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Predictors of Self-Injurious Behavior and Self-Restraint in Autism Spectrum Disorder: Towards a Hypothesis of Impaired Behavioral Control

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Abstract

Self-injury is common in autism spectrum disorder (ASD); however few studies have investigated correlates of self-injury or the putative associations with self-restraint. Questionnaire data on self-injury, self-restraint, health conditions, overactivity/impulsivity and repetitive/restricted behavior were collected on 208 children and 216 adults with ASD (mean age=24.10, range 6-61). Self-injury and self-restraint were frequent and significantly associated in both children (45.7% and 40.9%, $p<.001$) and adults (49.1% and 42.6%, $p<.001$). Severe self-injury was predicted by lower ability, health conditions and overactivity/impulsivity in children ($p<.001$) and repetitive/restricted behavior and overactivity/impulsivity in adults ($p<.001$). These data provide preliminary support for a developmental model of self-injury and self-restraint in which painful health conditions and compromised behavioral control influence the presence and trajectory of self-injury in ASD.

The prevalence of self-injury (SIB) in autism spectrum disorder (ASD) is significantly higher than that for individuals with intellectual disability of heterogeneous etiology, with estimates converging between 40% and 50% (Baghdadli, Pascal, Grisi & Aussilloux, 2003; Richards, Oliver, Nelson & Moss, 2012; Shattuck *et al.,* 2007). Despite the identification of ASD as a risk maker for SIB (McClintock, Hall & Oliver, 2003; Oliver & Richards, 2015) and the persistence of SIB in this population (Rice *et al.,* 2016; Richards, Moss, Nelson & Oliver, 2016), there has been limited research delineating characteristics associated with SIB in large cohorts of children and adults with ASD (Totsika & Hastings, 2009). These data are necessary in order to refine causal models of SIB for this high risk group. Guess and Carr (1991) proposed that SIB developed from repetitive behaviors through a process of social and non-social reinforcement. Oliver and Richards (2015) extended this model to integrate person characteristics that account for differences in the emergence, development and outcomes for SIB that are associated with individual difference. Oliver and Richards (2015) suggest causal roles for painful health conditions and impairments in behavioral inhibition in the etiology and trajectory of SIB. Whilst this model provides a useful theoretical framework to guide intervention, there is currently limited empirical evidence to evaluate the applicability of such a model to SIB in ASD.

Cross sectional studies of individuals with ASD have identified correlates of SIB including younger age (Esbensen, Seltzer, Lam & Bodfish 2009), greater impairment in adaptive skills (Baghdadli *et al.,* 2003; Duerden *et al.,* 2012; Richards *et al.,* 2012), greater degree of autism or social impairment (Baghdadli *et al.,* 2003; Duerden *et al.,* 2012; Richards *et al.,* 2012), abnormal sensory processing (Duerden *et al.,* 2012) and perinatal health conditions (Baghdadli *et al.,* 2003). There is emerging evidence that behavioral variables of overactivity, impulsivity and repetitive behavior correlate with the presence of SIB in individuals with ASD (Duerden *et al.,* 2012; Richards *et al.* 2012; Richman *et al.,* 2013) and predict the persistence of SIB longitudinally (Richards *et al.,* 2016). Whilst these results provide tentative support for the influence of person characteristics on the development of SIB in ASD, the putative demographic, behavioral and health correlates proposed by Oliver and Richards (2015) require evaluation in a sample with sufficient statistical power to model the independent contributions of each correlate to the presence and severity of SIB.

Painful physical health conditions have been proposed as one mechanism through which SIB may enter an individual's repertoire (Courtemanche, Black & Reese, 2016; Oliver & Richards, 2015). Empirical support for this casual association is drawn from the co-occurrence of health problems (such as otitis media, gastro-esophageal reflux or constipation) and SIB in individuals with intellectual disability (Carr & Owen-DeSchryver, 2007; Carr, Smith, Giacin, Whelan & Pancari, 2003; Christensen *et al*., 2009; Luzzani, Macchini, Valade, Milani & Selicorni, 2003; O'Reilly, 1997). Pain is conceptualized in the literature as a setting event for SIB (Carr *et al.,* 2003; Carr & Blakeley-Smith, 2006) and SIB has been hypothesized to 'gate' pain perception (Melzack & Wall, 1965). Whilst the association between pain and SIB has face validity and could lead to effective preventive and responsive interventions, there is currently no evidence associating painful health conditions with SIB in a cohort of individuals with ASD (de Winter, Jansen & Evenhuis, 2011). Thus, it is not possible to evaluate the plausibility of a causal role for pain and painful conditions in the development and maintenance of SIB in ASD.

In addition to evaluating the putative associations between pain and SIB, the identified correlations between SIB and impulsive/overactive and repetitive/restricted behaviors warrant further investigation. Attention deficit hyperactivity disorder (ADHD), and ADHD related

behaviors such as overactivity and impulsivity, are independently associated with SIB in individuals with intellectual disability (Arron, Oliver, Moss, Berg & Burbidge, 2011; Cooper *et al.,* 2009). Importantly, ADHD is thought to be underpinned by compromised development of inhibition, which, amongst other deficits, comprises the inability to prevent the initiation of a prepotent response to a stimulus and the inability to terminate an ongoing response (Nigg, 2005; Sonuga-Barke, 2002). Similarly, repetitive behaviors have been conceptualized as a deficit in executive function with response inhibition specifically implicated, which impacts on generating and controlling behavior (Turner, 1997; 1999). Thus, associations between SIB, repetitive behavior and impulsivity\overactivity might indicate a fundamental deficit in behavioral control. Drawing upon this neuropsychological interpretation, the presence of these behavioral correlates would predict more severe SIB that is either unrelated to environmental triggers, initiated more consistently in the presence of discriminative stimuli, or difficult for the individual to terminate (Oliver & Richards, 2015).

Key behaviors which indicate that SIB is difficult to control are, arguably, self-restraint and the preference for imposed restraint (see King, 1993). Self-restraint behaviors are those which involve the restriction of a person's body parts and/or movement through the use of clothing or material, the person's own body, or holding onto objects or others. Self-restraint is more common in males (Oliver, Murphy, Hall, Arron & Legget, 2003), in younger individuals (Fovel, Lash, Barron, & Roberts, 1989) and in those with a more severe intellectual disability (Fovel *et al.,* 1989). Importantly, self-restraint is associated with the presence of SIB in individuals with intellectual disability (Fovel *et al.,* 1989; Hyman, Oliver & Hall, 2002). The prevalence of self-restraint in those who engage in SIB is estimated at between 46 and 76 % (Oliver *et al.,* 2003, Powell, Bodfish, Parker, Crawford & Lewis, 1996). SIB significantly decreases when self-restraint occurs (Forman, Hall & Oliver, 2002; Kerth, Progar & Morales, 2009; Rojahn, Mulick, McCoy & Schroeder, 1978; Smith, Iwata, Vollmer & Pace, 1992) and consequently, self-restraint is conceptualized as a behavior exhibited by individuals in order to inhibit SIB.

