SELF-INJURIOUS BEHAVIOUR IN AUTISM SPECTRUM DISORDER

by

Caroline Ruth Richards

A thesis submitted to the
University of Birmingham
for the degree of
DOCTOR OF PHILOSOPHY

ABSTRACT

Background: Self-injurious behaviour is reported to be common in autism spectrum disorder (ASD), with ASD frequently identified as a risk marker for self-injurious behaviour in those with intellectual disability. However, there are limited robust data detailing the prevalence, persistence and person characteristics associated with self-injury in ASD. Additionally, there has been limited application of operant theory to an understanding of self-injury in this population.

Method: Three large scale survey studies were employed to establish the prevalence, persistence and risk markers for self-injury in individuals with ASD compared to contrast groups. Experimental functional analyses were conducted with a subsample of participants, including a fine grained temporal analysis of behaviours associated with self-injury.

Results: Self-injurious behaviour was displayed by 50% of the ASD sample and was persistent over three years in 77.8% of the group. Self-injury was associated with significantly higher levels of autistic behaviour in individuals without idiopathic autism. The presence of self-injury was associated with significantly higher levels of impulsivity and hyperactivity, painful health conditions, repetitive behaviours and significantly lower levels of adaptive behaviour in individuals with ASD. An 'ASD weighted' operant function for self-injury was identified for the majority of children with ASD.

Conclusions: Self-injurious behaviour is prevalent and persistent in individuals with ASD. The presence of ASD phenomenology, rather than a diagnosis of idiopathic autism, should be considered a risk marker for self-injury. Person characteristics associated with self-injury in ASD indicate a role for repetitive behaviours, pain and impaired behavioural inhibition in the development and persistence of self-injurious behaviour. Self-injury is likely to be maintained by operant reinforcement in many individuals with ASD, however this may be through 'ASD weighted' reinforcement contingencies rather than the typically identified reinforcement contingencies. A pro-active early intervention strategy for self-injury in ASD, utilising existing intervention techniques is warranted.

ACKNOWLEDGEMENTS

First and foremost, my thanks go to Professor Chris Oliver for supervising this thesis. Thank you for giving me a brilliant opportunity and for supporting and encouraging me through it. You have inspired and challenged me in equal measure, through your unfailingly high standards and continued dedication to the children and families you work with. Thank you also to Research Autism for funding this project and to the National Autistic Society schools and adult services. Without the support of both of these fantastic organisations, this thesis would not have been possible. I am also incredibly grateful to all of the families and children who gave so generously of their time and effort in these projects.

Many thanks go to the team of researchers at the Cerebra Centre for Neurodevelopmental Disorders. It has been a pleasure working with so many diligent and compassionate people. I am very appreciative of the help I received from Emma Clarkson and Rachel Johnson on research visits. I am also hugely grateful to Lucy, Kate and Darrelle for supporting the data collection for these projects – you have all become fantastic friends as well as colleagues. Special thanks go to the original Office 128; Laurie, Jane, Jess, Dan and Andy, and to all of Office 517. It has been a privilege to work with you and I cannot wait to see how all of your research shapes the field of Psychology in years to come.

My personal thanks go to my Mum and Dad, for their continual love, support and prayers. Thank you for your faith and the support and time you have given to all aspects of my work. My final and most important thanks go to Darren, for his unfailing support. From beginning to end, you have always believed that I could do this. Thank you for your encouragement and for always finding the time to listen. I am so grateful for all that you have given over the last few years - only one more degree to go now, I promise!

DEDICATION

For my brother, David

My inspiration and motivation for this thesis

TABLE OF CONTENTS

Page

LIST OF TABLES

LIST OF FIGURES

CHAPTER 1

Self-Injurious Behaviour and Autism Spectrum Disorder: An Introduction

1.1 Introduction

This chapter provides an overview of concepts, models and research relevant to the epidemiological and experimental work described in this thesis. This chapter also comprises a brief introduction to autism spectrum disorder (ASD) and self-injurious behaviour. Empirical research delineating the epidemiology, associated person characteristics and function of self-injurious behaviour in individuals with ASD is then summarised. An evaluation of this research, key findings and areas for further investigation are highlighted, providing a rationale to the ensuing empirical work.

1.2 Autism Spectrum Disorder

1.2.1 Definition of autism spectrum disorder

Autism is a behaviourally defined neurodevelopmental disorder first described by Leo Kanner and Hans Asperger in the mid 1900's. The defining features of autism are described as a triad of impairments: abnormalities or impairments in social interaction and communication with accompanying restricted or repetitive behaviours, activities or interests. As autism continues to be diagnosed behaviourally, standardised criteria, as specified in the Diagnostic and Statistical Manual of Mental Disorders, 4th Revision, (DSM-IV; American Psychiatric Association, 1994) or the International Classification of Diseases, 10th Revision (ICD-10; World Health Organization, 1992), are typically employed. With increasing research evidence, autism has been conceptualised as a 'spectrum' disorder, allowing for differing presentation on a number of dimensions. Consequently, the current version of DSM-IV includes behavioural criteria for Autistic Disorder, Asperger syndrome (where there is no clinically significant delay in language or cognitive development) and Pervasive Developmental Disorder – Not Otherwise Specified (PDD-NOS; where criteria are not met for autistic disorder).

For the purposes of this thesis, the commonly employed term 'autism spectrum disorder' (ASD) will be used to refer to groups of individuals included in studies with a diagnosis of Autistic Disorder, Asperger syndrome or PDD-NOS. Where studies have specified the precise diagnostic classification of participants, that diagnostic term will be used.

1.2.2 Epidemiology of autism spectrum disorder

Early epidemiological estimates of ASD reported a prevalence of between '4 and 10 autistic children in every 10,000 live births' (Happe, 1994, p25). However, in the seminal Camberwell Study, Wing and Gould (1979) reported a higher incidence rate for the triad of impairments, of 21 in 10,000 individuals. A recent review of all published epidemiological studies between 1966 and 2010 reported figures much closer to those published by Wing and Gould (1979) (Saracino, Noseworthy, Steiman, Reisinger & Fombonne, 2010). Saracino *et al.,* (2010) identified 61 studies and reported prevalence rates of 22 in 10,000 for autistic disorder and 70 in 10,000 for PDD-NOS. Saracino *et al.* (2010) also reported an average male to female ratio of 4.4:1 for autistic disorder. These figures are consistent with the most recent population study conducted in England (Baird *et al.,* 2006) in which the prevalence of childhood autism was reported as 24.8 per 10,000 and the prevalence of ASD was reported as 77.2 per 10,000. Most studies across the world report an increase in prevalence of all categories of ASD (Saracino *et al.,* 2010; Western Australia, Nassar *et al.,* 2009; California, Hertz-Picciotto & Delwiche, 2009; Japan, Kawamura, Takahashi & Ishii, 2008). Whether this reflects an increase in the incidence of ASD, or changes in diagnostic practice and service provision, is still unclear.

1.2.3 Consequences of autism spectrum disorder

Whilst some individuals with ASD are able to live independently, the social and communication deficits associated with ASD and the high prevalence of co-morbid intellectual disability (Matson & Shoemaker, 2009), means that many individuals require care throughout their lifetime. Parents of children with autism tend to report more stress and mental health problems than parents of children with intellectual disability of heterogeneous aetiology (Olsson & Hwang 2001; Totsika, Hastings, Emerson, Lancaster & Berridge 2011). Similarly, mothers of children with autism report more stress than mothers of children with Down syndrome, even after controlling for differences in child social competence and behaviour problems and maternal age (Griffith, Hastings, Nash & Hill, 2010).

In addition to the consequences for those caring for individuals with ASD, there are significant clinical implications for the individuals themselves. The presence of ASD is a predictor of both inpatient hospital admission and psychotropic medication use (Cowley, Newton, Sturmey, Bouras & Holt, 2005; Tsakanikos, Costello, Holt, Sturmey & Bouras, 2007). Consistent with the increased level of stress and poorer mental health identified in carers, there is emerging evidence that individuals with ASD are at an increased risk of developing mental health problems. Whilst the research findings are less consistent in adult populations (Cooper & van der Speck, 2009; Underwood, McCarthy & Tsakanikos, 2010), the evidence is robust in children and adolescents with ASD. Young people with ASD display higher levels of psychopathology than individuals with intellectual disability, specifically an elevated prevalence of anxiety and mood disorders (Bradley, Summers, Wood & Bryson, 2004; Brereton, Tonge & Einfeld, 2006; Gillberg & Billstedt, 2000). Finally, there is evidence that individuals with ASD exhibit higher levels of challenging behaviour than those with intellectual disability of heterogeneous aetiology (Ando & Yoshimura, 1979a, Bhaumik, Branford, McGrother & Thorp, 1997; Bradley *et al.,* 2004; McClintock, Hall & Oliver, 2003; Totsika *et al.,* 2011).

In addition to the clinical implications, there are also national financial implications of caring for children and adults with ASD. The yearly costs of supporting children with ASD are estimated at £2.7 billion and the yearly costs of supporting adults with ASD are estimated at £25 billion (Knapp, Romeo & Beecham, 2009). The individual lifetime costs of caring for someone with ASD and an intellectual disability are estimated at £1.23 million. Given the significant clinical and financial implications, clinically relevant research which can impact on the lives of individuals with ASD and those caring for them is warranted.

1.2.4 Trends in autism spectrum disorder research

A recent review of trends in ASD research identified that 0.02% of 820 sampled papers investigated challenging behaviour in individuals with ASD (Matson & LoVullo, 2009). However, this review did not determine the precise proportion of articles that specifically investigated self-injurious behaviour. Seth and Oliver (In Prep.) conducted a comprehensive review of ASD publication research trends between 2001 and 2010, from which data regarding publications on self-injurious behaviour can be detailed. When key research themes were analysed in two data sets, the review revealed a broad increase in research regarding individuals with ASD, from 5,271 research papers in 2001-2005 to 13,222 research papers in 2006-2010. The majority of the research in both time periods was concerned with identifying causes for ASD and differences in brain structure and function. These two areas accounted for almost 80% of the research between 2001 and 2010. Importantly, there was a large increase in the proportion of research focusing upon brain structure and function, from 36% of research papers in 2001-2005 to 40% of research papers in 2006-2010. Whilst both casual models and neuroanatomy are important research areas, the findings from this research are unlikely to result in immediate clinical benefits for individuals and families currently living and working with the consequences of ASD. Unfortunately, research regarding clinically relevant areas with immediate applications such as sleep, diet, mental health and intervention, constituted a disproportionately small percentage of the research, with less than 10% of papers published between 2001-2010 focusing on these areas. Importantly for this thesis the proportion of research on challenging behaviour, and more specifically self-injury, decreased between 2001 and 2010. From 2001 to 2005 1.3% of the identified research was concerned with self-injury, however between 2006 and 2010 this figure decreased to 0.9%. Given that the total prevalence of individuals engaging in self-injury is likely to have remained stable between 2001 and 2010, this trend in research is worrying.

Whilst it is acknowledged that research is, by definition, driven by future implications, it is also critical that research conducted in clinical populations is responsive to current need. In the following sections, the implications of self-injurious behaviour and the epidemiology of self-injury in ASD populations will be reviewed with a view to evaluating whether the existing research trends are reflective of the clinical needs of carers and parents of, and individuals with, ASD.

1.3 Self-injurious behaviour

1.3.1 Definition of self-injurious behaviour

Self-injurious behaviour has been defined as

> "Any non accidental behaviour, initiated by the individual, which directly results in physical harm to that individual. Physical harm (includes) bruising, lacerations, bleeding, bone fractures and breakages, and other tissue damage" (Murphy & Wilson, 1985, p. 15).

Alternatively, self-injurious behaviour may be categorised by the topography of behaviour shown, for example:

> "head punching, hitting or banging against hard objects, self-biting, hitting other body-parts and skin picking/piercing" (Cooper *et al.,* 2009, p.2)

Within intellectual disability populations, the term self-injury typically refers to repetitive types of behaviour in contrast to acts such as self-cutting or burning, which may be displayed by typically developing individuals. Within this thesis a similar distinction will be drawn, where the term self-injury will be used to indicate non accidental repetitive behaviours defined by their topography.

