Soke et al, 2019; Richards et al, 2016; Richman et al, 2013; Baghdadli et al, 2008; Lance et al, 2014; Dempsey et al, 2016), while three studies also included adult populations (Richards, Davis & Oliver, 2017; Gulsrud et al 2018; Poustka & Lisch, 1993).

Participants were recruited from different settings. This included clinic-based populations (Soke et al, 2017; Baghdadli et al, 2003; 2008, Soke et al, 2018; Rattaz et al, 2015) hospital based clinics (Gulsrud et al, 2018), community populations (Richards e al, 2016; Richards, Davies & Oliver, 2017; Soke et al, 2019; Dempsey et al, 2016) and inpatient hospital settings (Lance et al, 2014; Handen et al, 2018). In three studies, the setting from which individuals were recruited from was not stated (Richman et al, 2013; Poustka & Lisch, 1993; Duerden et al, 2012).

Of the studies reviewed, four involved the active recruitment of participants (Richards et al, 2016; Richards, Davis & Oliver, 2017; Poustka & Lisch, 1993; Gulsrud et al, 2018). Of these, two studies utilised a volunteer sampling method and recruitment via questionnaire packs (Richards et al, 2016; Richards, Davis & Oliver, 2017). One study involved a mixture of self-referral and referral by primary care physician or school for a neurodevelopmental evaluation. The method of advertising the neurodevelopmental evaluation was not specified (Gulsrud et al, 2018). The means of recruitment were not outlined by Poustka and Lisch (1993).

Eleven studies did not directly recruit a sample of participants, but instead used samples from other studies, subsets of existing study data, database repositories, or reviews of information pertaining to specific existing samples. Samples were extracted from: the Autism Inpatient Collection (Handen et al, 2018), the admission database for the Maryland Neurobehavioural Unit (Lance et al, 2014), the Autism and Developmental Disabilities Monitoring Network (Soke et al, 2019; 2017), the Autism Speaks- Autism Treatment Network (Soke et al, 2017), the Simon's Simplex Collection (Dempsey et

al, 2016), the National Database for Autism Research (Richman et al, 2013), Genetic studies at the Offord Centre or the Autism Research Unit in Canada (Duerden et al, 2012), the Study to Explore Early Development (Soke et al, 2018), and the EpiTED cohort in France (Rattaz, Michelon & Baghdadli, 2015). Baghdadli et al (2003; 2008) used data collected in another study by Aussilloux et al (2001). From the majority of these studies it was difficult to determine if a representative sample was obtained due to the nature of their recruitment.

Procedure

Studies collected data from a variety of sources. Soke et al (2019) collected information through reviewing summary files comprising health and education records as well as birth certificates. Data from health or school records were also accessed by Rattaz et al (2015). Lance et al (2014) collected data from inpatient admission medical records.

Most studies, with the exceptions of Soke et al (2019) and Poustka and Lisch (1993) involved the use of questionnaires. The sole use of questionnaires to collect data was implemented by Richards, Davis and Oliver (2017) and Richards et al (2016). While this method reduces interviewer bias, questionnaire designs are more prone to social desirability, potential sampling bias, and may not provide ‘rich’ data (Pattern, 2016).

The majority of studies involved standardised assessments (Deurden et al, 2012; Rattaz et al, 2015; Richman et al, 2013; Baghdadli et al, 2008; Soke et al, 2019; Dempsey et al, 2016; Handen et al, 2018; Soke et al, 2017; Gulsrud et al, 2018; Poustka & Lisch, 1993). The use of standardised assessments increases the validity and reliability of the studies. Four studies also conducted semi-structured interviews (Baghdadli et al, 2003; Baghdadli et al, 2008; Dempsey et al, 2016; Poustka & Lisch, 1993) or clinician observations (Rattaz et al, 2015; Baghdadli et al, 2003; Baghdadli et al, 2008).

Measures

SIB

Different measures were used to measure self-injury. Three studies (Handen et al, 2018; Richman et al, 2013; and Rattaz, 2015) used the *Aberrant Behaviour Checklist (ABC)*, which assesses problem behaviour in children and adults with developmental disabilities. This includes subscales of hyperactivity, irritability, impulsivity, stereotypy, and lethargy, where SIB can be derived from items on the subscales. Handen et al (2018) report the measure to be reliable in ASD populations.