King (1993) argues that the association between SIB and self-restraint provides evidence of a 'compulsive' nature of SIB. This hypothesis is supported by studies of individuals with Cornelia de Lange Syndrome (Hyman *et al.,* 2002) and adults with intellectual disability (Powell *et al.,* 1996), where sub-groups who engaged in both SIB and self-restraint displayed significantly more 'compulsive' behaviors than those who did not display SIB or self-restraint. This suggests the presence of 'compulsive' behavior moderates the relationship between self-restraint and SIB. In these preliminary studies and the model proposed by King (1993), 'compulsive' behavior was used to describe lining up, ritualistic behavior, spotless behavior and behaviors which could not be inhibited by the individual with no clear cognitive component. These may alternatively be conceptualized as 'impulsive' repetitive behaviors. Thus, the associations between SIB, self-restraint and 'compulsive' behavior further support a model of compromised inhibition in those who engage in SIB. However, thus far, there has been no investigation of the prevalence and topographies of self-restraint, or its interaction with SIB and variables indicative of impaired behavioral control (e.g., overactivity, impulsivity, repetitive behavior) in individuals with ASD.

In summary, there is a need to examine associations between behavioral and demographic characteristics and SIB in ASD, and the association between SIB and self-restraint in this group. In order to investigate these factors, a large sample of both children and adults with ASD will be assessed. This sample will provide the opportunity to evaluate putative associations between SIB, self-restraint, painful health conditions, repetitive behavior and

overactivity and impulsivity and thus the applicability of the model of SIB proposed by Oliver and Richards (2015) to individuals with ASD. The study aims to describe the prevalence, topographies and severity of SIB and the prevalence and topographies of self-restraint within and between children and adults with ASD. The following hypotheses are made:

i) Greater impairment in adaptive functioning, the presence of health problems, repetitive behavior, overactivity and impulsivity will be associated with the presence of SIB.
ii) SIB and behaviors indicative of impaired impulse control (overactivity, impulsivity and repetitive behavior) will be associated with the presence of self-restraint.
iii) The presence of overactivity and impulsivity will *predict* severe SIB. SIB and behaviors indicative of impaired impulse control will *predict* the presence of self-restraint.

Methods

Recruitment

This study was part of an audit of service need within the National Autistic Society (NAS). The NAS is the United Kingdom's largest provider of specialist ASD child and adult services. Service provision from the NAS necessitates an ASD diagnosis from a qualified medical professional, Psychologist, or Speech and Language Therapist and thus all included participants received a clinical diagnosis of ASD from one of these professionals. All National Autistic Society (NAS) schools and adult services were contacted and invited to participate using an opt-out consent procedure.

Procedure

All carers of individuals in the NAS adult services and schools received an information sheet detailing the study and the opt-out procedure. Questionnaire packs were then distributed to the schools and services and were completed by teachers or keyworkers who knew the participating individuals well. To avoid priming, the study was described as research into the behavior of children/adults with ASD. Schools and adult services returned completed questionnaires in a prepaid envelope. Ethical approval for this study was obtained from the School of Psychology ethical review committee at the University of Birmingham.

Participants

Data were collected on 515 individuals with ASD attending 12 NAS adult services and six NAS schools. The return rate was estimated at 60%. Questionnaires were excluded if: 1) 25% or more of the total items were incomplete (N=32), 2) participants were under the age of six and therefore had completed a differing assessment of ability level (N=2) or 3) age data were unavailable (N=57). This left a total of 424 individuals (82.3% of original sample; 208 < 18 years, 216 $\geq$ 18 years) for the analysis. All participants were between the ages of 6 and 61 years (mean age = 24.10; SD = 13.01) and 333 (78.5%) were male. Almost half of the sample were verbal (N = 208, 49.1%) and the majority of the sample had normal vision (N = 376, 88.7%), normal hearing (N = 405, 95.5%) and were ambulant (N = 392, 92.5%). Three items from the Wessex self-help score were included in the SAD-SQ to estimate ability (see measures). This was used to form a comparatively lower ability group for the purpose of analysis - those with some or substantial impairments in self-help skills (Score = 3 - 8) and a higher ability group - those without impairments in self-help skills (Score = 9). The categorical data showed that 188 (44.3%) participants comprised the lower ability group and 233 (55.0%) the higher ability group. Ability data were missing for 3 (0.7%) individuals.

Measures

The questionnaire pack comprised items regarding demographic information, the Self-injury, Aggression and Destruction Screening Questionnaire (SAD-SQ; Davies & Oliver, 2016), the Self-Restraint Checklist (Powell *et al.,* 1996) and the Challenging Behavior Questionnaire (CBQ; Hyman *et al.,* 2002).

Demographic information was collected on gender, age, ASD diagnosis, medication and contact with health professionals.

The SAD-SQ (Davies & Oliver, 2016) was developed as a screening measure to assess putative risk markers for challenging behavior. The measure was developed through a process of reviewing existing questionnaires measuring each putative risk marker that had been used previously with participants with an intellectual disability and had sufficient reliability and validity. These questionnaires were then systematically reduced so that the minimum number of items from each questionnaire was chosen whilst still reliably measuring the construct. The Ability subscale was measured via three items regarding adaptive functioning from the Wessex Behaviour Scale (Kushlick, Blunden & Cox, 1973). Four items from the Activity Questionnaire (Burbidge *et al.,* 2010) regarding difficulties waiting, wanting things immediately, acting as if driven by a motor, and finding it difficult to hold still were used to form the Overactive/Impulsive subscale. Two items from the behavior and emotional difficulties section of the Self-Help and Behaviour Rating Scale (Petty, 2006) regarding repetitive movements as well as obsessions and rituals formed the Repetitive/Restricted behavior subscale. This scale is an adapted version of the Wessex Behaviour Scale (Kushlick *et al.,*1973). Davies and Oliver (2016) report good reliability of the SAD-SQ. Additionally, Davies and Oliver (2016) demonstrate good concurrent and convergent validity of these behavioural risk markers through observational coding of naturalistic behavior, actigraphy and comparison to robust standardised measures. For example, participants classed as 'high risk' on the SAD-SQ, scored significantly higher on standardised measures of overactivity ($U = 33$, $p = .001$), impulsiveness ($Z = -2.727$, $p < .008$), repetitive ($U = .49$, $p = .003$) and restricted ($U = 61.5$, $p = .017$) behaviours than low risk participants (Davies & Oliver, 2016).