1.3.2 Prevalence of self-injurious behaviour in populations with intellectual disability

Large scale population studies have identified that between 5 and 10% of individuals with an intellectual disability of heterogeneous aetiology engage in challenging behaviour (Emerson *et al.,* 1997; Lowe *et al.,* 2007). Oliver, Murphy and Corbett (1987) conducted the first comprehensive total population study of self-injury in individuals with an intellectual disability. The study sampled the South East Thames Regional Health Authority of England (population approximately 3.5 million) and found that 12% of their sample engaged in self-injurious behaviour. Oliver *et al.,* (1987) also demonstrated that self-injury increased in both prevalence and severity between the ages of 15 and 25. Later prevalence studies have broadly supported these results, with prevalence rates varying between 4% (Cohen *et al.,* 2010; Cooper *et al.,* 2009; Holden & Gitlesen, 2006) and 17% (Collacott, Cooper, Branford & McGrother, 1998). Lower prevalence figures are typically associated with more stringent definitions of self-injury (e.g. *actual* physical harm, rather than *potential* physical harm; Cooper *et al.,* 2009). Likewise, less conservative prevalence figures include less severe and less frequent occurrences of self-injury within the prevalence data (e.g., Collacott *et al.,* 1998). Generally, prevalence estimates of self-injury for individuals with intellectual disability are estimated to lie between 4 and 10%.

1.3.2.1 <u>Person characteristics associated with self-injurious behaviour</u>

In addition to the variation in prevalence rates accounted for by differences in definitions and survey methodology, there are a number of person characteristics associated with a higher prevalence of self-injury in individuals with intellectual disability. Demographic characteristics such as younger age prior to the decrease in prevalence after 25 (Emerson *et al.,* 2001a; Collacott *et al.,* 1998), poorer adaptive functioning (Baghdadli, Pascal, Grisi, & Aussilloux, 2003), poorer expressive language (Collacott *et al.,* 1998; Emerson *et al.,* 2001a; Murphy *et al.,* 2005) and the presence of painful health conditions (Carr & Owen-DeSchryver, 2007) are all associated with a higher prevalence or the presence of self-injurious behaviour. There is also an emerging literature demonstrating that behavioural characteristics such as repetitive behaviour (Collacott *et al.,* 1998; Emerson *et al.,* 2001a; Oliver, Petty, Ruddick & Bacarese-Hamilton, In Prep.) and overactivity and impulsivity (Collacott *et al.,* 1998; Schneider *et al.,* 1996) significantly increase the prevalence of self-injury. These behavioural and demographic characteristics will be further discussed in Section 1.5.1, with a specific focus on the evidence for an association with self-injury in those with heterogeneous aetiology of intellectual disability and those with ASD.

In addition to the evidence associating person characteristics with the presence of self-injurious behaviour, there are also data illustrating an association between genetic syndromes and the presence and severity of self-injury. Self-injurious behaviour has been identified to be more common in individuals with Lesch-Nyhan, Fragile-X, Cornelia de Lange, Cri du Chat and Smith-Magenis syndromes (Arron, Oliver, Moss, Berg & Burbidge, 2011; Christie *et al.,* 1982; Colley, Leversha, Voullaire & Rogers, 1990). The elevated prevalence of self-injury in these syndromes has afforded an opportunity to evaluate causal models for self-injury. Whilst there is evidence from some syndromes to support a biological model (e.g., Lesch-Nyan syndrome; Baumeister, Frye & Schroeder, 1985) the most convincing models draw upon a phenotype x environment interaction to account for the heightened prevalence and severity of self-injury (Langthorne & McGill, 2008; Oliver, 1993; Tunnicliffe & Oliver, 2011). Causal theories of self-injurious behaviour are discussed below.

1.3.3 Dominant causal theories of self-injurious behaviour

1.3.3.1 <u>Operant theories of self-injurious behaviour</u>

Operant theories suggest that self-injury is a functional, learned behaviour which is broadly ‘adaptive’ (Emerson, 1998). Self-injurious behaviour is conceptualised as being associated with environmental and/or internal stimuli in the form of antecedents and consequences. Antecedents, such as low levels of attention or demanding tasks, are theorised to occasion self-injury; reinforcement contingencies then maintain the behaviour. Positive reinforcement occurs when the presentation of stimuli increases the response strength of self-injurious behaviour; conversely negative reinforcement occurs when the removal of stimuli increases the response strength of self-injurious behaviour. Internal or automatic reinforcement contingencies also occur, where self-injurious behaviour is reinforced by a positive internal sensation (positive automatic reinforcement) or by the removal of an aversive internal sensation (negative automatic reinforcement).

The application of operant theory has two important corollaries regarding the study of self-injurious behaviour, relevant to the empirical work in this thesis. Firstly, an operant account posits that functional self-injury will vary systemically across environmental conditions, relative to the antecedents and consequences present. Secondly, the application of operant theory indicates that behaviours become organised in an individual's repertoire, dependant upon the consequences the behaviours evoke in the environment. Investigation of this behavioural organisation has led to the delineation of response classes, where temporally proximal behaviours are found to group into functionally equivalent classes (Baer, 1982). By definition, each behaviour in the class is evoked by the same antecedents and maintained by the same reinforcement contingencies. Behaviours within a response class may be organised in a hierarchy in which the initial response requires the least effort but is also the least likely to receive the reinforcing consequence. The systematic variation of behaviour across environmental conditions, and investigation of temporally organised response classes afford an opportunity to evaluate the utility of operant theory in explaining the presence, development and maintenance of self-injurious behaviour.

Operant theories of self-injury have been supported by experimental studies which employ manipulations of antecedents and consequences in order to identify systematic fluctuations in levels of behaviour (Carr & Durand, 1985; Iwata, Dorsey, Slifer, Bauman & Richman, 1994). Iwata *et al.* (1994) demonstrated that for 152 people with intellectual disability and self-injurious behaviour, 38% evidenced self-injury maintained by social negative reinforcement, 26% by social positive reinforcement, 21% by positive automatic reinforcement, 5% by multiple controlled reinforcement (positive and negative) and only 10% showed undifferentiated patterns. Carr and Durand (1985) used a similar methodology to that of Iwata *et al.* (1994), experimentally manipulating antecedents over a number of standardised conditions. Carr and Durand (1985) again demonstrated that self-injury was occasioned by environmental antecedents. The study also demonstrated that it was possible to reduce self-injury through the introduction of a functionally equivalent behaviour, occasioned and reinforced by the same antecedents and consequences as the self-injury. These original studies, using experimental functional analyses, have now been widely replicated and extended. A recent review of functional analyses describes the broad success of these experimental techniques in providing evidence to support the role of operant learning in the maintenance of self-injury (Hanley, Iwata & McCord, 2003). However, it should be acknowledged that the functional analysis literature may be biased by the selective submission and publication of assessment and intervention studies where the initial experimental functional analysis successfully identified maintaining antecedents and consequences.

In addition to explaining the maintenance of self-injury, operant theory has also been invoked to account for increases in the frequency and severity of self-injury. Hall, Oliver and Murphy

(2001) demonstrated that escalation in the early development of self-injurious behaviour was associated with an antecedent of low levels of adult attention. Oliver (1993) explains the evolution of self-injurious behaviour, from minor occurrences to severe self-injury, drawing on the application of operant theory. Delineating a principle of mutual reinforcement, Oliver (1993) suggests that damaging self-injurious behaviour, be it more severe, frequent or dangerous, is more aversive to caregivers. These behaviours are therefore more likely to be reinforced, and in turn are therefore more likely to occur again. This differential reinforcement leads to a cycle in which self-injurious behaviour can become increasingly frequent and severe.

Operant theories have been refined over time, delineating concepts such as mutual reinforcement (Hall & Oliver, 1992; Oliver, 1993), setting events (Bijou & Baer, 1978) and establishing operations (Michael, 1982) which add to an understanding of how self-injury may be maintained and shaped by environmental contingencies. However, although operant theories can explain the maintenance and changes in severity and frequency of self-injury, operant theory alone does not adequately account for the first episodes of self-injurious behaviour. In a significant theoretical development, Guess and Carr (1991) proposed a model which could account for the initial emergence and subsequent development of self-injury. They suggested a three stage model, in which self-injury develops from stereotypic and repetitive behaviours through a process of social and non-social reinforcement. At Stage One, repetitive behaviours emerge in young children with a self regulatory function. These rhythmic behaviours are initially not influenced by environmental factors, however they become sensitive to the environment during Stage Two. At Stage Two, stereotyped and repetitive behaviours modulate arousal levels; as environmental stimulation lowers arousal, rhythmic behaviours increase to compensate. Likewise, as environmental stimulation increases arousal, rhythmic behaviours decrease. Finally, during Stage Three, the rhythmic behaviours are shaped by social negative and positive reinforcement into self-injurious behaviour.

Although the model proposed by Guess and Carr (1991) is theoretically parsimonious, there has been little empirical evaluation. Richman and Lindauer (2005) conducted a small cohort study and provided some evidence for a longitudinal shaping of proto-injurious (or stereotyped) behaviours into self-injury. Similarly, Oliver, Hall and Murphy (2005) demonstrated that increases in early self-injurious behaviour over a two year period were correlated with social contact consistent with a social reinforcement paradigm. Finally, Petty, Allen and Oliver (2009) demonstrated temporal associations between repetitive, proto-injurious and self-injurious behaviours, indicating that these behaviours may be part of a single response class which has been shaped over time. However, whilst these studies provide preliminary support for Guess and Carr's model (1991), all three studies sampled relatively small cohorts. Additionally, Guess and Carr's model (1991) does not account for the association between person characteristics, such as lower ability or the presence of health problems, and self-injury, discussed in Section 1.3.2.1. Therefore, further empirical research which delineates associated person characteristics in larger samples is warranted, in order to evaluate the validity of this model in accounting for the emergence and development of self-injury.

In summary, operant theories can provide a useful explanation for the maintenance and changes in severity of self-injury. Further empirical research is needed to evaluate the application of operant principles to the early emergence and initial development of self-injury. It will be useful to consider principles from operant theory in relation to the development and maintenance of self-injury in individuals with ASD.

1.3.3.2 Neurobiological theories of self-injurious behaviour

There are three dominant theories regarding neurobiological differences in individuals who engage in self-injury. These theories have focused upon differences in neurotransmitters and nueromodulators; dopamine, serotonin and opioid peptides.

Self-injurious behaviour has been associated with disturbances in the basal ganglia dopaminergic system, which among many other functions, is hypothesised to be involved in regulating motor activities (Schroeder & Tessel, 1994). A recent review of animal models for self-injury highlighted the utility of work with the deer mouse which has provided evidence to support a dopaminergic theory of stereotyped and self-injurious behaviour (Schroeder, Loupe & Tessel, 2008). Substantial evidence for the role of dopaminergic involvement in self-injury comes from populations with Lesch-Nyan syndrome, with tangential evidence coming from typically developing individuals with Obsessive Compulsive Disorder (Schroeder *et al.,* 2001). Additionally, repetitive behaviours in ASD have been associated with decreased basal ganglia volume (Estes *et al.,* 2011). However, associations between self-injury and abnormalities in dopamine receptors and the basal ganglia are tentative and only occur under prescribed circumstances.

A second theory proposes the involvement of the serotoninergic system in self-injurious behaviour. Serotonin is known to be involved in arousal, appetite control, anxiety and depression, and there is some limited evidence that serotonin may be implicated in more 'obsessive' forms of self-injury (Bodfish *et al,* 1995; Kolevzon *et al.,* 2010; Thompson, Hackenberg, Cerutti, Baker, Axtell, 1994). However, there is currently no coherent explanation as to why differences in serotonin levels would impact upon self-injurious behaviour.

The most coherent neurobiological theory for self-injury involves the release of opioid neuromodulators such as β-Endorphin in response to self-injurious behaviour. Endorphins have analgesic and euphorogenic properties which are purported to lead to dependence. Drawing upon operant theories, it has been hypothesised that self-injury is positively reinforced by the release of endorphins (e.g., Sandman & Hetrick, 1995). This theory is supported by evidence that levels of endorphins are elevated in those who engage in self-injurious behaviour (Sandman, 1992; Sandman, Barron, Chicz-DeMet & DeMet, 1990). Concurrent evidence has also been reported in intervention studies which utilise Naltrexone and Naloxone, which both act as opiate blockers. These studies have demonstrated a decrease in self-injury for some individuals following drug administration (Barrett, Feinstein, & Hole, 1989; Sandman *et al.,* 1983; Sandman *et al.,* 2000). Finally, there is also emerging evidence that self-injury is, in some cases, targeted towards body sites that lead to a release in endorphins (Symons, Clark, Hatton, Skinner & Bailey, 2003; Symons & Thompson, 1997).