SIB was also measured by items on the *Challenging Behaviour Questionnaire* (Richards et al, 2016; Richards, Davies & Oliver, 2017) by the self-injurious subscale of the *Repetitive Behaviour Scale-Revised* (Handen et al, 2018; Deurden et al, 2012), and in three studies, item 83 of the *Autism Diagnostic Interview-Revised* was used as a measure of SIB (Dempsey et al, 2016; Soke et al, 2018; Duerden et al, 2012). SIB was also coded 'yes/no' from observational data and records (Soke et al, 2019; Soke et al, 2019; Lance et al, 2014), and rated by clinical judgement (Baghdadli et al, 2008; Baghdadli et al, 2003; Poustka & Lisch, 1993).

Autism

A range of measures were used to confirm the diagnosis of autism. Five studies used the *Autism Diagnostic Interview – Revised (ADI-R)* measure, which assesses the presence of the core domains of autism (Gulsrud et al, 2018; Poustka & Lisch, 1993; Soke et al, 2018; Rattaz et al, 2015; Deurden et al, 2012). The properties of the ADI-R were reported in one study (Gulsrud et al, 2018) as having good inter-rater reliability for the three core domains, between 0.62 and 0.89, and good internal consistency with domains ranging between 0.69 and 0.95. Use of such a measure suggests increased validity in the participants' ASD diagnosis. Baghdadli et al, (2003) also used the ADI-R, but as a measure of expressive speech.

Two studies referred to confirmation of diagnosis using the *Autism Diagnostic Observation Schedule (ADOS)* (Handen et al, 2018; Soke et al, 2017), which is an assessment tool that is used to examine the core components of autism. Neither study included information about the reliability or validity of the assessment. Eight studies did not use standardised measures to explicitly confirm a diagnosis of

autism (Richards et al, 2016; Richards, Davies & Oliver, 2017; Soke et al, 2019; Lance et al, 2014; Richman et al, 2013; Baghdadli et al, 2003; Baghdadli et al, 2008; Dempsey et al, 2016).

Associated Variables

However, most studies conducted an assessment of autism severity. Seven studies used the *Autism Diagnostic Observation Schedule (ADOS)* to assess the presence and severity of autism (Duerden et al, 2012; Richman et al, 2013; Soke et al, 2017; Soke et al, 2018; Gulsrud et al, 2018; Handen et al, 2018; Poustka & Lisch, 1993). Autism severity was also assessed using the *Childhood Autism Rating Scale (CARS)* which measures autism between 1 (normal) and 4 (maximum severity). Three studies used this measure whereby after a 20 minute video recording of the participant and an adult, the participant's autism severity was observed and rated by two independent clinicians (Baghdadli et al, 2003; Baghdadli et al, 2008; Rattaz et al, 2015).

The *Vineland Adaptive Behaviour Scales (VABS)* was used in eight studies as a measure of adaptive functioning (Soke et al, 2017; Gulsrud et al, 2018; Handen et al, 2018; Soke et al, 2018; Duerden et al, 2012; Rattaz et al, 2015; Baghdadli et al, 2003; Baghdadli et al, 2008). The VABS is used across age groups, typically completed in a semi-structured interview with parents, and is comprised of communication, daily living skills, and socialisation domains. Gulsrud et al (2018) reported internal consistency as 0.86 to 0.98, and test-retest reliability ranging from 0.83 to 0.96, suggesting this is a reliable measure.