The Self-Restraint Checklist (Powell *et al.,* 1996) describes seven topographies of self-restraint and caregivers are asked to endorse whether the individual has displayed each of the behaviors. The scale has good inter-rater reliability of 91% (Powell *et al.,* 1996).

The CBQ (Hyman *et al.,* 2002) evaluates the presence of SIB, physical aggression, destruction of property and stereotyped behavior over the last month. The measure also examines eight topographies of SIB that were adapted from Bodfish *et al.* (1995). Items evaluating SIB only were used for the current study. Previous examination of the psychometric properties of the questionnaire has demonstrated good inter-rater reliability with reliability coefficients ranging from .61 to .89 (Hyman *et al.,* 2002).

Data analysis

Where multiple tests were conducted, the alpha level was set to $p < .01$. Data are presented for two groups; those under 18 (child sample) and those 18 years and over (adult sample). To investigate the prevalence and topographies of SIB and self-restraint, the percentage of the sample showing each behavior was derived from the SAD-SQ, CBQ and Self-Restraint Checklist respectively. Severity of SIB was derived from three items from the SAD-SQ. These items rate the frequency, management difficulties and concern caused by the SIB displayed. They were scored on a five point Likert scale ranging from zero (never, not

difficult, not at all concerning) to four (very often, extremely difficult, extremely concerning). Scores for concern about SIB correlated very strongly with the scores for frequency of SIB (r_s = .92, p <.001), as did the scores for management difficulties (r_s = .92, p <.001). Therefore, as frequency of SIB was a more accessibly defined construct, frequency was used as a proxy measure of severity. If an individual scored three or four on frequency, they were deemed to show severe SIB.

For all analyses of topography and severity, those showing the behavior were compared to the total sample of those not showing the behavior. For example, when identifying the prevalence of 'hits self with body part', the prevalence is calculated by comparing those showing the behavior to those who do not show any SIB *and* those who do not show this topography of behavior, but may display other topographies of SIB.

In order to investigate variables associated with the presence and severity of SIB, a number of categorical groups were created. For health problems, the sample was categorized into those displaying no health problems, and those displaying one or more health problems. For Ability, a median split was conducted on the SAD-SQ self-help score forming two groups; those with lower ability (score < 9) and those with higher ability (score = 9). A repetitive and restricted behaviors and interests (Repetitive/Restricted) composite was formed by summing two items in the SAD-SQ referring to repetitive movements and obsessions and rituals. The items were scored on a five point Likert scales ranging from 0 (never) to 4 (very often). A median split was conducted on the Repetitive/Restricted composite forming two groups; those without high levels of repetitive and restricted behaviors and interest (score < 4 on Repetitive/Restricted composite) and those with high levels of repetitive and restricted behaviors and interest (score $\geq$ 4 on Repetitive/Restricted composite). An overactivity and impulsivity (Overactive/Impulsive) composite was formed by summing four items in the SAD-SQ referring to overactive and impulsive behaviors. The items were scored on a five point Likert scales ranging from 0 (never) to 4 (very often). A median split was conducted on the Overactive/Impulsive composite forming two groups; those without high levels of repetitive and restricted behaviors and interested (score < 5 on Overactive/Impulsive composite) and those with high levels of repetitive and restricted behaviors and interests (score $\geq$ 5 on Overactive/Impulsive composite).

Relative risk analyses (with 99.9% confidence intervals), were conducted to appraise the associations between each variable and the presence and severity of SIB and the presence of self-restraint. Relative risks are deemed significant if the lower confidence interval is greater than one. In order to further investigate the associations between health problems and SIB, Chi Square tests were utilized in order to test for differences in specific forms of health problems.

Finally, in order to control for the overlap between variables in the relative risk analysis and to develop predictive models for the presence and severity of SIB and the presence of self-restraint, binary logistic regressions were conducted.

Results

Sample characteristics and age differences

Prior to investigating the aim of the study and testing the hypotheses, prevalence data for characteristics of the sample were generated for the child and adults samples. Table 1 displays the prevalence of males, those with lower ability, those with one or more health

problems, those with high Repetitive/Restricted behaviors and those with high Overactive/Impulsive behaviors for both groups.

++++++++++++Insert Table 1 about here++++++++++++++++

The results reveal that there were significantly more males in the child sample. There were significantly more individuals with lower ability, with one or more health problems, and significantly more individuals with high levels of Repetitive/Restricted behavior in the adult sample. There were no significant differences between the groups in the proportion of individuals with high levels of Overactive/Impulsive behavior.

Prevalence and topographies of SIB and self-restraint

In order to investigate the aim of the study, prevalence data were generated for the child sample and adult sample for SIB, severity of SIB; topographies of SIB, self-restraint and topographies of self-restraint (see Table 2).

++++++++++++Insert Table 2 about here++++++++++++++++

Table 2 reveals that 45.7% of the child sample and 49.1% of the adult sample engaged in SIB; 18% of the child sample and 19.9% of the adult sample engaged in severe SIB. For both groups, the most frequent topography of SIB was hitting self with a body part; the least frequent was hitting self with an object. There were no differences between the groups for prevalence, severity or topography of SIB. Table 2 also shows that 40.9 % of the child sample and 42.6 % of the adult sample engaged in self-restraint behavior. Additional analysis revealed that significantly more individuals with SIB engaged in self-restraint than those without SIB in both the child ($\chi^2(1) = 19.97$, $p < .001$) and adult ($\chi^2(1) = 21.55$, $p < .001$) samples. Significantly more children than adults engaged in a particular topography of self-restraint: holding onto others or holding onto others' clothing. Interestingly, this was the only topography of self-restraint that involved other people. The groups did not differ in prevalence of any other form of self-restraint. For both groups, the least prevalent topography of self-restraint was choosing orthoses. The number of topographies of self-restraint displayed by both groups was broadly similar.

Variables associated with SIB

In order to investigate the first hypothesis, relative risk statistics were calculated to assess the association between demographic and behavioral variables and the presence and severity of SIB. The results are shown in Figure 1.

++++++++++++++++Insert Figure 1 about here++++++++++++++

Figure 1 displays relative risk statistics for the child and adult samples for each variable and the presence and severity of SIB. Being male was not associated with SIB. Lower ability was associated with SIB for the adult sample, and with severe SIB for the child sample. Health problems were associated with SIB and severe SIB for the child sample. High levels of Repetitive/Restricted behavior were associated with SIB for the child sample and with severe SIB for both samples. High levels of Overactive/Impulsive behavior were associated with SIB, and severe SIB for both samples.