Although there is some convincing evidence for the involvement of opioid peptides in self-injury, the data and model are still limited. Naltrexone and Naloxone trials are only effective in a limited number of cases, and the model can not account for less severe self-injury where endorphin release is unlikely. Neither can the model account for self-injury directed towards body sites that do not lead to endorphin release, nor how self-injury initially entered the individual's behavioural repertoire. Support for all neurobiological theories is limited by the evidence that self-injury is often influenced by environmental contingencies. Consequently, more parsimonious neurobiological theories of self-injurious behaviour should seek to synthesise operant and biological accounts, clearly stating where one account or both accounts are most applicable, in order to provide a more comprehensive model (e.g., Oliver, 1993).

1.3.3.3 Pain related theories of self-injurious behaviour

Individuals with intellectual disabilities have been reported to experience a high prevalence of painful health problems (Berg, Arron, Burbidge, Moss & Oliver, 2007, Haveman *et al.,* 2000; Jansen, Krol, Groothoff, & Post, 2004; van Schrojenstein Lantman-De Valk *et al.,* 2000; van Schrojenstein Lantman-de Valk, Linehan, Kerr & Noonan-Walsh, 2007), with pain experiences increasing proportionally with the level of cognitive impairment (Breau, Camfield, McGrath & Finley, 2003). However, there is also evidence that pain and painful health conditions are rarely identified and treated in individuals with intellectual disability (Howells, 1986; Stallard, Williams, Lenton & Velleman, 2007). Several single case and small sample cohort studies have identified an association between pain and self-injury in individuals with intellectual disabilities. Self-injurious behaviour was shown to be associated with the presence of gastro-oesophageal reflux in individuals with Cornelia de Lange syndrome (Luzzani, Macchini, Valade, Milani & Selicorni, 2003). A single case study using a validated measure of pain related behaviour (Non-Communicating Children's Pain Checklist-Revised, NCCPC-R; Breau, Finley, McGrath & Camfield, 2002) demonstrated that severe self-injurious behaviour was significantly associated in time with higher levels of pain related behaviour (Symons & Danov, 2005). Similarly, O' Reilly (1997) and Christensen *et al.*

(2009) report case studies of children with intellectual disabilities who display self-injury associated with otitis media and constipation. Two cohort studies present data associating physical illness (e.g., coughs, colds, constipation, cuts, ear infections etc.) and self-injury, and menstrual discomfort and self-injury (Carr & Owen-Deschryver, 2007; Carr, Smith, Giacin, Whelan & Pancari, 2003). Finally, in two recent studies screening for risk markers[1] of challenging behaviour, the presence of one or more health problems was significantly correlated with the presence and severity of self injury in children aged between two and twelve (Davies, 2010) and with the presence of self-injury in children under five (Petty, Bacarese-Hamilton & Oliver, In Prep).

In addition to studies associating the presence of self-injury with pain, there is also a small body of evidence demonstrating that treating pain leads to a reduction in self-injury. Bosch, Van Dyke, Smith and Poulton (1997) present data on a cohort of seven children for whom previously unidentified health conditions were treated with medical intervention. For six of the seven children, medical treatment resulted in a significant reduction in self-injury. Christensen *et al.*'s (2009) study also demonstrated that medical intervention to reduce chronic constipation reduced levels of self-injurious and aggressive behaviours to near zero. Further large scale studies, evaluating self-injury following medical treatment for pain, are required in order to extend these preliminary findings. Taken together, studies associating pain and self-injury and studies detailing reductions in self-injury following treatment for pain, suggest that pain may be causally implicated in the development and maintenance of self-injury.

[1] Throughout the thesis, the term 'risk markers' is used to describe those attributes which are associated with an increased likelihood of self-injurious behaviour. They are differentiated from risk factors, in which the attribute contributes causally to the likelihood of self-injury.

The putative association between pain and self-injury may be accounted for in a number of different ways. One hypothesis is that specific sites of pain or painful health conditions occasion self-injury directly at the location of pain. Thus, self-injurious behaviour may directly function to remove the painful stimuli e.g., scratching at the site of skin conditions or finger poking in the ear during an ear infection. Supporting this hypothesis, Mailis (1996) presents four case studies of self-injury in typically developing individuals experiencing pain. In all four cases, the self-injury was targeted toward the symptomatic body site. In this cohort study, Mailis (1996) notes that the self-injury was intermittent and associated with episodic increases in pain, which supports a hypothesis that the self-injurious behaviour directly functions to remove the painful stimulation or the assumed cause of pain.

Linked to the first hypothesis is a second model in which self-injury acts to 'gate' pain. Melzack and Wall's (1965) gate control theory of pain suggests that individuals may engage in self-injury at one body site in order to gate the pain experienced at another body site. It could be hypothesised that in these cases, self-injury would be less random and more focused towards body sites that effectively gate chronic pain. In support of this, Breau *et al.* (2003) demonstrated that children with chronic pain conditions (e.g., gastrointestinal reflux, musculoskeletal conditions, chronic ear infections) engaged in more 'targeted' self-injury than those without chronic pain conditions. Typically, those with chronic pain engaged in self-injury across significantly less body surface and across significantly fewer body sites. Both of these hypotheses present a model with face validity that may explain how self-injury enters into an individual's behavioural repertoire. It is highly likely that these models may interact with an operant model; that once established in an individual's behavioural repertoire through pain, self-injury becomes sensitive to environmental reinforcement. This account would be compatible with the theory proposed by Guess and Carr (1991), with painful health conditions influencing the emergence of self-injury, alongside or instead of the shaping of repetitive behaviours.

A final model detailing the interaction between pain and self-injury is that of pain as a setting event (Carr & Smith, 1995). As a setting event, pain from these conditions is hypothesised to increase the aversive properties of an antecedent such as a task demand, and therefore increase the likelihood of self-injury being displayed in response to the antecedent. Carr *et al.* (2003) and Carr and Blakeley-Smith (2006) present robust studies evidencing pain as a setting event for self-injury occasioned by tasks. Carr *et al.* (2003) demonstrate that severe self-injurious and aggressive behaviours occurred more frequently in a cohort of four women with intellectual disability when menstrual related pain and demanding tasks co-occurred. Similarly Carr and Blakeley-Smith (2006) demonstrated that challenging behaviour in a cohort of 21 children increased during demand conditions when the children were unwell. In both studies, a combination of medical and behavioural intervention effectively reduced self-injurious behaviour.

Despite the emerging evidence presented from case and cohort studies, and the hypothesised models associating pain with self-injury, there are few large scale studies which validate the association between self-injury and pain (de Winter, Jansen & Evenhuis, 2011). This may be due to methodological difficulties intrinsic in the study of pain and self-injury. Self-injurious behaviour is highly associated with limited expressive language abilities (McClintock *et al.,* 2003). Self-report, is the ‘gold standard’ for pain assessment and consequently it is unsurprising that pain is rarely diagnosed or treated in individuals with an intellectual disability (Stallard *et al.,* 2007). In order for our understanding of the role of pain in self-injury to move forward, effective methods of assessing pain in non-verbal populations must be identified. Promising research methods have been developed using third party report of individual's facial expressions in order to identify undiagnosed pain (LaChapelle, Hadjistavropoulos & Craig, 1999; Messmer, Nader & Craig, 2008; Nader, Oberlander, Chambers & Craig, 2004). However, this has methodological difficulties, being heavily reliant upon the carer of the individual with an intellectual disability noticing potentially small variations in facial expression. It also requires extensive observations, ideally incorporating measures to independently confirm the presence of pain, which limit the potential sample size.

In order to further evaluate the role of pain in influencing self-injury, a more robust method may be to examine associations between identified health conditions which are known to be painful (e.g., eye and ear infections, eczema, gum disease, reflux) and self-injury (e.g., Carr & Owen-DeSchryver 2007; Davies, 2010; Petty, Bacarese-Hamilton, Oliver, In Prep). Whilst this method may underestimate the presence of pain in individuals with intellectual disabilities, it will provide a conservative measure of a high probability correlate of pain occurrence and a method through which associations between pain and self-injury can be assessed in larger populations.

1.3.4 Consequences of self-injurious behaviour

Despite the contrasts in causal explanations for the presence and development of self-injury, research findings are broadly consistent when detailing the consequences of self-injurious behaviour. Self-injury is reported to have a significant impact upon the individual who displays the behaviour and upon those caring for the individual. Self-injury in children with

ASD significantly increases the child's risk of psychiatric hospitalisation (Mandell, 2008) and hospital treatment for injury (McDermott, Zhou & Mann, 2008). The presence of self-injury is known to increase the likelihood of reactive physical interventions and emergency medication use (Allen, Lowe, Brophy & Moore, 2009). Both ASD and challenging behaviour independently contribute to lower quality of life scores for individuals (Beadle-Brown, Murphy & DiTerlizzi, 2009). Parents of children with ASD are significantly more likely to be a member of support group if their child shows self-injurious behaviour (Mandell & Salzer, 2007). Mothers of children with autism are significantly more likely to experience stress if their child displays challenging behaviour (Hastings, 2003) and are more likely to experience dysregulation of the levels of their stress hormone, cortisol (Seltzer, *et al.,* 2010). The effects of self-injurious behaviour have bi-directional consequences for both parents and children (Greenberg, Seltzer, Hong, & Orsmond, 2006) and also contribute to staff burnout in care settings (Hastings & Brown, 2002). In summary, the effects of self-injury in individuals with ASD are pervasive and damaging.

1.4 Self-Injurious Behaviour in Autism Spectrum Disorder: Epidemiology

Given the significant social and economic implications of both ASD and self-injurious behaviour detailed above, there is a pressing need to determine the prevalence, severity and persistence of self-injury in ASD populations. The following sections describe and evaluate the research literature published to date, detailing the size and scale of the problem of self-injury in ASD samples.

1.4.1 Prevalence

Studies reporting prevalence data for self-injurious behaviour in ASD were identified through literature searches of major scientific databases using key terms including 'self-injury', 'self-injurious behaviour', 'self-mutilation', 'ASD' and 'autism'. In order to ensure a breadth of literature was considered, hand searches of key journals were also conducted. Finally, reference sections of sourced papers were examined for further literature. A total of 17 studies were identified which reported, or provided the data necessary to ascertain, the prevalence of self-injury in a sample with ASD. Table 1.1 describes the articles; their sample sizes and demographics, the ability level of the sample and the measures used to define ability, the ASD diagnoses of the sample, and the measures used to define diagnosis, the measurement of self-injurious behaviour and the reported or calculated prevalence of self-injury.