Ten studies completed a measure of intelligence (IQ), which varied depending upon the age and ability of the participant (Handen et al, 2018; Richman et al, 2013; Duerden et al 2012, Gulsrud et al, 2018; Soke et al, 2017; Dempsey et al, 2016; Soke et al, 2018; Baghdadli et al, 2008; Rattaz et al, 2015; Poustka & Lisch, 1993). Measures used across these studies were the *Leiter International Performance Scale,* the *Mullen Scales of Early Learning (MSEL)*, the *Wechsler Preschool and Primary Scale of Intelligence 4th edition (WPPSI-IV),* the *Wechsler Intelligence Scale for Children Third Edition (WISC-III)* the *Wechsler Intelligence Scale for Children Fourth Edition (WISC-IV),* the *Wechsler Intelligence Scale for Children Fifth Edition (WISC-V),* the *Stanford-Binet Intelligence*

Scales, Fifth Edition, Wechsler Abbreviated Scales of Intelligence- Second Edition (WASI-II), the *Wechsler Adult Intelligence Scale Fourth Edition (WAIS-IV)*, the *Differential Ability Scales-II*, the *Peabody Picture Vocabulary Test*, and the *Brunet-Lezine Test*. Poustka and Lisch (1993) used German translated versions of the WISC and WAIS. The range of tools used to measure IQ makes it difficult to compare findings across studies.

Particular hypotheses were tested in individual studies, for example, exploration of the role of executive functioning, or affect. Relevant measures were utilised to assess such hypotheses, namely the *Behaviour Rating Inventory of Executive Function (BRIEF)* and the *Mood Interest and Pleasure Questionnaire- Short (MIPQ-S)* respectively. A full list of additional measures is listed in Appendix 2. Richards et al (2016) and Richards, Davies & Oliver (2017) generally report good reliability of their measures. However, limited information is provided about measures in a number of other studies, which calls into questions the robustness of findings based on these measures.

Synthesis of Main Findings

Autism Severity

Mixed findings were reported regarding the association between autism severity and SIB. Four studies found that severity of ASD or increased ASD phenomenology was associated with SIB (Richards et al, 2016) and that it is a risk factor for SIB (Baghdadli et al, 2003; Baghdadli et al, 2008; Rattaz et al, 2015). On the other hand, no associations were found between ASD severity and SIB in five studies (Handen et al, 2018; Gulsrud et al, 2018; Soke et al, 2017;Deurden et al, 2012;Richman et al, 2013). Is has been suggested that this discrepancy could be due to sampling differences, measurement variables, data analysis procedures and the characterisation of autism severity (Duerden et al, 2012; Handen et al, 2018).

Characteristics of ASD

A number of studies explored core characteristics of ASD as factors associated with SIB. Lower levels of social communication and social interactions were found to be related to, or predictive of, increased SIB (Gulsrud et al, 2018; Richards et al, 2016; Deurden et al, 2012). An association between SIB and insistence on sameness was found in two studies, (Richards et al, 2016; Deurden et al, 2012). An increased level of RRB was identified as a factor increasing SIB in two papers (Richards, Davies & Oliver, 2017; Richards et al, 2016). However, Deurden et al (2012) and Gulsrud et al (2018) contradicted this, reporting that RRB was not a significant predictor of SIB. Poustka and Lisch (1993) reported no associations between SIB and the core features of ASD.

IQ

Results generally suggested a negative association between IQ and increased levels of SIB. Studies reported a fairly consistent finding that lower IQ is associated with SIB (Handen et al, 2018; Gulsrud et al, 2018; Baghdadli et al, 2008; Soke et al, 2019; Rattaz et al, 2015; Duerden et al, 2012; Dempsey et al, 2016) and is a risk factor for SIB (Rattaz et al, 2015; Richman et al, 2013). Soke et al (2017) reported a negative association between IQ and SIB, however this did not reach statistical significance. Unlike other studies, Soke et al (2018) conversely reported a positive relationship between IQ and SIB. Authors suggested that decreased IQ might affect functional ability to engage in SIB. No association was found between SIB and IQ by Poustka and Lisch (1993).

Adaptive Behaviour

Findings were generally consistent for adaptive behaviour. Five studies found an association between low adaptive ability and increased SIB (Baghdadli et al, 2003; Soke et al, 2018; Baghdadli et al, 2008; Soke et al, 2017; Rattaz et al 2015). Two studies (Richards, Davies, & Oliver, 2017; Richards et al, 2016) did not explore adaptive behaviour per se, but explored ability levels; they reported an association between lower ability and increased SIB. Although findings were generally consistent, no association was found between adaptive behaviour delay and higher levels of SIB in Gulsrud et al. (2018). Authors suggested that this finding may be due to small sample size. Findings for the role of

adaptive behaviour were not explicitly reported in one study exploring this variable (Handen et al, 2018).