In order to further evaluate the relationship between health problems and SIB in the child population, Chi square tests were employed to assess the association between specific types

of health problems and presence of SIB. Table 3 displays the prevalence of health problems and Chi square tests for the child sample for each health problem and the presence of SIB.

++++++++++Insert Table 3 about here++++++++++++++++

The results in Table 3 reveal that there were significantly more skin and digestive problems in the sample of children who engaged in SIB. Additionally, the difference in levels of ear problems between the sample that engaged in SIB and the sample that did not approaches significance.

Variables associated with self-restraint

In order to test the second hypothesis of the study, relative risk indices were calculated; first investigating the relative risk of self-restraint given differing severity of SIB and second the relative risk of self-restraint given behavioral markers associated with SIB (Ability, Repetitive/Restricted and Overactive/Impulsive). As can be seen in Figure 2, all variables were significantly associated with self-restraint except for the presence of low ability in the adult sample.

++++++++++++++Insert Figure 2 about here++++++++++++++

Logistic regression for predictors of SIB and self-restraint

In order test the third hypothesis and to control for the overlap between variables and produce predictive models of SIB and self-restraint, binary logistic regressions were conducted. If the relative risk of SIB or self-restraint was significantly greater in participants with a given characteristic, then this characteristic was entered into the regression analysis as a predictor variable. Due to the differences in relative risk given the characteristics across age groups, predictive models were generated for the child and adult samples separately. All models for both children and adults were statistically significant, indicating that the models were able to distinguish between those displaying SIB and those not, those displaying severe SIB and those not, and those displaying self-restraint and those not. Table 4 reveals the results of the logistic regressions, and indicates the variables which made a significant independent contribution to each of the models. The results indicate that the models explained between 13% (Adult SIB model; Cox and Snell R square) and 39% (Child Severe SIB model; Nagel-kerke R squared) of the variance and correctly classified between 67.6% (Adult SIB model) and 84.9% (Child Severe SIB) of cases.

++++++++++Insert Table 4 about here++++++++++++++++

Figure 3 outlines the significant findings of these models. The logistic regressions revealed that, independently of other factors, children and adults with high levels of Overactive/Impulsive behavior were significantly more likely to show SIB. In addition to contributing to the presence of SIB, high Overactive/Impulsive behavior scores predicted increased likelihood of severe SIB in both the child and adult samples. Children with high levels of Repetitive/Restricted behavior were more likely to show SIB but high levels of Repetitive/Restricted behavior did not contribute to the adult model for presence of SIB. For the child sample, the presence of health problems significantly increased the likelihood of SIB and severe SIB. Low ability only contributed to the presence of severe SIB in the child sample.

Finally, children with high Overactive/Impulsive behavior scores, high Repetitive/Restricted behavior scores and SIB were more likely to display self-restraint whereas only the presence of SIB contributed to self-restraint in the adult sample.

++++++++++++++++++Insert Figure 3 about here++++++++++++

In summary, repetitive, overactive and impulsive behaviors, lower ability and the presence of health conditions predicted SIB and severe SIB. The presence of SIB, repetitive, overactive and impulsive behaviors predicted the presence of self-restraint.

Discussion

This study was the largest description of the prevalence and topography of SIB in a sample of children and adults with ASD from the United Kingdom with a novel, theory driven evaluation of self-restraint and painful health conditions in relation to SIB in ASD. Statistical evaluation of putative demographic and behavioral correlates of SIB and self-restraint afforded the opportunity to evaluate an explanatory model combining operant and neuropsychological theory to understand SIB and self-restraint in individuals with ASD (Oliver & Richards, 2015). Potential confounds of age, ability and heterogeneity were controlled through recruiting a large sample in which ASD diagnosis had been confirmed previously, conducting separate analyses for the child and adult samples, and including ability as a variable within analyses. The results confirm a high prevalence of SIB in individuals with ASD and provide preliminary support for the model proposed by Oliver and Richards (2015), by demonstrating the significant contribution of painful physical health conditions and behaviors indicative of compromised behavioral control to the presence and severity of SIB and self-restraint in individuals with ASD.

The results indicated a high prevalence of SIB (45.7% for children; 49.1% for adults), which is commensurate to the prevalence figures of between 40 and 50% reported in other samples with ASD (Baghdadli *et al.*, 2003; Duerden *et al.*, 2012; Richards *et al.*, 2012; Shattuck *et al.*, 2007). There were no significant differences in the prevalence or severity of SIB for the child and adult samples. Although limited conclusions can be drawn from cross sectional data, these findings support longitudinal research demonstrating the persistence of SIB in ASD (Rice *et al.*, 2016; Richards *et al.*, 2016) and may indicate that severity does not differ across age groups. There were no significant differences between the child and adult samples in the topography of SIB displayed, and, supporting previous findings, the most prevalent form of SIB was hitting self with a body part (Rice *et al.*, 2016; Richards *et al.*, 2012).

When investigating variables associated with SIB in ASD, gender was not associated with the presence or severity of SIB. This supports previous research (Bagdadli *et al.*, 2003; McClintock *et al.*, 2003, Richards *et al.*, 2012; Richards *et al.*, 2016) and indicates that in populations with ASD, being male does not increase the likelihood of SIB. Lower ability was significantly associated with SIB in the adult sample, but not in the child sample. However, the logistic regression revealed that lower ability was not independently predictive of SIB for adults. The relative risk analysis revealed that lower ability was associated with more severe SIB in the child sample, and the logistic regression confirmed an independent effect of lower ability for severe SIB. These results partially support previous findings associating lower ability and SIB in ASD (Bagdadli *et al.*, 2003; Duerden *et al.*, 2012; Richards *et al.*, 2012; Shattuk *et al.*, 2007). However, the results from this study are equivocal, with differing results dependent on age and severity of SIB. Further research utilizing robust cognitive measures of ability could usefully augment these results.