Table 1.1. Sample size, demographic information and prevalence of self-injury in identified research papers

Authors	N	Age in years Mean *(SD)* Range	% Male	Ability measure	% with ID (IQ<70)	Diagnostic measure	Diagnoses included	SIB frequency/ severity	SIB Prevalence
Ando & Yoshimura (1979a; 1979b)	47	No mean/SD 6.0 – 14.0	83.0	Performance scale of WISC	95.7	Professional diagnosis	Autism	Presence or absence of SIB	42.6
Baghdadli *et al.,* (2003)	222	5.0 *(1.2)* 2.0-7.0	(M:F) 4.7:1	Developmental Quotient from Vineland	96.2	ICD-10 criteria	Infantile autism	Presence or absence of SIB	53.0
Baghdadli *et al.,* (2008)	185	8.0 *(1.3)* 5.0 – 10.0	80.2	Developmental Quotient from Vineland	95.5	Childhood Autism Rating Scale	Pervasive Developmental Disorder	Presence or absence of SIB in last 15 days	32.7
Bartak & Rutter, (1976)[2]	19 17	No mean/SD <11 No mean/SD <11	100.0 100.0	Non-verbal tests from Merril-Palmer Scales of Mental Tests or WISC	0.0 100.0	A definition of autism was used for both groups	Autism	SIB ever shown	31.6 70.6
Bradley *et al,* (2004)	12	16.33 *(2.2)* No range	66.7	WAIS –R, WISC-R, Merril Palmer Scales of Mental Tests, VABS	100.0	ADI-R	Autism	Score above clinical cut off on DASH II	58.0

[2] Two samples included in this study, one with IQ > 70, and one with IQ< 70. Results from both samples are reported here

Table 1.1 Cont. Sample size, demographic information and prevalence of self-injury in identified research

Authors	N	Age in years Mean *(SD)* Range	% Male	Ability measure	% with ID (IQ<70)	Diagnostic measure	Diagnoses included	SIB frequency/ severity	SIB Prevalence
Billstedt, Gillberg & Gillberg, (2005)[3]	120	Not reported	70.0	Tested age <10 with neuropsychological developmental or social development test	81.7	DSM-III-R criteria Handicaps, Behaviours and Skills Schedule Autistic Behavior Checklist	Autistic disorder Infantile autism Autistic-like conditions Atypical autism	Moderate or severe SIB shown in lifetime	50.0
Cooper *et al.*, (2009)	77	Not reported	Not reported	*C21st Health Check*	Not reported	*C21st Health Check* Clinical assessment by Consultant Psychiatrist	Autism	SIB that requires clinical assessment/ intervention and causes tissue damage	13.0
Dominick *et al.*, (2007)[4]	54	91.2 *(29.8)* No range	87.0	Pre-school or school age differential ability scales	Mean IQ = 81.0	DSM-IV criteria ADI-R score ADOS score	ASD	SIB present for at least three months in lifetime	32.7

[3] ID and diagnostic classifications based upon total sample at T1 (N=120), but SIB prevalence based upon sample at T2 (N=108)
[4] Age data reported in months

Table 1.1 Cont. Sample size, demographic information and prevalence of self-injury in identified research

Authors	N	Age in years Mean *(SD)* Range	% Male	Ability measure	% with ID (IQ<70)	Diagnostic measure	Diagnoses included	SIB frequency/ severity	SIB Prevalence
Janicki and Jacobson (1983)	314	≥ 21	72.0	None reported	85.0	None reported	Autism	None reported	20.0
Lecavalier (2006)	487	Not reported	82.6	Scales of Independent Behavior - Revised	66.0	Prior diagnosis by professional or school district	Pervasive Developmental Disorders	Occurring often/ moderate problem/ occurring a lot/severe problem	Parent = 11.0 Teacher = 10.3
Matson *et al* (1996)	185	37.94 *(12.84)* No range	64.0	American Association on Mental Retardation criteria	100.0	DASH-II	PDD Autism	Score above clinical cut off on DASH II	34.0
McTiernan *et al* (2011)[5]	174	8.00 *(2.38)* 3.0 – 14.0	82.0	None reported	87.0	DSM IV	Autism	Occurred in last two months; causes damage	48.9
Murphy, Healy & Leader (2009)[6]	157	8.5 *(2.17)* 3.0–14.2	82.8	None reported	79.8	None reported Participants all attended ABA or ASD schools/units	ASD	Occurrence of monthly or more	45.2

[5] ID data only reported for 63.2% (N = 110) of sample
[6] ID data only reported for 69.4% (N=109) of sample

Table 1.1 Cont. Sample size, demographic information and prevalence of self-injury in identified research

Authors	N	Age in years Mean *(SD)* Range	% Male	Ability measure	% with ID (IQ<70)	Diagnostic measure	Diagnoses included	SIB frequency/ severity	SIB Prevalence
Poustka & Lisch (1993)	61	15.3[7] 5.00 – 33.00	82.6	German version of WAIS/WISC	78.3	ADI score ADOS	Autism	Presence or absence of SIB	52.5
Richler *et al* (2007)[8]	165	29.41 *(4.86)* No range	84.4	Mullen Scales of Early Learning	Mean IQ = 49.44	ADI-R score ADOS if necessary	Autism PDD-NOS	Presence or absence of SIB	29.7
Seltzer *et al.*, (2010)	86	24.7 *(7.24)* 18.0-53.0	79.1	Wide Range Intelligence Test (WRIT) Vineland Screener	57.0	Professional diagnosis and ADI-R score	Autistic disorder Asperger's Disorder PDD-NOS	SIB shown at least once during eight day study	24.0
Shattuck *et al.*, (2007)	241	22.0 *(9.7)* 10.0 – 52.0	75.5	Vineland Screener WRIT Clinical consensus Review of history	68.5	Professional diagnosis and ADI-R score	Autistic disorder Asperger's Disorder PDD-NOS	SIB shown in last six months	46.1

[7] Median age
[8] Age data reported in months

The prevalence rates displayed in Table 1.1 indicate that self-injury is relatively common in individuals with ASD, with a large proportion of the papers estimating the prevalence of self-injury between 40 and 60% (Ando & Yoshimura, 1979a; 1979b; Baghdadli *et al.,* 2003; Bradley *et al,* 2004; Billstedt, Gillberg & Gillberg, 2005; McTiernan, Leader, Healy & Mannion, 2011; Murphy, Healy & Leader, 2009; Poustka & Lisch, 1993; Shattuck *et al.,* 2007). These figures are significantly higher than those reported in populations with intellectual disability of heterogeneous aetiology (Cohen *et al.,* 2010; Cooper *et al.,* 2009; Holden & Gitlesen 2006; Oliver *et al.,* 1987), and indicate that there is an association between ASD phenomenology and self-injurious behaviour. However, as can be seen in the table, there is also significant variation in the reported prevalence rates, ranging from 10.3% to 70.6%. Given the heterogeneity of ASD populations, it is likely that the variation in prevalence rates is due to a combination of differences in sample characteristics including level of intellectual disability and inclusion of individuals with genetic syndromes, as well as differences in sample sizes, time periods of self-injury specified and definitions of self-injury.

Of the 17 studies, five did not conduct an independent assessment of ASD and relied upon previous clinical diagnoses for their inclusion criteria. As ASD is diagnosed solely through behavioural markers, it is essential that studies reporting prevalence data ensure that all individuals within the study meet diagnostic criteria for ASD. Failure to do this, threatens the external validity of the study, and renders the reported prevalence rates unusable for generalisation. Several international ASD diagnostic tools and screening measures are now available (e.g., Autism Diagnostic Observation Schedule, Lord *et al.,* 2000; Autism Diagnostic Interview Revised, Lord, Rutter & Lecouteur, 1994; Social Communication Questionnaire; Rutter, Bailey & Lord, 2003) with well established reliability and validity.

Future studies should seek to conduct direct observation, interviews or questionnaires in order to ensure the homogeneity of ASD diagnoses within their samples.

Interestingly, most studies contained a majority of individuals with an intellectual disability (with the exception of the subsample included in Bartak and Rutter's study, 1976 and the study conducted by Cooper *et al.,* 2009, where no data are provided on the intellectual function of the ASD sample). Consequently, this increases the homogeneity across the samples and removes a key confound that may have contributed to the variation in reported prevalence rates. As with ASD diagnosis, assessment of intellectual disability was conducted with varying degrees of rigor across the studies. It is likely that this is due to inherent difficulties in comprehensively assessing intellectual disability when utilising survey methods. Studies that employed robust neuropsychological tests of intellectual ability have a significantly smaller sample (e.g., Bradley *et al,* 2004), and thus there appears to be a trade off between sample size and quality of assessment. One possible strategy to overcome this may be to assess adaptive behaviour through a screening tool, as a proxy measure of intellectual ability (e.g., Baghdadli *et al.,* 2003). This may allow for assessment of larger sample sizes, whilst collecting robust data regarding adaptive functioning. Given the widely reported association between degree of disability and challenging behaviour (McClintock *et al.,* 2003) it is critical that assessment of intellectual disability or adaptive functioning is a core component of any epidemiological study of self-injury in ASD.

An additional factor that may contribute to the divergence in reported prevalence rates are the different definitions of self-injurious behaviour. The studies which reported a higher prevalence of self-injury predominantly reported the presence or absence of self-injury without a specific time period (Ando & Yoshimura, 1979a; 1979b; Baghdadli *et al.* 2003; Bartak & Rutter, 1976; Poustka & Lisch, 1993). This may be better conceptualised as a lifetime prevalence estimate. There is a broad trend towards a decrease in prevalence estimates when a time period (Baghdadli *et al.,* 2008; Seltzer *et al.,* 2010; Shattuck *et al.,* 2007), or longevity of self-injury (Dominick, Davis, Lainhart, Tager-Flusberg & Folstein, 2007) or greater severity of self-injury (Cooper *et al.,* 2009; Lecavalier, 2006) is specified within the definition. With a view towards clinical utility, this divergence in definitions provides an opportunity to evaluate the severity of self-injury displayed by individuals with ASD. Whilst there is a reportedly high prevalence of self-injury in ASD populations, these results suggest that the prevalence of enduring and severe self-injury in ASD is lower. In order to delineate this further, additional research is required in a single ASD population, where the presence/absence of self-injury is ascertained, alongside a measure of severity and persistence.

In summary, despite significant variation between the prevalence rates, and the differences in sample characteristics and self-injury definitions, the vast majority of studies report higher levels of self-injury in ASD populations than those reported in the general intellectual disability population. However, many of the datasets are limited by small sample sizes or insufficient evaluation of ASD diagnosis and intellectual disability status. There is a need for robust research on the prevalence, severity and persistence of self-injury, in large samples where ASD phenomenology and intellectual or adaptive functioning are also assessed.

1.4.2 Association between self-injury and autism spectrum disorder diagnosis and phenomenology

In addition to those studies reporting the prevalence of self-injury in ASD populations, a number of studies have evaluated the broader association between ASD diagnosis and the presence of self-injury through the use of group comparisons. Cohen *et al.,* (2010) demonstrated that self-injury was associated strongly with autism diagnosis. In a comprehensive study, Bodfish, Symons, Parker and Lewis (2000) evaluated the differences in repetitive behaviour between matched groups of adults with ASD and intellectual disability of heterogeneous aetiology. They found that a higher percentage of individuals with ASD engaged in self-injury, however the sample sizes of the groups were relatively small. Other studies utilising group comparison designs have yielded similar results. When compared to children with a history of language impairment (Dominick *et al.,* 2007), children with Down syndrome and typically developing children (Griffith *et al.,* 2010), children with ASD exhibit significantly higher levels of self-injury. Similarly, in a 'gold standard' meta-analysis, McClintock *et al.,* (2003) demonstrated that individuals with ASD were 6.41 times more likely to show self-injury than individuals with intellectual disability of heterogeneous aetiology. These studies all provide evidence of elevated levels of self-injury in ASD samples when compared to non-ASD populations. Thus, in addition to the elevated prevalence of self-injury in ASD populations, there is also evidence that the presence of self-injury is associated with the presence of ASD diagnosis. This work could be extended to contrast an ASD sample with multiple comparison groups in which levels of self-injury are already known. This would serve to better anchor the elevated prevalence levels of self-injury in ASD against the prevalence of self-injury in known populations.

Building upon work comparing self-injury in ASD and non-ASD populations, there is a substantial body of evidence describing a broader association between self-injury and ASD phenomenology. In two large scale regional surveys with samples of over 2000 individuals, the presence of self-injury was significantly associated with the number of identified autistic traits and symptoms (Bhaumik *et al.,* 1997; Collacott *et al.,* 1998). Collacott *et al.* (1998) demonstrated this association to be independent of effects of age, developmental ability, hearing status and mobility through the use of logistic regression modelling. Interestingly, there was also a significant association between the need for medical attention following self-injury and the number of autistic traits, suggesting that the presence of ASD type behaviours may be related to the severity of self-injury displayed (Bhaumik *et al.,* 1997).