Impulsivity and Over-activity

Consistent findings in four studies reported an association between SIB and increased impulsivity and aberrant behaviours such as over-activity and stereotypy (Richman et al, 2013; Richards et al 2016; Rattaz et al 2015; Richards, Davies & Oliver, 2017). Over-activity, which appears to be conceptualised in the same way as hyperactivity, and Impulsiveness was found to be predictive of SIB in both child and adult populations.

Language/ Lower Speech ability

Lower speech level, lower levels of functional language, and non-verbal communication is found to be a risk factor associated with increased levels of SIB, whereas higher levels of communicative ability is found to be a protective factor against SIB (Baghdadli et al, 2003; Baghdadli et al, 2008; Richards et al 2016; Rattaz et al, 2015). These findings were not supported by Deurden et al (2012), where functional communication was not significantly predictive of SIB.

Atypical Sensory Processing

A further factor associated with increased SIB was atypical sensory processing, where individuals with ASD can present with a number of abnormalities in processing sensory stimuli which could cause stress (Soke et al 2017; Dempsey et al, 2016; Soke et al, 2019; Deurden et al, 2012). In two studies (Deurden et al, 2012; Dempsey et al, 2016) atypical sensory processing was the single biggest predictor of SIB. However, in both studies this only accounted for a small proportion of the overall variance, suggesting other relevant factors may be unaccounted for.

Sleep

Although sleep was only investigated as a variable in three studies (Soke et al, 2017; Soke et al, 2018; Soke et al, 2019), all reported sleep to be a significant variable associated with SIB. These studies which found sleep to be a significant factor were all focused on child samples.

The factors described above are those with the strongest evidence. However, a number of other factors were found to be associated with SIB which includes, externalising behaviours such as aggression and behavioural difficulties (Handen et al, 2018; Soke et al, 2018; Soke et al, 2019), and mood (Richards et al, 2016). Mixed findings were found for the role of regression (reverting back to 'younger' behaviours) where one study reported no association (Lance et al, 2014) and others identified a role for developmental regression and SIB (Soke et al, 2017; Soke et al, 2019).

Discussion

The purpose of this review was to determine what psychosocial and behavioural factors are associated with self-injurious behaviour for individuals with autism spectrum diagnoses. 15 studies were systematically identified, reviewed and critically appraised. The quality of the studies was generally good overall, although the methodology was poor across a number of them, potentially influencing the validity of the results due to bias. A number of behavioural and psychosocial factors were associated with, or predictive of, increased levels of SIB in individuals with ASD. These factors include lower levels of IQ, adaptive ability, speech and language skills, sleep, atypical sensory processing, and higher levels of impulsivity and overactivity. Mixed findings were reported regarding the association between increased levels of SIB and autism severity and phenomenology.

While the finding of an association between factors does not elucidate the function of the behaviour, the results of the studies can offer insights into theoretical perspectives regarding SIB. The finding that impulsivity and over-activity has been consistently found to be associated with increased SIB lends support to a theory that SIB is underpinned by an impaired behavioural inhibition (Richards et

al, 2016). This theory is in line with previous research in the Attention Deficit Hyperactivity Disorder (ADHD) literature, where individuals with ADHD display similar impulsive and over-active behaviours. Here, individuals experience deficits in response inhibition, which are considered a primary form of executive dysfunction contributing to such impulsive behaviours (Barkley, 1997; Scheres et al, 2004). Thus it could be intuitively argued that if SIB is associated with impulsivity in individuals with ASD, there could be a similar link to deficits in response inhibition (Richman et al, 2013).