The presence of health problems was significantly associated with the presence and severity of SIB in the child sample. The logistic regression supported these findings with health problems predicting the presence and severity of SIB in the child sample. This novel finding extends previous research associating painful health conditions and SIB to individuals with ASD (Christensen *et al*., 2009; Luzzani *et al.,* 2003), and indicates a key area for intervention. Further analysis revealed specific associations between skin and digestive problems and SIB; the association between SIB and ear problems (e.g., ear infections) approached significance. These data provide valuable information regarding causal hypotheses of pain and SIB. Whilst skin problems may be a result of SIB (e.g., skin picking, scratching or biting), it is less likely that digestive problems and ear problems are a direct consequence of SIB. Therefore, it can be hypothesized that these specific health problems, which have also been identified in other populations as associating with SIB (digestive problems: Luzzani *et al.,* 2003; ear problems: O'Reilly, 1997) may be causally implicated in the development and maintenance of SIB. Although health problems were predictive of SIB in the child sample, they were not associated with SIB in the adult sample. The lack of association between health problems and SIB in the adult sample may be due to higher proportion of adults who experienced health problems compared to the child sample. Alternatively, the results may indicate developmental differences, with health problems implicated in the emergence of SIB (most likely in childhood) but not in the maintenance of SIB in adulthood. Thus, these data lend preliminary support to the hypothesis that painful health conditions provide one mechanism by which SIB can enter an individual's repertoire (Oliver & Richards, 2015); this hypothesis requires further investigation using longitudinal designs. Taken together, these novel findings associating painful health conditions and SIB in individuals with ASD suggest that pain may be causally implicated in SIB, particularly in children. Therefore, clinical evaluations of SIB should include a full health assessment in order to rule out or treat the influence of pain upon SIB.

High levels of repetitive/restricted behavior, and overactive/impulsive behavior were significantly associated with the presence of SIB in both samples. The logistic regression revealed that high levels of repetitive/restricted behavior predicted the presence of SIB in the child sample and the severity of SIB in the adult sample. The association between repetitive behavior and *severe* SIB supports an assertion that repetitive/restricted behaviors are not solely precursors that are shaped into SIB, but are also implicated in moderating the severity of SIB, perhaps through a fundamental difficulty in regulating behavior (Oliver, Petty, Ruddick & Bacarese-Hamilton, 2012). The association between SIB and behavioral characteristics was most consistent for overactive and impulsive behaviors, which significantly predicted the presence and severity of SIB for both groups. Taken together, these findings support those reported in previous research (Cooper *et al.,* 2009; Duerden *et al.,* 2012; Richards *et al.,* 2012; Richards *et al.,* 2016; Richman *et al.,* 2013). The consistent associations between ADHD like behaviors of overactivity and impulsivity with SIB lends tentative support to the model proposed by Oliver and Richards (2015) in which impairments in inhibition are implicated in the development, and trajectory, of SIB.

The results of the self-restraint analysis revealed that 40.9% of the child sample, and 42.6% of the adult sample engaged in self-restraint behaviors. There were no significant differences in the prevalence of self-restraint in the child and adult samples, which contrasts with previous findings indicating that lower age was associated with self-restraint (Fovel *et al.,* 1989). In line with previous research in individuals with intellectual disability, the presence of SIB was significantly associated with self-restraint in both child and adult samples (Fovel *et al.,* 1989; Hyman *et al.,* 2002). The results extend previous findings by demonstrating that

severe SIB was associated with self-restraint, suggesting an interaction between severity of SIB and self-restraint. These findings provide support for the theory that self-restraint is displayed by those who engage in SIB in order to reduce their SIB, particularly those showing severe SIB (Forman *et al.,* 2002; Kerth *et al.,* 2009; Rojahn, *et al.,* 1978; Smith *et al.,* 1992). Both the child and adult groups engaged in very low levels of seeking mechanical restraint; however this may be due to policy decisions regarding the utilization of restraints within the service. Interestingly, there were differences in the topographies of self-restraint displayed between the child and adult groups. The child sample engaged in significantly more 'holding onto others or holding onto others' clothing', than the adult sample, suggesting that perhaps in childhood, self-restraint behaviors are more dependent on external support than in adulthood. It is possible that gaining support from others provides the mechanism by which independent self-restraint enters a behavioral repertoire. It may be that parents and carers initially provide physical support and restraint for a child engaging in SIB, and through a process of learning the child eventually begins to engage in self-restraint. This warrants further investigation.

The results also revealed that lower ability was significantly associated with self-restraint in the child sample, and the confidence intervals for the adult sample approached significance. This finding supports previous research which associated greater level of intellectual disability with the presence of self-restraint (Fovel *et al.,* 1989). However, lower ability did not independently predict the presence of self-restraint in the final model for either sample. Similarly, high levels of repetitive and restricted behaviors were associated with self-restraint in both groups, but did not independently predict self-restraint for either sample. These findings highlight the importance of conducting statistical analyses which control for the interaction between variables. This is particularly important when the variables in question are known to interact with one another (e.g., age and ability; ability and SIB; SIB and self-restraint, ability and repetitive behavior).

Finally, high levels of overactive and impulsive behavior were significantly associated with self-restraint in both populations. This finding builds upon the reported association between compulsive behavior and self-restraint in individuals with intellectual disability (Hyman *et al.,* 2002; King, 1993; Powell *et al.,* 1996), and indicates that self-restraint may be associated with hypothesized impairments in behavioral control which contribute to the presence of SIB (Oliver & Richards, 2015). The logistic regression revealed that overactivity and impulsivity independently predicted the presence of self-restraint for the child sample. Overactive and impulsive behaviors did not predict self-restraint in the adult model; however the presence of SIB did. In interpreting these differences in findings, it is important to highlight that there were no significant differences in the presence of SIB, self-restraint or overactive and impulsive behaviors between the child and adult samples. Consequently, the differences in predictors of self-restraint can be viewed as potentially developmental differences.

These findings suggest that impaired behavioral control, as evidenced through impulsive and overactive behavior, may lead to the early development of self-restraint during childhood. This supports the assertion made earlier that self-restraint may not only function to inhibit SIB, but may help control a variety of behaviors. This theory would explain why SIB was not predictive of self-restraint in the child sample; it may be that during childhood, self-restraint occurs to control poor behavioral inhibition and is displayed in order to inhibit a range of behaviors, not specifically SIB. However, in adulthood, it is merely the presence of SIB, and not the presence of overactive and impulsive behaviors, that predicts the presence of self-restraint. It would seem that with development, self-restraint comes to function primarily to

reduce SIB, rather than as a response to general impairments in behavioral control. This may happen through a process of operant learning, as over time self-restraint is negatively reinforced by the avoidance of SIB, and is shaped into a functional behavior within an individual's repertoire. This tentative model requires significant further research, specifically detailing the development of SIB, self-restraint and behavioral inhibition. Ideally, this research would contain a behavioral test of inhibition in order to delineate the precise nature of impairment in inhibition. Results from this future research may open the possibility of interventions to improve behavioral control. It is plausible that an intervention for behavioral inhibition may result in a decrease in SIB, self-restraint behavior and overactive and impulsive behaviors.