Lowe *et al.* (2007) and Murphy *et al.* (2005) conducted total population surveys and found that self-injury was significantly associated with the triad of impairments. The very large populations from which these samples were obtained (total populations of 1.2 million and almost 35 thousand respectively) strengthen the findings and indicate a robust association between the presence of ASD phenomenology and self-injury. However, given the sample sizes assessed in these studies, it was not possible to conduct individual assessments in which ASD diagnosis was confirmed. Consequently, many of the studies relied upon informant report using measures such as the Disability Assessment Schedule (DAS; e.g., Collacott *et al.,* 1998; Lowe *et al.,* 2007). Thus, the results imply that the presence of ASD type behaviours, rather than a clinical diagnosis of ASD, is associated with self-injury. Whilst this may initially appear to be a limitation of the research, the findings offer an opportunity to evaluate the effect of ASD phenomenology rather than ASD diagnosis per se, upon the presence of self-injury. The results suggest that ASD phenomenology may be a useful putative risk marker for self-injurious behaviour within multiple populations. Supporting this assertion, Arron *et al.* (2011) used the Social Communication Questionnaire to demonstrate that for individuals with Cornelia de Lange, Fragile X, Prader-Willi and Lowe syndromes, specific areas of the triad of impairments were associated with self-injury. Taken together, this research evidence demonstrates that it is the presence of ASD phenomenology, rather than a diagnosis of idiopathic autism per se, that is associated with the presence of self-injury.

Despite the substantial evidence associating ASD with self-injury, there is also a significant body of research that has failed to find an association between ASD phenomenology/ diagnosis and self-injury. Bradley *et al.,* (2004) and Matson *et al.,* (1996) used the DASH II to compared individuals with ASD and intellectual disability. Both studies found no significant differences between the two groups on the self-injury scale. However, these results should be interpreted with caution as the reported prevalence rates for self-injury in the intellectual disability groups were strikingly high (50.0% and 38.0%). This suggests that the measure used to assess self-injury was not valid or reliable. In a group comparison study, Richler, Bishop, Kleinke and Lord (2007) compared prevalence rates of self-injury in ASD in typically developing and intellectual disability samples. Richler *et al.,* (2007) found no significant differences between the three groups. However, the study was specifically designed to investigate repetitive behaviours in very young children (average age of sample ≈ 2 years). The authors discuss that at this early age, there are no significant differences between the groups, but that these differences must emerge during development, as other research demonstrates that self-injury decreases in the typically developing group and increases in the ASD group. Very little is currently known about the early emergence of self-injury, particularly in ASD samples. This study provides a useful basis for further longitudinal research to delineate the development of self-injury in individuals with ASD.

Several longitudinal studies have also failed to find an association between ASD and persistence of self-injury over time. Cooper *et al.* (2009) reported that ASD diagnosis did not predict self-injury at the second time point of a two year longitudinal study. However, ASD diagnosis was strongly associated with self-injury at the first time point. The study included a relatively small sample of individuals with ASD (N=77, of whom 10 engaged in self-injury) compared to the total cohort (N=1023). Therefore it is plausible that the lack of association between ASD and self-injury over time is due to a lack of statistical power. In another longitudinal study, Chadwick, Piroth, Walker, Bernard and Taylor (2000) failed to find an association between self-injury and informant report of autism diagnosis at time point one. However, at follow up, an independent measure of ASD was included and higher total score on this measure was associated with higher levels of total problem behaviour (Chadwick *et al.,* 2008). A final longitudinal study investigated factors predictive of continued self-injury in an intellectual disability service (Danquah *et al.,* 2009) and also found no association with ASD. Taken together, despite the limitations of these studies, the results do appear to provide evidence that ASD may not be associated with self-injury longitudinally. Further research is needed, with substantial sample sizes of participants with ASD, in order to evaluate the association between ASD and self-injury over time.

1.4.3 Associations between autism spectrum disorder severity and self-injurious behaviour severity

Progressing from research demonstrating that ASD phenomenology is associated with self-injury, there is also a small body of research associating ‘severity’ of ASD with ‘severity’ of self-injury. In a large scale interview study, Bhaumik *et al.* (1997) demonstrated that the number of autistic traits were associated with the presence of self-injury, and with the need for medical attention following self-injury. Similarly, Matson and Rivet (2008) found that individuals who were identified as having severe rather than mild ASD symptoms had significantly higher levels of self-injury. Baghdadli *et al.* (2003) report very similar findings, demonstrating that in a sample of children with ASD, more severe self-injury was associated with a higher endorsement of autism symptomatology.

These findings suggest an association between ASD severity and self-injury severity. However, in most studies, the 'severity of ASD' is poorly defined; it is unclear whether this term reflects an increased number of ASD traits or symptoms, or a greater level of impairment in a limited number of ASD traits or symptoms. Likewise, self-injury is poorly operationalised, thus it is not clear whether increased severity reflects an increased number of topographies, increased rate, increased frequency or increased damage resulting from self-injury. Therefore, in order to better understand the relationship between ASD and self-injury severity, future studies must explicitly define ASD severity and self-injury severity.

1.4.4 Persistence of self-injurious behaviour in autism spectrum disorder

Within intellectual disability populations, self-injury has been identified as a relatively persistent behaviour. Emerson *et al.,* (2001b) demonstrated that over a seven year period, 71% of individuals continued to display self-injury. Similarly, Taylor, Oliver and Murphy (2011) demonstrated that self-injurious behaviour was persistent in 84% of people with intellectual disabilities over a 20 year period. However, there are limited longitudinal data evidencing the persistence of self-injury in ASD populations. Esbensen, Seltzer, Lam and Bodfish (2009) evaluated the stability of a variety of repetitive behaviours in a large cross sectional study.

They found that older age was significantly associated with lower levels of self-injury. However, when the change in self-injurious behaviour was plotted for different age groups, the gradient of change in self-injury declined less steeply with age than other forms of repetitive behaviour. This may indicate that self-injury is more persistent than, for example, restricted interests and stereotyped behaviours. Similarly, Matson, Mahan, Hess, Fodstad and Neal (2010) utilised a cross sectional survey design and demonstrated that self-injury was consistent, and therefore persistent, across subsamples of young children, children and young adolescents with ASD. However, the results of both of these studies must be interpreted with caution; despite assessing samples with well validated ASD diagnosis, the studies utilised cross sectional designs to imply persistence. Ideally, longitudinal designs should be employed in order to fully explore the persistence of self-injury in individuals with ASD.

To date, two longitudinal studies evaluating self-injury stability in ASD populations have been conducted. Shattuck *et al.,* (2007) investigated the stability of ASD phenomenology and maladaptive behaviours in a cohort of adolescents and adults with ASD. The study revealed that the prevalence of self-injury decreased significantly over four and half years. However, the study recruited individuals through service agencies and clinics specifically and consequently may have recruited individuals who would be most likely to receive interventions to reduce self-injury. Baghdadli *et al.,* (2008) report similar results in a three year follow up of children with Pervasive Developmental Disorders (PDD). At the first time point, 49% of their sample engaged in self-injury, whereas at the second time point, only 32.7% did. However, there are two caveats to these results. Firstly, the sample of children were relatively young (mean age of children at second time point was 8 years) and the prevalence of self-injury is known to peak between the ages of 15 and 25 (Oliver *et al.,* 1987). Therefore, a longer outcome study, or broader age range of sample may provide more valuable information regarding the persistence of self-injury in this population. Secondly, at the first time point, all of the children in the study reported by Baghdadli *et al.* (2008) were receiving treatment, with 98% of them attending day hospitals that provide services for children with psychiatric disorders. Therefore, as with the study presented by Shattuck *et al.* (2007), it is not possible to evaluate whether the remission in self-injury is the natural course of development of self-injury in ASD or a result of intensive intervention. Consequently, there is a need to establish the persistence of self-injury in a population with ASD that has not been recruited from clinical services and is not engaged in therapeutic intervention.

1.4.5 Summary of self-injurious behaviour in autism spectrum disorder: epidemiology

The research reviewed suggests that a significant proportion of individuals with ASD engage in self-injury. Even the most conservative estimates of prevalence indicate an elevated presence of self-injury in samples with ASD compared to those with intellectual disability of heterogeneous aetiology (e.g., Cooper *et al.,* 2009; Richler *et al.,* 2007; Seltzer *et al.,* 2010). Additionally, there is tentative evidence to suggest an association between ASD severity and self-injury severity (Baghdadli *et al.,* 2003; Matson & Rivet, 2008). However, despite the broad trends detailed in the literature, many of the findings are limited by inadequate assessment and description of both self-injury and ASD sample characteristics. Additionally, there are very limited data evaluating the persistence of self-injury in ASD populations. Therefore, further research is required, utilising robust measures of group characteristics and self-injury, in large longitudinal samples, in order to delineate the size and scale of the problem of self-injury in ASD populations.

1.5 Self-Injurious Behaviour in Autism Spectrum Disorder: Person Characteristics and Operant Function

In addition to a description of the epidemiology of self-injury in ASD populations, there is also a need to investigate both person characteristics and operant function of self-injury, in order to inform an interpretation of the epidemiological data. As highlighted above, certain person characteristics and specific genetic disorders have been identified which influence both the presence and severity of self-injury (see Section 1.3.2.1). Given the purported elevated prevalence of self-injury in individuals with ASD, it is prudent to assess and identify any person characteristics associated with self-injury in this population. The identification of person characteristics associated with self-injury in ASD will allow for the delineation of risk markers for self-injurious behaviour and will afford an opportunity to build a model of self-injury which accounts for the elevated prevalence in individuals with ASD.

Similarly, operant theory has been applied usefully in order to understand the occurrence, maintenance and severity of self-injury (see Section 1.3.3.1). One corollary of operant theory indicates that functional self-injury will systematically fluctuate given the appropriate environmental conditions. A second corollary suggests that functional self-injurious behaviour will appear within a response class in an individual's behavioural repertoire. An investigation and understanding of both of these corollaries will afford an opportunity to evaluate the utility of operant theory in explaining self-injury in individuals with ASD. As an application of operant theory offers explanations for the emergence (Guess & Carr, 1991), development and maintenance of self-injury, an evaluation of operant behaviour in ASD will also contribute to a model of self-injury accounting for the elevated prevalence in ASD.

In summary, an investigation of both person characteristics associated with self-injury, and the utility of operant theory in accounting for the presence of self-injury, will directly influence the interpretation of data regarding the epidemiology of self-injury in ASD.

1.5.1 Association between self-injurious behaviour and person characteristics

Associations between a variety of behavioural and demographic variables and self-injury have been investigated in populations with intellectual disability of heterogeneous aetiology (see Section 1.3.2.1). However, little research has evaluated these associations in populations with ASD. The following sections describe and evaluate research investigating person characteristics and self-injury conducted in populations with ASD, contrasted with research conducted in individuals with intellectual disability of heterogeneous aetiology.

1.5.1.1 Age

Reported associations between age and self-injury in individuals with intellectual disability vary, with the majority of studies reporting that younger age is associated with the presence of self-injury (Emerson *et al.,* 2001a; Collacott *et al.,* 1998). Similarly, within a population with ASD, Esbensen *et al.,* (2009) demonstrated that older age was correlated with significantly lower levels of self-injury. However, in a younger sample of children with ASD, age was not significantly associated with the presence or severity of self-injury (Bagdhadli *et al.,* 2003). The conflicting data may be reflective of a more complex curvilinear relationship between age and self-injury. Oliver *et al.,* (1987) demonstrated this curvilinear relationship in a total population sample of individuals with intellectual disability of heterogeneous aetiology. The results demonstrated that the prevalence of self-injury peaks between the ages of 15 and 25. An operant model successfully accounts for the increasing prevalence of self-injury as behaviour becomes learnt and is shaped by the environment (see 1.3.3.1). However, the resulting decrease in prevalence as age increases is yet to be fully explained. Given the complex association between age and self-injury, it is necessary to evaluate this association in a sample of individuals with ASD which contains a sufficient number of participants across the lifespan. Delineation of the association between age and self-injury will inform theoretical understandings of the development of self-injury in ASD populations.

1.5.1.2 Gender

Whilst gender has been associated with aggressive behaviours (McClintock *et al.,* 2003), the majority of studies have found no significant differences between males and females with regard to self-injury (Collacott *et al.,* 1998; Griffin, Williams, Stark, Altmeyer, & Mason, 1986). A meta-analysis of 22 prevalence and cohort studies of challenging behaviour in individuals with intellectual disability found no evidence to suggest an association between self-injurious behaviour and gender (McClintock *et al.,* 2003). In a sample of individuals with ASD, Baghdadli *et al.* (2003) replicated these results demonstrating no association between self-injury and gender. However, in a recent study by Cohen *et al.* (2010) in which self-injury was significantly associated with ASD, this association was stronger for females. Therefore, the association between self-injury and gender in ASD samples remains unclear. This association warrants further research, and is of particular importance for individuals with ASD, given the heightened prevalence of ASD diagnoses in males (Saracino *et al.,* 2010).