Research indicating that SIB is associated with lower speech abilities may support a theory that SIB is used as a way of communicating. This converges with previous research indicating that deficits in communicative ability is associated with more behavioural problems in individuals with developmental difficulties (Sigafoos, 2000) and learning disabilities (Chamberlain, Chung & Jenner, 1993). It has also been demonstrated that interventions to increase functional communication reduce 'maladaptive behaviours' in individuals displaying behaviours such as self-injury and aggression (Carr & Durand, 1985). The association between communication and behaviour is embedded in wider research. For example, literature on self-harm suggests that a function of the behaviour may be to communicate distress (Nock, 2009).

Consideration that SIB is a means of communication opens wider channels of theoretical exploration. Literature highlights that lower communicative ability is related to adaptive functioning, severity of autism symptomology, and IQ (Klin et al, 2007; Kjellmer et al, 2012). A relationship has also been identified between lower communicative ability and increased abnormalities in sensory processing (Patten, 2013). This complex association between SIB and deficits in communication, IQ, sensory processing and adaptive ability may relate to information processing and how individuals with ASD make sense of the world around them.

Limitations of Included Studies

Although the studies were generally appraised as being of good quality, several limitations need to be considered before drawing conclusions from the research. First, definitions used to refer to SIB are inconsistent and research continues in its struggle to distinguish SIB and self-harm in a way which

might be meaningful to readers and participant groups. This is of particular importance considering that research has relied predominantly on parent report, and that data has largely been collected through questionnaires, where researchers may not have chance to qualify terms, meaning that parents may report higher levels of SIB or be referring to self-harm or proto-SIB.

Second, the majority of studies used secondary data, most of which relied on databases or case files/ chart reviews. Acknowledged by most authors, there remain limitations associated with this around recruiting a representative sample, differing definitions of variables, and only analysing data which is readily available. Several studies were insufficiently transparent with their methodology to allow replication of results, calling into question the scientific value of the research. A number of studies were limited by selection bias, and potential under/over reporting of data.

Third, studies which explored variance reported that significant factors predicting SIB accounted for a small proportion of the overall variance. Findings from Deurden et al (2012) and Dempsey et al (2016) identified a number of significant variables predicting SIB. However, overall there is still up to 71% variance unaccounted for by these models, which has led to criticism regarding the extent to which we can draw conclusions about factors influencing SIB (Forgeot D'Arc et al, 2012). Thus, even significant and precise findings regarding factors associated with SIB may only give us limited understanding of aetiology.

Limitations of Current Review

The critique and analysis tool presented in this review has been undertaken by an individual researcher. The protocol was not registered with an open science platform. This introduces potential subjectivity and bias, where there is a lack of inter-rater reliability. This said, the use of PRISMA guidelines, specific eligibility criteria and verification from a research supervisor could sufficiently reduce subjectivity. Although the current appraisal tool lacks formal validity and reliability, items on the new appraisal tool were taken directly from valid appraisal tools which were systematically amalgamated and verified by a supervisor to reduce subjectivity.

Furthermore, researchers should be cautious in drawing concrete conclusions based solely on this review due to its strict inclusion criteria. Here, SIB has been studied as a single construct presenting in ASD populations. Although the reasons for this are understandable due to its prevalence, persistence, and association with negative outcomes, it limits and possibly simplifies the presentations observed across the literature for this population. Beyond the scope of this review, SIB is also classified as a challenging behaviour, and as a repetitive and restricted behaviour in wider areas of research. Readers are therefore directed to consult literature in these domains which may reveal further insights to factors associated with SIB.

Implications

This review has highlighted the complexity of presentations of SIB in ASD populations and has indicated a broad range of factors which could be significant to consider. Assessments in clinical practice need to be mindful of this and be sufficiently comprehensive to explore behavioural, social and psychological factors which could relate to the behaviour. Assessments should endeavour to include direct interview with parents to offer clarity of terms and explore SIB thoroughly, as questionnaires may offer limited information. Furthermore, parental involvement is necessary for more effective treatments for children with ASD (Burrell & Borrego, 2012). There is also a need for direct assessment of SIB, such as conducting a functional behavioural assessment, which is deemed as one of the most prominent means of assessing any challenging behaviour, including SIB (Neidert et al, 2013; see Emerson, 1995).