The findings of this study have significant clinical implications. Taken together, the results suggest that SIB, self-restraint and overactive and impulsive behaviors may cluster together through the process of development. Additionally, the results implicate health problems in the development and maintenance of SIB. The identification of individuals at risk of SIB may be aided by attempts to identify those with high levels of health problems, overactivity, impulsivity and displaying self-restraint behaviors. In order to further support this model, future research should now progress to include longitudinal assessments of SIB and self-restraint, and their associated behavioral and demographic risk markers. Importantly, future research should also include an intervention component in order to provide causal evidence for these risk markers.

A number of possible caveats to these findings must be considered. There may be a sample bias as all children and adults were recruited through the NAS. However, the NAS is the largest specialist service provider for individuals with ASD in the UK, and the purpose of this study was to evaluate a model of SIB and self-restraint in ASD, the sample choice seems sensible. Additionally, the use of a screening tool prevented extensive evaluation of risk markers, thus impairments in cognitive ability were evaluated by proxy measure of adaptive functioning and constructs of repetitive/restricted behavior and overactive/impulsive behavior were drawn from a limited number of items. It was also beyond the scope of this study to evaluate all possible risk markers and protective factors and thus the relative contribution of communication and social skills deficits and the benefits of ongoing interventions were not evaluated. However, these limitations must be evaluated in the context of the very large and representative sample recruited through using a brief screening tool. Future research should seek to evaluate these risk markers with greater specificity in smaller samples to complement the large dataset reported in this study.

In summary, the results indicated that lower ability, health problems, overactive, impulsive, and repetitive behavior significantly independently predicted SIB and severe SIB. The presence of overactivity and impulsivity independently predicted self-restraint for children, and the presence of SIB independently predicted self-restraint for adults. A developmental model of SIB and self-restraint, underpinned by impairments in behavioral control is hypothesized to account for these findings.

Compliance with ethical standards:

Funding: This study was funded by Research Autism and Cerebra.

Ethical approval: All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Informed consent: Informed consent was obtained from all individual participants included in the study.

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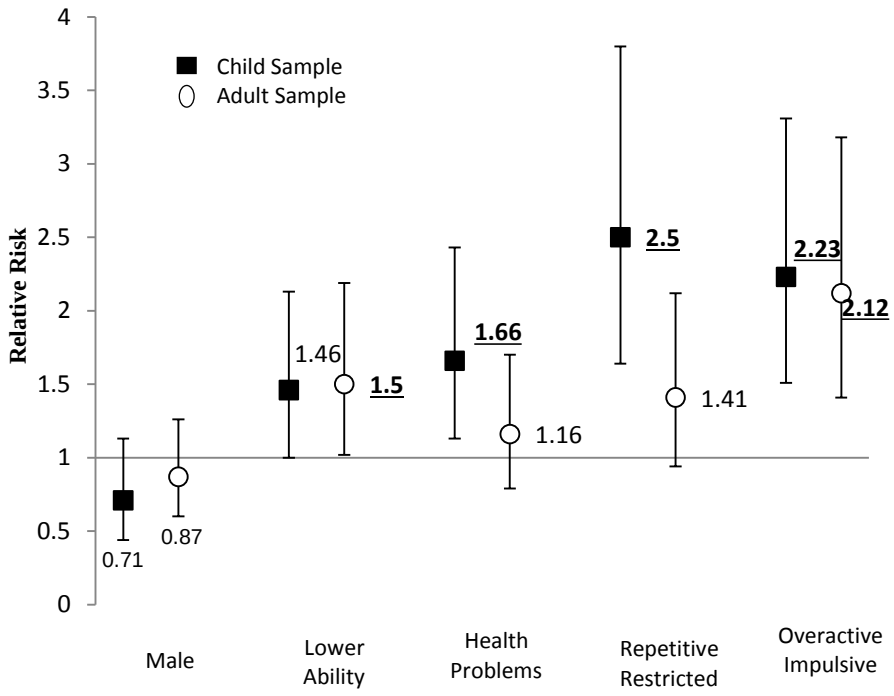
Figure Captions

Figure 1. Relative risk statistics (99% confidence intervals) for putative correlates of SIB and severe SIB in the child and adult samples. Significant relative risk statistics underlined.

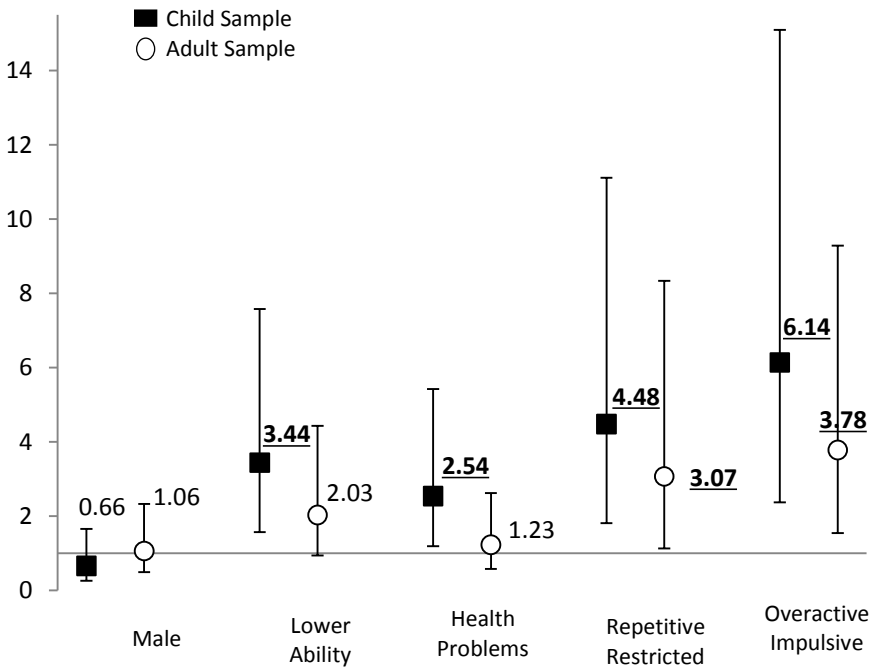
Figure 2. Relative risk statistics (99% confidence intervals) for putative correlates of self-restraint in the child and adult samples. Significant relative risk statistics underlined.