1.5.5.3 Adaptive functioning

Low levels of adaptive functioning have been associated with self-injury in individuals with intellectual disability. A greater severity of intellectual disability is strongly associated with the presence of self-injury (Collacott *et al.,* 1998; McClintock *et al*., 2003; Schroeder, Schroeder, Smith & Dalldorf, 1978). Poor mobility has also been identified as a risk marker for self-injury (Murphy, Hall, Oliver & Kissi-Debra, 1999). In addition, impaired expressive language (Collacott *et al.,* 1998; Emerson *et al.,* 2001a; Murphy *et al.,* 2005; Schneider, Bijam-Schulte, Janssen & Stolk, 1996) and social skills functioning (Murphy *et al.,* 2005) have been identified as correlates of self-injury in individuals with intellectual disability.

The relationship between impaired social-communicative behaviours and self-injurious behaviour should be considered with respect to operant models which propose challenging behaviours to be analogous to communication. Given that impaired communication is a necessary characteristic for a diagnosis of autism, it is expected that this deficit will lead to increased susceptibility to develop self-injury in individuals with ASD. Unsurprisingly therefore, deficits in expressive language (Dominick *et al.,* 2007) along with low levels of adaptive functioning (Baghdadli *et al.,* 2003) and IQ (Dominick *et al.,* 2007; McTiernan *et al.,* 2011) are all associated with self-injury in individuals with ASD. Importantly, lower speech levels were found to predict the persistence of self-injurious behaviour in children with ASD over time (Baghdadli *et al.,* 2008), indicating that lower speech levels may serve as a putative risk marker for self-injury in individuals with ASD. This hypothesis requires further investigation in both adult and child samples with ASD, given that adaptive functioning, expressive language and ASD symptoms are often confounded (McClintock *et al.,* 2003).

1.5.5.4 <u>Aggressive behaviours</u>

Almost half of all individuals with intellectual disability of heterogeneous aetiology who display self-injury also engage in aggressive behaviours (Emerson *et al.,* 2001a; Griffin *et al.,* 1986). Studies have consistently found an association between self-injury and aggression (Schneider *et al.,* 1996; Collacott *et al.,* 1998), however, aggression does not appear to be associated with long term and persistent self-injury (Emerson *et al.,* 2001b; Griffin *et al.,* 1986). Additionally, temporal relationships have been identified between self-injurious and aggressive behaviours in individuals with intellectual disability of heterogeneous aetiology (Petty *et al.,* 2009), indicating that mutual reinforcement contingencies may operate to maintain these different behaviours. Whilst the focus of this thesis is on self-injurious behaviour in ASD, investigation of the temporal associations between aggressive and self-injurious behaviours in this population is warranted, given the significant clinical implications of both behaviours.

1.5.5.5 <u>Sensory sensitivity</u>

Unusual sensory responses have frequently been documented within both preschool (Ornitz, Guthrie & Farley, 1977) and school aged children (Kientz & Dunn, 1997) with ASD when compared to typically developing controls. Using parental report questionnaires, children with ASD have been found to have significantly more symptoms of sensory sensitivity compared to comparison groups of children with intellectual disabilities and typically developing children (Rogers, Hepburn & Wehner, 2003). This profile of heightened sensory sensitivity is comparable to the levels of sensitivity seen in children with Fragile X syndrome and sensory modulation disorder (Miller, Reisman, McIntosh, & Simon, 2001). Theories regarding the development of self-injury have postulated that self-injurious behaviour may function to modulate levels of sensory arousal (Hutt & Hutt, 1965). However, despite the high levels of sensory sensitivity reported in individuals with ASD, there has been little consideration of the relationship between function of self-injury and sensory sensitivity in this population.

1.5.5.6 Repetitive behaviour

Self-injury is often viewed as a maladaptive repetitive or stereotyped behaviour. Consequently, research has evaluated associations between these behaviours in an attempt to delineate the aetiology of self-injury. However, whilst there have been models proposed to delineate the development of self-injury from stereotypies (e.g., Guess & Carr, 1991), the database of evidence directly assessing these behaviours is lacking (Symons, Sperry, Dropik & Bodfish, 2005). The available research appears to link stereotyped and self-injurious behaviours, showing that individuals with intellectual disability who engage in stereotypy are twice as likely to display self-injury (Collacott *et al.,* 1998). Additionally, stereotyped behaviours are associated with self-injurious behaviour in individuals with intellectual disability (Emerson *et al.,* 2001a).

Importantly the presence of repetitive and restricted behaviours are a diagnostic feature of ASD (DSM-IV; American Psychiatric Association, 1994; ICD-10; World Health Organization, 1992), and individuals with ASD are reported to display higher levels of repetitive behaviour than individuals with intellectual disability of heterogeneous aetiology (Estes *et al.,* 2011; Turner, 1999) or typically developing individuals (Richler *et al.,* 2007). Associations have been identified between broad categories of repetitive behaviour and self-injury in individuals with ASD (Dominick *et al.,* 2007). However, there has been little investigation of the associations between specific forms of repetitive behaviour and self-injury in ASD. Additionally, the predictive value of repetitive behaviours as a risk marker for self-injury in individuals with ASD has not been delineated.

In addition to the utility of repetitive behaviour as a risk marker for self-injury, repetitive behaviour has also been hypothesised as a stage in the development of self-injury (Guess & Carr, 1991). Longitudinal study has associated stereotyped, proto-injurious and self-injurious behaviour (Richman & Lindaur, 2005), supporting a theory of progression from stereotypies to self-injury. This relationship has been further supported in populations with intellectual disability, where temporal relationships have been identified between repetitive behaviours and self-injury (Petty *et al.,* 2009). Given the heightened prevalence of stereotyped and repetitive behaviours in individuals with ASD, it is essential that this relationship is further understood, in order to delineate the development of self-injury in individuals with ASD.

1.5.5.7 <u>Overactive and impulsive behaviour</u>

Despite the tentative causal models linking repetitive and self-injurious behaviours, evidence suggests that repetitive behaviours are displayed by typically developing children but rarely develop into self-injury. This may indicate that other factors are contributing to this relationship in individuals with intellectual disability and/or ASD. One hypothesis has been that an underlying executive dysfunction, evidenced by impulsivity, overactivity and compulsive behaviours, mediates the relationship between repetitive/stereotypic behaviours and self-injury (Bodfish *et al.,* 1995).

This model is supported by evidence that self-injury is associated with overactivity in adults with intellectual disability (Collacott *et al.,* 1998) and in individuals with Cornelia de Lange syndrome (Oliver, Sloneem, Hall & Arron, 2009). Hyperactivity has been found to be more common in individuals who display self-injury than a matched comparison group who do not display self-injury (Schneider *et al.,* 1996). In addition to this, many individuals who engage in self-injurious behaviour also actively seek out and engage in self-restraint behaviours (Forman, Hall & Oliver, 2002; Fovel, Lash, Barron, & Roberts, 1989; Oliver, Murphy, Hall, Arron & Leggett, 2003). This may suggest that self-injury is not fully under the individual's control, perhaps due to a compromised ability to inhibit the behaviour. Compromised inhibition may influence self-injury in one or more ways (Barkley, 1997). It is plausible that impaired inhibition drives the individual to engage in self-injury as a prepotent response to triggering stimuli. Additionally, compromised inhibition may result in individuals being unable to terminate self-injury and thus self-restraint behaviours may reflect an attempt to suppress self-injury.

The relationship between self-injury and self-restraint was investigated in a group of individuals with CdLS (Hyman, Oliver & Hall, 2002). Hyman *et al.*, (2002) found a significant association between self-injury and self-restraint. Importantly they also found that those individuals with CdLS who engaged in both self-injury and self-restraint displayed significantly more 'compulsive'[9] behaviours than those who did not display self-injurious or self-restraint behaviour. It is plausible that such an association exists within individuals with ASD. This hypothesis is supported by results which show that a group of individuals with ASD display significantly higher numbers of stereotypies and compulsions than a comparison group of individuals with intellectual disability. The group with ASD also had significantly greater severity ratings for 'compulsions', stereotypy and self-injurious behaviour (Bodfish *et al.,* 2000).

---

[9] 'Compulsive' behaviour was used as a term in this study to describe impulsive repetitive behaviours, as opposed to the anxiety relieving compulsive behaviours typically seen in conditions such as Obsessive Compulsive Disorder.

These preliminary findings may indicate a model of self-injury in ASD in which poor behavioural inhibition, as evidenced by overactivity/impulsivity and self-restraint behaviours, drives the development and maintenance of self-injury. However, thus far, no empirical study has quantified the associations between these behaviours and self-injury in individuals with ASD. This area warrants significant further attention, as the findings may lead to a more complete model of self-injury that could explain prevalence, maintenance and severity differences which are difficult to account for using purely operant and biological models.

1.5.5.8 Painful health conditions

Given the putative causal association between pain and self-injury (see Section 1.3.3.3), it is important to establish whether such an association may exist in individuals with ASD. Several studies have included individuals with ASD and provide preliminary evidence that pain and painful health conditions may have a causal role (Carr & Owen-DeSchryver, 2007; Christensen *et al.,* 2009). However, despite the emergent evidence of an association between painful health conditions and self-injurious behaviour, to date no empirical research has evaluated this association in large groups of individuals with ASD. Given the purported high levels of self-injury in individuals with ASD, it is critical that painful health conditions are quantified and evaluated in this population.

1.5.2 Operant function of self-injurious behaviour in autism spectrum disorder

As detailed in Section 1.3.3.1, operant theories have influenced assessment and intervention techniques for self-injury. Due to the success of operant based interventions, significant research attention has been given to developing assessments which are effective at identifying the antecedents and consequences, or ‘function’ of self-injury in individuals with intellectual disability (Hanley *et al.,* 2003). Experimental functional analytic techniques based on those pioneered by Iwata *et al.* (1994) and Carr and Durand, (1985) have been shown to identify functions for self-injurious behaviour as evidenced through effective interventions (see Hanley *et al.,* 2003 for a comprehensive review). However, several problems with using experimental functional analyses have been identified. Experimental functional analyses may be considered complex and difficult to implement in clinical settings, lacking in ecological validity and unable to test for idiosyncratic antecedents and consequences. There have also been concerns regarding the ethics of reinforcing challenging behaviours during functional analysis and potentially creating a new behavioural function (Hall, 2005). Therefore, descriptive and informant based methods of eliciting behavioural function in individuals with intellectual disability have been developed.

Informant report measures such as the Questions about Behavioral Function (QABF, Paclawskyj, Matson, Rush, Smalls & Vollmer, 2000) have good validity and reliability in assessing behavioural function. Informant report measures are quick and simple to administer in clinical settings, and often produce similar results to experimental functional analysis assessments (Hall, 2005; Tarbox *et al.,* 2009). However, informant report measures rely on correlational evidence and therefore conclusions regarding causality cannot be drawn. Additionally, as they assess a selection of common antecedents and consequences delineated prior to the initiation of the research, they are also unable to responsively identify idiosyncratic functions to behaviour, such as accessing materials involved in repetitive behaviour or escaping sensory stimulation. Given the limitations of informant report measures, they are most successfully used as an adjunct or precursor to experimental methods.

Descriptive methods are those in which behaviours are observed in naturalistic conditions and no direct manipulations of antecedents or consequences occur. Through the comparison of conditional and unconditional probabilities, naturally occurring antecedents and consequences are determined, and operant function is assumed based on these. Descriptive assessments provide an ecologically valid method; however concordance with experimental functional analyses in individuals with intellectual disability is very poor. From this poor concordance, it is often assumed that experimental functional analyses have the most robust methodology, leading the majority of researchers to identify experimental assessments as the preferred method for assessing operant function (Hall, 2005; Pence, Roscoe, Bourret & Ahearn, 2009; Tarbox *et al.,* 2009). Additionally, collecting data for descriptive assessments is very time consuming and analysis methods are more complex than those used for experimental functional analysis. Given the limitations of both descriptive and informant based methods, research in individuals with intellectual disabilities has focused predominantly upon limiting the flaws in experimental functional analyses by improving ecological validity, developing methods using brief analogue sessions, identifying idiosyncratic functions and limiting potential ethical problems (Hanley *et al.,* 2003).