These findings have also supported the view that assessments should be offered as early as possible so proactive early interventions could be targeted for individuals identified as ‘at risk’ of SIB (Richards et al, 2016; Soke et al, 2017; Gulsrud et al, 2018). It has been suggested that intervention and prevention of SIB could begin before diagnosis, as similar factors associated with SIB have been identified in very young children pre-diagnosis, at risk of autism (Dimian et al, 2017).

Clinical practice should focus on the development of targeted treatment protocols and differential treatments (Dempsey et al, 2016; Richman et al, 2013). Results have emphasised the need for specific treatments aiming to develop interventions to focus on factors including communicative abilities, sensory processing, and the need for sameness (Rattaz et al, 2015; Baghdadli et al, 2008; Deurden et al, 2012). With this is mind, there are implications for formulation from Clinical Psychologists to make sense of such complex assessments and offer insights to guide treatment plans.

The various factors highlighted in influencing SIB and the identification of target areas for intervention give rise to the need for multi-disciplinary working in autism services. Difficulties with behaviour and affect suggest the need for mental health professionals such as psychologists and Board Certified Behavioural Analysts to implement psychological and behavioural approaches, while factors associated with SIB such as communication, adaptive ability, sensory processing difficulties, and sleep suggest that Speech and Language Therapists, Psychiatrists, and Occupational Therapists could have a significant role in providing intervention for individuals presenting with SIB.

Lastly, there are clinical implications around the wider impact that the development of such interventions may have. Namely, early intervention to reduce SIB could significantly reduce hospital admissions for this population and improve their quality of life through minimising the long term negative physical effects of self-injury. Reduction in SIB through awareness of risks and early intervention may also have a distinct positive impact on parents, carers, and teachers, who experience a person's self-injury as distressing. In other words, targeting a reduction in SIB holds implications not just for individuals with SIB, but for their carers, support systems, and for health services.

Future Research

Future research should attempt to address the limitations outlined in this review. The inconsistent definitions of SIB make comparisons across studies difficult, and definitions may be compounded with

descriptions of self-harm. Clarity is needed not only in terms, but in methodology. A need for more studies employing methodologies to investigate associations with SIB which collect primary data is called for.

It is noted that the majority of research included in this review predominantly collected data from parental reports. This may be natural given that parents are generally the primary caregiver and may hold the most insight to a child, indeed often acting as an advocate or 'voice' for a child with ASD during health appointments (Boshoff, Gibbs, Phillips, Wiles, & Porter, 2016). However, considering that autism presentations are persistent across different settings, perspectives from other carers, respite workers, teachers, and the individuals themselves is lacking. Wider insights may impact on perceptions of important factors associated with SIB.

The majority of research was also cross sectional in design. To explore risk markers for SIB and factors predicting SIB it is suggested that an emphasis be placed on longitudinal research to further explore the variables highlighted by current research. To achieve this successfully, researchers would be encouraged to recruit sufficiently sized samples.

Furthermore it is recommended that research explore a wider population base beyond the emphasis on children. This focus potentially limits our understanding of the course of SIB since different factors have shown to be associated with SIB in children and adults (Richards, Davies & Oliver, 2017). Autism is a lifelong disorder, and presentations of SIB are also observed in older adult populations (Kats et al, 2015). In light of this, research is encouraged to be more age inclusive in their samples to make findings more generalizable.

Conclusion

This review aimed to establish the current understanding of factors associated with SIB in ASD populations. Current research lacks robustness due to methodological issues and a reliance on secondary data. Findings demonstrated that a number of factors are associated with or predictive of

SIB, including levels of adaptive functioning, atypical sensory processing, communicative ability, IQ, sleep, and impulsivity and over-activity. Mixed findings were revealed as to the role of severity of autism phenomenology with SIB. There is evidence that SIB may be associated with different factors during different stages of life, although it would be recommended that future research explore this further through longitudinal designs. Multidisciplinary teams could be utilised to provide early assessment, develop and provide differential targeted treatments given the wide range of factors associated with SIB in this population, although further research is needed to inform such treatments.

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Conflicts of Interest

There are no conflicts of interest to report.