Figure 3. Significant independent variables predictive of SIB, severe SIB and self-restraint for the child and adult samples

Figure 1 Top



Putative Correlates of Self-injury



Putative Correlates of Severe Self-injury

Figure 2 Top

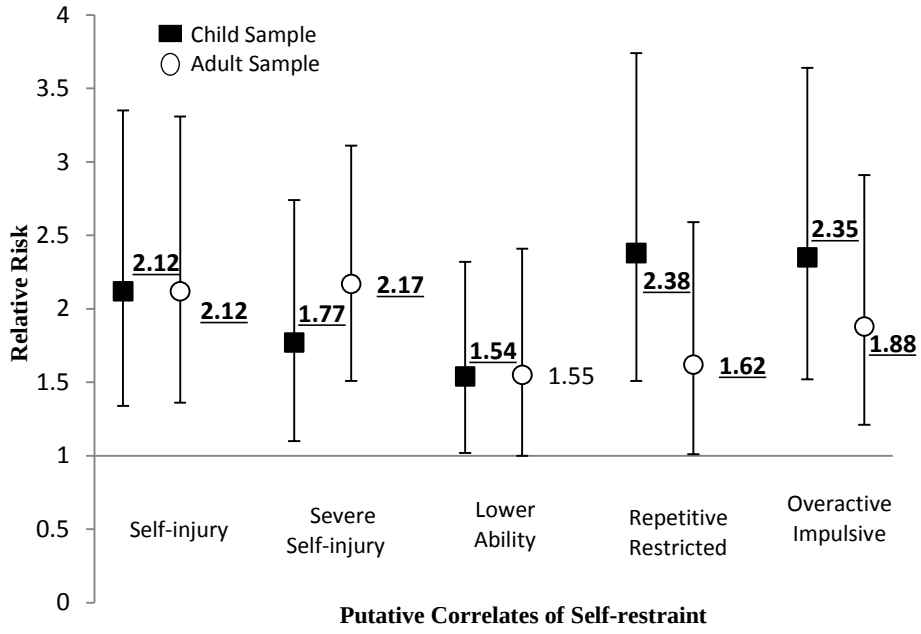

Child Sample
Adult Sample
Relative Risk
4
3.5
3
2.5
2
1.5
1
0.5
0
2.12
2.12
1.77
2.17
1.54
1.55
2.38
1.62
2.35
1.88
Self-injury
Severe Self-injury
Lower Ability
Repetitive Restricted
Overactive Impulsive
Putative Correlates of Self-restraint

Figure 3 Top

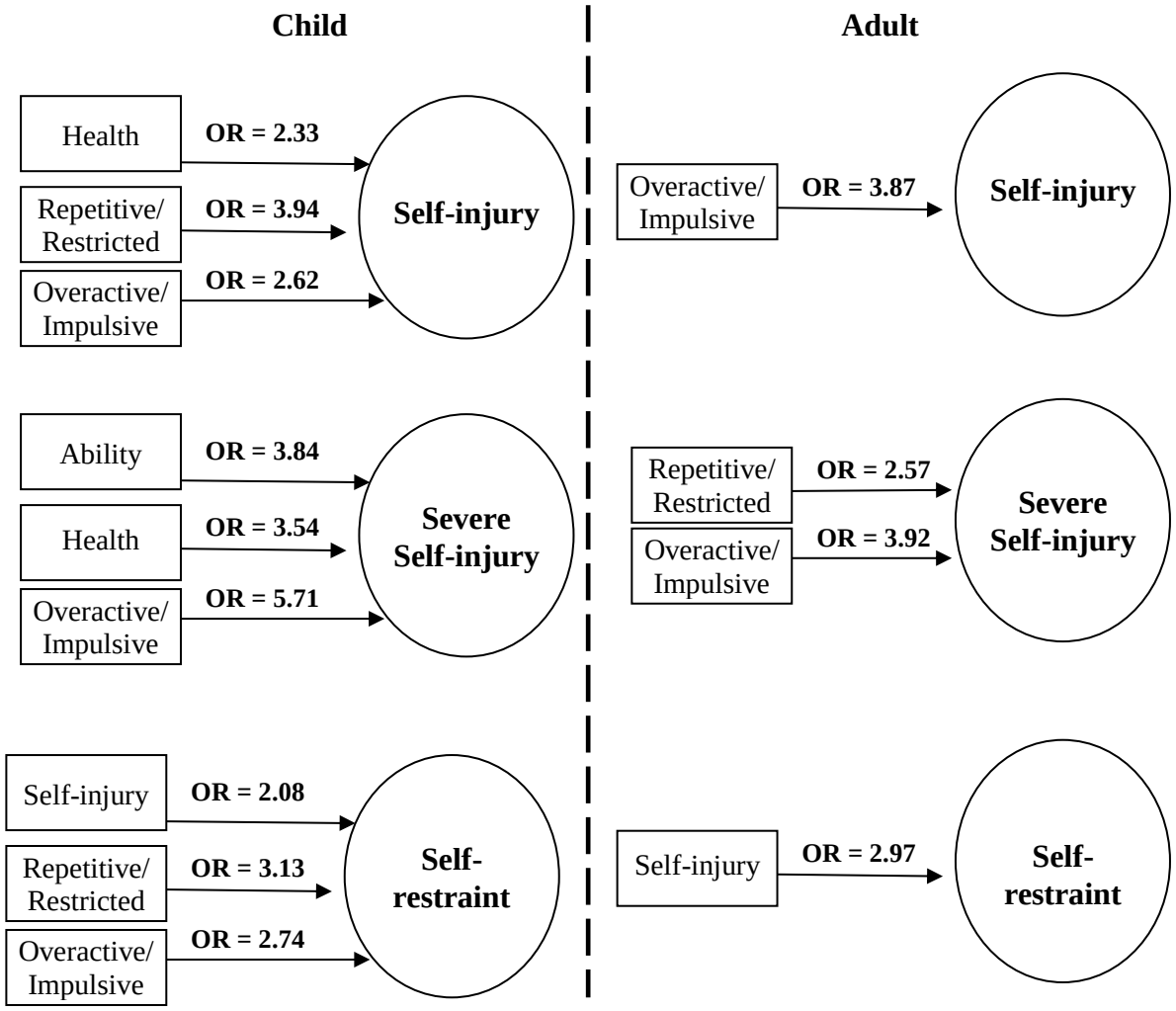

Child
Adult
Health
OR = 2.33
Repetitive/ Restricted
OR = 3.94
Overactive/ Impulsive
OR = 2.62
Self-injury
Overactive/ Impulsive
OR = 3.87
Self-injury
Ability
OR = 3.84
Health
OR = 3.54
Overactive/ Impulsive
OR = 5.71
Severe Self-injury
Repetitive/ Restricted
OR = 2.57
Overactive/ Impulsive
OR = 3.92
Severe Self-injury
Self-injury
OR = 2.08
Repetitive/ Restricted
OR = 3.13
Overactive/ Impulsive
OR = 2.74
Self-restraint
Self-injury
OR = 2.97
Self-restraint

Table 1. Demographic and behavioral characteristics for child and adult samples. Significant differences are highlighted in bold (p <.01; 2 tailed)

Characteristic	% (N)		Chi Square	*p* value
	Child (N = 208)	Adult (N = 216)		
Male	87.0 (181)	70.4 (152)	**16.81**	**<.001**
Lower ability	36.1 (75)	52.3 (113)	**12.30**	**<.001**
Health problems	38.5 (80)	61.1 (132)	**20.19**	**<.001**
High Repetitive/ Restricted	42.8 (89)	60.6 (131)	**14.48**	**<.001**
High Overactive/Impulsive	39.4 (82)	50.0 (108)	4.79	.029

Table 2 Prevalence, severity and topographies of SIB and self-restraint.