Given the reportedly high prevalence of self-injury in individuals with ASD (Section 1.4.1) and the overwhelming body of research supporting operant based assessments and interventions for self-injurious behaviour (Section 1.3.3.1), it might be assumed that operant functions for self-injury in individuals with ASD have been studied extensively. However, the number and range of robust experimental studies of operant function for self-injury in ASD is limited. Studies utilising standardised experimental functional analyses have been conducted, however the majority report results from single case studies and do not verify the diagnosis of

ASD (Devlin, Healy, Leader & Reed, 2008; Hagopian, Bruzek, Bowman & Jennett, 2007; Hagopian, Wilson & Wilder, 2001; Hausman, Kahng, Farrell & Mongeon 2009; Healey, Ahearn, Graff & Libby, 2001; McComas, Hooch, Paone & El-Roy 2000; McKerchar, Kahng, Casioppo & Wilson, 2001; Tiger, Fisher, Toussaint & Kodak, 2009). Where robust cohort studies have been conducted, functions for challenging behaviour, rather than self-injurious behaviour specifically have been identified (O' Reillly *et al.,* 2010). Therefore, there is a need for robust cohort studies investigating the operant function of self-injury in samples of individuals where ASD diagnosis and phenomenology have been independently assessed.

Recently, idiosyncratic functions for self-injurious behaviour in cohorts with ASD have been identified using informant report measures (Reese, Richman, Belmont & Morse, 2005; Reese, Richman, Zarcone & Zarcone, 2003). Single case studies have also begun to experimentally identify idiosyncratic functions for self-injury in ASD (e.g., Hagopian *et al.,* 2001; Hausman *et al.,* 2009; Tiger *et al.,* 2009). These idiosyncratic functions have included access to materials involved in repetitive behaviours, escape from sensory stimulation and access to completion of routines. Importantly, all of these idiosyncratic functions can be seen as an interaction between the environment and ASD characteristics, such as a preference for routine or low tolerance for sensory stimulation. In this way, the phenotype of ASD may interact with the environment and result in operant functions for behaviour which are less common in other populations. This phenotype x environment interaction may provide a useful framework for understanding further the elevated prevalence of self-injury in ASD. However, thus far no cohort studies utilising experimental functional analyses and appropriate assessments of ASD diagnosis to investigate idiosyncratic functions have been conducted. Therefore, there is a need for robust experimental assessments of idiosyncratic functions for self-injury in individuals with ASD.

Finally, whilst the function of self-injury has predominantly been explored through an analysis of environment x behaviour interactions, operant theory also affords an opportunity to evaluate functional relationships through behaviour x behaviour interactions. An important corollary of operant theory indicates that through environmental interactions, behaviour becomes functionally and hierarchically organised within an individual's repertoire (Section 1.3.3.1). Thus, an investigation of the temporal associations between self-injury and other behaviours shown by an individual, allows for an analysis of the organisation of behaviour and consequently an evaluation of the utility of operant theory to explain that behaviour. In an application of this reasoning, Petty *et al.,* (2009) conducted a cohort study of response classes and temporal behavioural organisation in individuals with intellectual disability of heterogeneous aetiology. Petty *et al*., (2009) demonstrated that repetitive and proto-communicative behaviour appeared to be functionally organised around self-injury with both theoretical and pragmatic implications. These temporal associations allowed for an examination of the utility of Guess and Carr's (1991) model of the development of self-injury *and* suggested useful implications for the development of functional communication interventions.

Despite the proven utility of the method and analysis conducted by Petty *et al.,* (2009), no cohort study of temporal behavioural associations with self-injury has been conducted in individuals with ASD. Although several single case studies have demonstrated temporal associations between repetitive behaviour and self-injury (Herscovitch, Roscoe, Libby, Bourret & Ahearn, 2009; Langdon, *et al.,* 2008) or proto-imperative communications and self-injury (Herscovitch *et al.,* 2009; Smith & Churchill, 2002) these data are limited as no independent assessment of ASD phenomenology was conducted. This prevents the results from being generalised to wider populations with ASD (Section 1.4.1). Therefore, there is a need to explore operant theory through the temporal associations between self-injury and other behaviours in a cohort of individuals with ASD, where ASD phenomenology has been independently assessed.

1.5.3 Summary of associated person characteristics and operant function of self-injury in ASD

The research reviewed indicates a number of person characteristics for which robust associations with self-injury have been identified in populations with intellectual disability of heterogeneous aetiology. However, despite the reportedly high levels of self-injury in individuals with ASD, there has been limited research investigating these variables in ASD populations. Similarly, the substantial evidence supporting operant models of assessment and intervention for self-injury has not resulted in any comprehensive research detailing the antecedents and consequences for self-injury in cohorts of individuals with ASD. Nor has there been any systematic investigation of the organisation of behavioural repertoires around self-injury in individuals with ASD. An improved understanding of risk markers for, and operant function of, self-injury in ASD may lead to a more complete model of self-injurious behaviour in this population. Specifically, an understanding of ASD phenotype x environment interactions may help to account for the elevated prevalence of self-injury in individuals with ASD. In addition to improved theoretical understanding, pragmatic outcomes such as more targeted and efficacious interventions may also follow. Therefore, it is critical that further research is conducted in order to delineate the associated person characteristics and operant function of self-injury in individuals with ASD.

1.6 Conclusion and aims

The research delineated above has described the pervasive and often detrimental consequences of self-injury in populations with ASD. This thesis has a number of specific aims motivated by the reviewed research and the limitations of the research described in this chapter.

- The prevalence of self-injury is purported to be elevated in individuals with ASD (Section 1.4.1). However, the existing prevalence data are limited by small sample sizes and inconsistent or less robust measures of self-injury, ASD phenomenology and intellectual disability (Section 1.4.1). Thus, Chapter 2 presents a robust epidemiological study detailing the prevalence of self-injurious behaviour in individuals with ASD. These data are contrasted with prevalence data from multiple comparison groups who evidence reliably documented levels of self-injury.

- Self-injury is evidenced to be a persistent behaviour in individuals with intellectual disability of heterogeneous aetiology (Section 1.4.4). However, evaluation of the persistence of self-injury in individuals with ASD has been limited by reliance on data collected in samples receiving ongoing clinical intervention (Section 1.4.4). Therefore, Chapter 3 presents a three year longitudinal follow up of the data presented in Chapter 2, allowing for an evaluation of the persistence of self-injury in ASD in a non-clinical sample.

- The prevalence of self-injury in populations with intellectual disability is known to be influenced by the presence of a number of key person characteristics (Section 1.3.2.1 & 1.5.1). However, despite the purported elevated prevalence of self-injury in ASD, data evaluating the association between person characteristics and self-injury in individuals with ASD are minimal or non-existent. Therefore, delineation of person characteristics associated with self-injury in ASD is provided in both Chapters 2 and 3. Building upon these preliminary findings, Chapter 4 provides an extensive evaluation of person characteristics associated with self-injury in ASD, with a specific focus on the interaction between self-injurious behaviour and self-restraint.

- Operant theory has been employed usefully to explain self-injury in individuals with intellectual disability of heterogeneous aetiology (Section 1.3.3.1). One corollary of the operant account is that self-injury can be observed to systematically fluctuate given certain environmental conditions. However, despite the wide application of techniques to evaluate these systematic fluctuations, there has been limited employment of these methods to study self-injury in cohorts of individuals with ASD (Section 1.5.2). Additionally, there are limited experimental data investigating idiosyncratic functions of self-injury in the context of phenotype x environment interactions in individuals with ASD. Therefore, Chapter 5 details an experimental study evaluating both typical and idiosyncratic functions of self-injury in a cohort of children with ASD.

- A second corollary of operant theory is that self-injury can be observed to be functionally organised in an individual's behavioural repertoire, resulting in response classes and temporal configurations of behaviour (Section 1.3.3.1). However, despite the important theoretical and pragmatic consequences of this behavioural organisation (Section 1.5.2), temporal associations between self-injury and other behaviours have not been examined in individuals with ASD. Thus, Chapter 6 presents an exploratory evaluation of the temporal associations between self-injury, repetitive behaviour, challenging behaviour, proto-imperative communication and self-restraint in a cohort of children with ASD.

- Finally, the development and maintenance of self-injurious behaviour has been modelled by several researchers, most notably Guess and Carr (1991). However, this model can not fully account for the purported elevated prevalence of self-injury in ASD, nor the interactions between person characteristics and the prevalence and function of self-injury (Section 1.3.3.1). Therefore, Chapter 7 synthesises the data generated in this thesis and extends the model proposed by Guess and Carr (1991) to allow for a more complete account of the development and maintenance of self-injurious behaviour in individuals with ASD.

CHAPTER 2

Self-Injurious Behaviour in Autism Spectrum Disorder

2.1 Preface

In the previous chapter, research investigating the prevalence and phenomenology of self-injury was reviewed. A number of areas for future research in individuals with ASD were identified. This included a pressing need for epidemiological estimates of self-injury in ASD, where data were generated from large sample sizes and robust measures of self-injury, ASD phenomenology and intellectual disability or adaptive functioning were employed. Therefore, this chapter aims to assess the prevalence and associated behavioural and demographic characteristics of self-injury in a group of individuals with ASD. Through the employment of standardised survey methodology and between group comparisons, reliable and valid data will be generated.

2.2 Introduction

Prevalence estimates for self-injury are significantly higher for individuals with ASD than for individuals with intellectual disability of heterogeneous aetiology (see Sections 1.3.2 & 1.4.1). Variation in prevalence estimates in ASD samples is likely due to differing participant inclusion criteria, specifically the definition and diagnosis of ASD, and varying definitions of self-injury. Additional variation may also be accounted for by a divergence in degree of ability in the ASD groups. Bartak and Rutter (1976) reported a prevalence rate of 71% for self-injury in children with ASD and IQ below 70, compared to a prevalence rate of 32% in children with ASD and IQ above 70. Despite this variation in the reported rates of self-injury in ASD, all studies demonstrate a higher prevalence of self-injury in individuals with ASD compared to those in the wider intellectual disability population. In order to establish a valid and reliable prevalence estimate of self-injury in ASD it is important that inclusion criteria for ASD groups are rigorous, with careful delineation of ability levels and diagnostic status.

Studies have identified ASD as a risk marker for self-injury in individuals with intellectual disability (Emerson *et al*., 2001a; Baghdadli, Pascal, Grisi & Aussilloux, 2003; McClintock, Hall & Oliver, 2003). The presence of ASD is associated with an increased persistence and severity of self-injury (Bodfish, Symons, Parker & Lewis, 2000). Within populations with ASD, self-injury is associated with increased severity and quantity of autistic behaviours and a 'severe' rather than 'mild' presentation of characteristics (Bhaumik, Branford, McGrother & Thorp, 1997; Matson & Rivet, 2008). Matson and Rivet (2008) assessed the association between the triad of impairments (impaired social interaction, impaired communication and restricted and repetitive behaviour) and the presence of self-injury. Higher total scores and scores on the restricted and repetitive behaviour subscale of their autism diagnostic tool

(Autism Spectrum Disorders – Diagnosis for Intellectually Disabled Adults; Matson & Rivet, 2008) were found to be associated with self-injury. However, these results must be interpreted with caution due to the small sample size, low interrater reliability (.30) and test-retest reliability (.39) of their diagnostic tool. McClintock *et al.* (2003) conducted a meta-analysis of prevalence and cohort studies of challenging behaviour in order to establish the increased risk of self-injury in ASD. They compared rates of self-injury in those with ASD and those without from two studies that provided the necessary prevalence data (Ando & Yoshimura, 1979a; Bhaumik *et al.,* 1997). They found that across the studies, those with ASD were significantly more likely to engage in self-injury than those without ASD. In summary, the majority of studies have shown that both the presence and severity of ASD are associated with self-injury. However, little is still known about how self-injury associates with the behaviours that constitute the triad of impairments both in those with ASD, and in other groups.