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Appendix 1. Quality Assessment Tool

1. Were the aims/objectives of the study clear?
2. Was a sufficient background and rationale for the study provided?
3. Was the study design appropriate for the aims?
4. Was the cohort recruited in an acceptable way?
5. Was the sample size justified?
6. Were variables accurately measured to minimise bias?
7. Have the authors identified all important confounding factors?
8. Was the follow up of subjects long enough?
9. Was the follow up of subjects complete enough?
10. Were the methods (including statistical methods) sufficiently described to enable them to be repeated?
11. Was ethical approval or consent of participants attained?
12. What are the results and do you believe the results?
13. Were the results presented for all the analyses described in the methods?
14. Are the results precise? (What are the confidence intervals?)
15. Were the authors' discussions and conclusions justified by the results?
16. Were the limitations of the study discussed?
17. Are there implications of this study for practice, theory or future research?
18. Were there any funding sources or conflicts of interest which may affect the authors' interpretation of the results?

Appendix 2. Table to show additional measures used across included studies

Name of measure	Authors who utilised measure	Description of measure
The Social Communication Questionnaire	Richards et al (2016)	A carer report questionnaire, based on the Autism Diagnostic Interview, this 40 item measure screens for ASD
The Social Responsiveness Scale, second edition	Gulsrud et al (2018)	A parent report used to assess the level of ASD related impairment
The Repetitive Behaviour Scale-Revised (RBS-R)	Deurden et al (2012), Dempsey et al (2016), Handen et al (2018).	A 44-item parent-completed questionnaire that measures repetitive behaviours in children and adolescents with ASD.
The Activity Questionnaire	Richards et al (2016)	A carer report measure to assess overactivity, impulsivity and impulsive speech across 18 items, not validated
The Self-Restraint Checklist	Richards, Davies & Oliver (2017)	A carer report questionnaire which describes seven topographies of self-restraint, caregivers are asked to endorse whether the behaviour is present.
The Wessex Scale	Richards et al (2016)	An carer report of ability in children and adults with intellectual disabilities, not validated
The Child Behaviour Checklist (CBCL)	Gulsrud et al (2018), Dempsey et al (2016)	A parent-report questionnaire assessing social, emotional and behavioural functioning. Dempsey et al (2016) used this questionnaire as a measure of anxiety.
The Adult Behaviour Checklist	Gulsrud et al (2018)	A parent report questionnaire assessing social, emotional and behavioural functioning.
The Behaviour Rating Inventory of Executive Functioning (BRIEF)	Gulsrud et al (2018)	A parent-report questionnaire used to measure executive functioning in real life situations. For children ages 2-5years, the BRIEF-Preschool Version was used.

The Parental Concerns Questionnaire	Soke et al (2017)	A screening tool for identifying problem behaviour for young children at risk of developmental delays
Self-Injury Aggression and Destruction Screening Questionnaire (SAD-SQ)	Richards, Davies & Oliver (2017)	Developed as a carer report screening measure to assess putative risk markers for challenging behaviour - Overactivity/Impulsivity, Repetitive/Restricted Behaviours and Ability.
Seibert and Hogan's Scale	Baghdadli et al (2008)	A measure for clinician's use of person-related cognition consisting of subscales for social interaction, joint attention and behaviour adjustment.
The Early Social Communication Scale	Rattaz et al (2015)	Items were taken from this scale for clinicians to assess person-related cognitive functioning including theory of mind, symbolic play, imitation and joint attention
The Oral and Written Language Scales (OWLS)	Duerden et al (2012)	A clinician's measure to assess expressive and receptive language skills.
The Mood Interest and Pleasure Questionnaire-Short	Richards et al (2016)	A carer report questionnaire comprising of 12 items to assess affect across mood and interest and pleasure.
The Child Sleep Habit Questionnaire	Soke et al (2018)	A standardised parent-report instrument for assessing sleep across 5 domains.
The Gastrointestional Symptom Inventory	Soke et al (2018)	No description provided. An assessment of gastrointestinal symptoms, not validated.
The Parental-developmental-disorders-Quality of Life (PAR-DD-QoL)	Rattaz et al (2015)	A parent-report questionnaire to measure parental quality of life.