	Behavior	% (N) Child (N = 208)	% (N) Adult (N = 216)	Chi Square	*p* value
Presence of SIB	All SIB	45.7 (95)	49.1 (106)	0.43	.513
Severity of SIB	Severe SIB	18.8 (39)	19.9 (43)	0.49	.825
Topography of SIB	Hits self with body part	24.5 (51)	28.2 (61)	0.86	.354
	Hits self against surface or object	15.9 (33)	16.2 (35)	0.17	.897
	Hits self with object	6.3 (13)	2.8 (6)	2.94	.087
	Bites self	17.3 (36)	15.7 (34)	0.16	.687
	Pulls (e.g., hair or skin)	8.7 (18)	10.2 (22)	0.32	.571
	Rubs or scratches	11.1 (23)	15.3 (33)	1.74	.187
	Inserts finger or objects	6.3 (13)	5.1 (11)	0.25	.619
	Other (incl. cutting self, bending fingers)	3.8 (8)	3.2 (7)	0.10	.747
Presence of self-restraint	All self- restraint	40.9 (85)	42.6 (92)	0.03	.959
	Self-restraint if SIB occurs	*56.8 (54)*	*57.5 (61)*	-	-
	Self-restraint if SIB does not occur	*26.8 (30)*	*28.2 (31)*	-	-
Topography of self-restraint	Wraps self in own clothing	7.7 (16)	6.0 (13)	0.61	.453
	Holds onto others or holds onto others clothing	23.6 (49)	12.0 (26)	**10.84**	**.001**
	Positions self to restrain	6.7 (14)	4.2 (9)	1.56	.209
	Hold hands together, holds onto self	8.7 (18)	13.9 (30)	2.48	.115
	Holds or squeezes objects	17.3 (36)	16.7 (36)	0.12	732
	Chooses to wear a particular item of clothing most of the time	11.1 (23)	17.6 (38)	3.14	.076
	Chooses mechanical restraint	0.0 (0)	0.5 (1)	-*	1.00
	Other form of self-restraint	0.5 (1)	0.5 (1)	-*	1.00

* Fishers exact calculated as 50% of cells had expected count < 5.

Table 3. Prevalence and Chi square statistics for specific forms of health problems associated with the presence of SIB in the child sample. Significant Chi square statistics are highlighted in bold (p <.01; one tailed)

Health Problem	% (N) SIB (N = 95)	% (N) No SIB (N = 112)	Chi Square	P value
Eye Problems e.g., infections	3.2 (3)	3.6 (4)	-*	.500
Ear Problems e.g., infections	8.4 (8)	2.7 (3)	3.36	.034
Dental Problems e.g., cavities, gum problems	10.5 (10)	7.1 (8)	0.62	.215
Digestive Problems e.g., reflux, stomach problems	18.9 (18)	7.1 (8)	**5.83**	**.008**
Skin Problems e.g., eczema, dry skin	25.3 (24)	9.8 (11)	**7.93**	**.003**
Other Problems[1]	8.4 (8)	9.8 (11)	0.15	.352

* Fishers exact calculated as 50% of cells had expected count < 5

[1] Examples cited included dietary allergies, colds, hay fever, arthritis, scoliosis

Table 4. Logistic regression predicting likelihood of displaying SIB, severe SIB and self-restraint in the child sample and adult samples (bold text indicates predictor variables where p <.05)

Model	Chi Square	*Df*	*p*	Cox and Snell R square	Nagel-kerke R squared	Correct Classific-ation of cases (%)	Predictor Variables	*B*	S.E	Wald	*Df*	*p*	Odds Ratio	95.0% C.I. for Odds Ratio	
														Lower	Upper
Presence of SIB															
Child (N = 200)	51.22	3	<.001	.23	.30	76.0	**Health**	0.85	.33	6.50	1	**.011**	2.33	1.22	4.47
							Repetitive/ Restricted	1.37	.35	15.74	1	**<.001**	3.94	2.00	7.75
							Overactive/ Impulsive	0.96	.35	7.53	1	**.006**	2.62	1.32	5.20
Adult (N = 213)	28.80	2	<.001	.13	.17	67.6	Ability	0.41	.31	1.84	1	.175	1.51	0.83	2.75
							Overactive/ Impulsive	1.35	.30	19.83	1	**<.001**	3.87	2.14	7.03
Severe SIB															
Child (N = 192)	54.82	4	<.001	.25	.39	84.9	**Ability**	1.34	.45	9.10	1	**.003**	3.84	1.60	9.19
							Health	1.27	.44	8.25	1	**.004**	3.54	1.49	8.40
							Repetitive/ Restricted	0.90	.48	3.54	1	.060	2.46	0.96	6.29
							Overactive/ Impulsive	1.74	.48	13.00	1	**<.001**	5.71	2.22	14.72
Adult (N = 206)	23.31	2	<.001	.11	.17	79.1	**Repetitive/ Restricted**	0.95	.46	4.14	1	**.042**	2.57	1.04	6.39

							Overactive/ Impulsive	1.37	.42	10.50	1	**.001**	3.92	1.72	8.95
Self-restraint															
							SIB	0.73	.35	4.33	1	**.037**	2.08	1.04	4.13
							Ability	0.38	.35	1.22	1	.270	1.47	0.74	2.90
Child (N = 194)	43.09	4	<.001	.20	.27	73.7	**Repetitive/ Restricted**	0.76	.38	4.08	1	**.043**	2.13	1.02	4.45
							Overactive/ Impulsive	1.01	.36	8.00	1	**.005**	2.74	1.36	5.52
							SIB	1.09	.31	12.21	1	**<.001**	2.97	1.61	5.47
Adult (N = 205)	29.89	3	<.001	.13	.18	67.3	Repetitive/ Restricted	0.54	.33	2.69	1	.101	1.71	0.90	3.24
							Overactive/ Impulsive	0.60	.33	3.45	1	.063	1.83	0.97	3.46