In order to better understand the phenomenology and prevalence of self-injury in ASD, it is important that degree of intellectual disability or adaptive functioning is assessed, as both ASD and a greater degree of intellectual disability are associated with a higher prevalence of self-injury (McClintock *et al.,* 2003). Previous studies have utilised a 'test group, control group' model in which individuals with ASD (the 'test group') are compared to individuals with intellectual disability of heterogeneous aetiology (the 'control group'; Ando & Yoshimura, 1979a; Bhaumik *et al.,* 1997). However, this may be problematic due to potentially high levels of undiagnosed/unreported ASD and genetic syndromes within the control group which could increase the identified prevalence of self-injury. In order to delineate self-injury in ASD, any comparison groups need to be as homogenous as possible, both in terms of diagnosis and behavioural phenotype. Moving away from a 'test group, control group' model allows for a more refined understanding of self-injury in ASD. Using standardised measures across multiple homogeneous contrast groups would allow the prevalence and phenomenology of self-injury in ASD to be detailed and contrasted to that of other groups with known characteristics and greater homogeneity.

One possible design is to compare individuals with ASD to individuals with syndromes that have well documented aspects of behavioural phenotypes relevant to the contrasts to be made. Down and Fragile X syndromes each have comparatively well documented behavioural phenotypes. Individuals with Down syndrome evidence a low prevalence of ASD (Cohen *et al.,* 2005; Moss & Howlin, 2009) and a low prevalence of self-injurious behaviour which is similar to that documented in individuals with intellectual disability of heterogeneous aetiology (Blacher & McIntyre, 2006; Chadwick, Piroth, Walker, Bernard & Taylor, 2000). In contrast, those with Fragile X syndrome show high levels of ASD characteristics (Hall *et al.,* 2008; Lewis *et al.,* 2006; Moss & Howlin, 2009) and a high prevalence of self-injury similar to that documented in individuals with ASD (51% - 58%; Arron, Oliver, Moss, Berg & Burbidge, 2011; Hall, Lightbody & Reiss, 2008). Additionally, a specific topography of self-injury, self-biting, is reported to be unusually common in Fragile X syndrome (Arron *et al.,* 2011; Hall *et al.,* 2008, Symons *et al.,* 2010). The selection of these groups enables the association between ASD characteristics and self-injury to be approached in a number of ways. Firstly, the groups have predictable characteristics of relevance. Secondly, it enables a comparison of those with high levels of ASD (ASD and Fragile X syndrome groups) to those with low levels of ASD (Down syndrome group). Finally, comparisons of topography of self-injury can be made across the groups.

In addition to considering the contrasts between groups, a within group analysis of the ASD sample will allow further investigation of factors that may be associated with the increased prevalence of self-injury in ASD. As detailed in Chapter 1, there may be a role for environmental and biological factors in self-injury displayed by individuals with ASD. Several experimental studies have demonstrated that challenging behaviour in individuals with ASD is associated with environmental reinforcement (Love, Carr & LeBlanc, 2008; Reese, Richman, Belmont, & Morse, 2005). Single case studies have also begun to demonstrate the presence of ASD specific functions to challenging behaviour (Hausman, Kahng, Farrell & Mongeon, 2009; Murphy, McDonald, Hall & Oliver, 2000; Tiger, Fisher, Toussaint & Kodak, 2009). Further investigation of the function of self-injury in individuals with ASD is explored in Chapter 5. Although there is strong evidence for an operant explanation of self-injury, a purely environmental model can not explain the increased prevalence of self-injury reported in ASD populations. Consequently non-environmental factors must also be considered.

A number of person characteristics, such as the presence of repetitive behaviour and impulsive/overactive behaviour, are known to be associated with an elevated prevalence of self-injury (see Sections 1.3.2.1 & 1.5.1). Stereotypic and repetitive behaviours are common in ASD populations and contribute to the diagnosis of ASD using DSM-IV criteria (American Psychiatric Association, 1994). These stereotypic and repetitive behaviours may be associated with the high prevalence of self-injury in ASD. It has been proposed that self-injury develops from stereotypic and repetitive behaviour through a process of social and non-social reinforcement (Guess & Carr, 1991). Empirical data provide support for a progression from repetitive behaviours to potentially injurious and self-injurious behaviour (Petty, Allen &

Oliver, 2009; Richman & Lindauer, 2005) suggesting that these behaviours may have common cognitive underpinnings. It has been hypothesised that individuals with ASD have deficits in executive function and response inhibition and that these deficits impact on the individuals' ability to generate and control repetitive behaviours (Turner, 1999). Therefore this underlying executive deficit may contribute to both the repetitive and stereotypic behaviours frequently seen in individuals with ASD and to self-injurious behaviour (Sayers, Oliver, Ruddick & Wallis, 2011). Initial evidence to support this hypothesis demonstrated that a group of individuals with ASD displayed significantly higher numbers of stereotypies and compulsions, behaviours that may be indicative of impaired inhibition, than a comparison group of individuals with intellectual disability (Bodfish *et al.,* 2000). The group with ASD also had significantly greater severity ratings for compulsions, stereotypy and self-injury (Bodfish *et al.,* 2000). However, although this provides preliminary support for the hypothesised link between repetitive behaviour and the presence and severity of self-injury, no research has considered these behaviours within the context of an executive deficit.

The associations between behaviours indicative of poor behavioural inhibition and self-injury have been explored in more detail in those with intellectual disability. Attention deficit hyperactivity disorder (ADHD), overactivity and hyperactivity are independently associated with self-injury (Cooper *et al.,* 2009; Collacott, Cooper, Branford & McGrother, 1998; Oliver, Sloneem, Hall & Arron, 2009; Schneider, Bijam-Schulte, Janssen, & Stolk, 1996). Importantly, ADHD is thought to be underpinned by a delayed development of inhibition, which, amongst other deficits, comprises both an inability to stop the initial prepotent response to a stimulus, and the inability to stop an ongoing response (Barkley, 1997). Were this delay or deficit to be present in individuals with self-injury, it could result in an individual being unable to inhibit their self-injury in response to a learned environmental antecedent *and* being unable to cease self-injury once they had started. Therefore, a delay or deficit in behavioural inhibition could be seen to influence both the presence and severity of self-injury. Preliminary evidence to support this hypothesis is found in individuals who engage in self-injury and also actively seek out and engage in self-restraint behaviours (Forman, Hall & Oliver, 2002; Fovel, Lash, Barron, & Roberts, 1989, Oliver, Murphy, Hall, Arron & Legget, 2003). The presence of self-restraint suggests that the self-injurious behaviour may not be fully under the individuals' control, and may in fact, reflect a compromised inhibition system in which the drive to engage in self-injury cannot be repressed.

This association between self-injurious behaviour and self-restraint was investigated in a group of individuals with Cornelia de Lange syndrome (CdLS; Hyman, Oliver and Hall, 2002). CdLS is a rare genetic syndrome associated with autism spectrum behaviours and severe to profound levels of intellectual disability (Jackson, Kline, Barr & Koch, 1993; Moss *et al.,* 2008; Oliver, Arron, Hall & Sloneem, 2008). Hyman *et al.,* (2002) found a significant association between self-injury and self-restraint in individuals with CdLS. Importantly they also found that those individuals with CdLS who engaged in both self-injury and self-restraint displayed significantly more compulsive behaviours than those who did not display self-injury or self-restraint. Thus, there is preliminary evidence for an association between behaviours indicative of poor behavioural inhibition and self-injurious behaviour. It is plausible that such an association exists within individuals with ASD and may be evidenced through the presence of ADHD type behaviours such as overactivity and impulsivity.

A final characteristic that has been associated with self-injury in ASD is low mood. Hill and Furniss (2006) found that compared to those without autism, individuals with autism had higher scores on a low mood scale, and this difference was significant for those with severe autism. Challenging behaviour has been associated with low mood in individuals with severe and profound intellectual disabilities (Hayes, McGuire, O'Neill, Oliver & Morisson, 2011). However, Ross and Oliver (2002) failed to find a relationship between challenging behaviour and low mood in individuals with intellectual disability, and suggest that low mood may not be a contributor to challenging behaviour, but a correlate of it. Thus, it is unclear whether the association between low mood and self-injury arises because self-injury is a 'behavioural depressive equivalent' (Marston *et al.,* 1997) or, alternatively, whether low mood is related to pain that has repeatedly been demonstrated to be underlying self-injurious behaviour in some people (Breau *et al.,* 2003; Carr & Owen-Deschryver, 2007; Luzzani *et al.,* 2003). Results from Berg, Arron, Burbidge, Moss and Oliver (2007) suggest that the correlation between low mood and self-injury may be accounted for by health problems, which are undetected or unresolved. In this case, both self-injury and low mood are a result of pain experiences and, consequently, co-occur. This may be particularly important for individuals with ASD, who experience sensory dysfunctions (Rogers, Hepburn & Wehner, 2003) that may also cause discomfort. The relationship between pain and self-injury is currently poorly understood (see Section 1.3.3.3), but it is plausible that this is a contributory factor to self-injury in ASD.

In conclusion, using between and within group analyses, this study aims to delineate the prevalence, phenomenology and associated behavioural characteristics of self-injury in ASD. Through analysing data from ASD, Fragile X syndrome and Down syndrome groups, four key areas will be investigated:

i) The prevalence of self-injury in ASD will be delineated in comparison to the other groups. Based on previous literature, it is predicted that there will be a higher prevalence of self-injury in the ASD group, compared to the Down syndrome group (Blacher & McIntyre, 2006; Chadwick *et al.,* 2000), and that there will be a comparable prevalence of self-injury in the ASD group compared to the Fragile X syndrome group (Arron *et al.,* 2011; Symons *et al.,* 2003). Additionally, data will be analysed to describe the topography and severity of self-injury in all groups. In line with previous research, it is hypothesised that individuals with Fragile X syndrome will show homogeneity of self-injury with an elevated prevalence of self-biting (Hall *et al.,* 2008).

ii) The association between ASD behaviours typified as the triad of impairments and self-injury within each of the three groups will be investigated. Consistent with previous research, it is predicted that self-injury will be related to higher levels of autistic behaviour within all groups (Bhaumik *et al.,* 1997; Matson and Rivet, 2008).

iii) Factors which may be associated with self-injury in ASD will be investigated within the ASD group, including repetitive behaviours, activity behaviours, affect and demographic characteristics. Consistent with previous research, it is predicted that the presence and severity of self-injurious behaviour will be associated with lower ability and speech (Ando & Yoshimura, 1979a; Bartak & Rutter, 1976), higher levels of repetitive behaviour and activity (Bodfish *et al.,* 2000; Cooper *et* *al.,* 2009; Collacott *et al.,* 1998; Schneider *et al.,* 1996) and lower levels of affect (Berg, Arron, Burbidge, Moss & Oliver, 2007).

iv) The prevalence of self-injury in a ‘more able’ subsample of individuals with ASD will be investigated. Additionally, factors which may contribute to the prevalence of self-injury in ASD will be examined within this subsample to explore how far these factors are associated with self-injury when ability level is controlled.

2.3 Methods

2.3.1 Recruitment

Participants with ASD, Fragile X syndrome and Down syndrome were recruited via the National Autistic Society, Fragile X Society and the Down's Syndrome Association respectively. 288 carers of individuals with ASD (return rate 19.63%), 144 carers of individuals with Down Syndrome (return rate 28.80%) and 212 carers of individuals with Fragile X syndrome (return rate 44%) completed the questionnaire pack.

2.3.2 Procedure

All carers received an information sheet, cover letter, consent form, demographic questionnaire and questionnaire pack (see Appendix A). To avoid priming, the study was described as investigating behaviours associated with the relevant syndrome group. Carers returned completed questionnaires and consent forms in a prepaid envelope.

Ethical approval for this study was obtained from the School of Psychology ethical review committee at the University of Birmingham.

2.3.3 Participants

Participants under the age of four were excluded from the study as some measures were not appropriate for young children. Additionally, participants were excluded from the analysis if they did not have a confirmed diagnosis of the respective syndrome from a professional. For individuals with Fragile X and Down syndromes, the diagnosis was deemed to be confirmed if received from a General Practitioner, Clinical Geneticist or Paediatrician. For individuals with ASD, the professionals additionally included Psychiatrists, Clinical Psychologists and

Educational Psychologists. For individuals with ASD, the diagnoses included autism, Asperger syndrome, autism spectrum disorder and pervasive developmental disorder. If a large proportion of the data (25% or more of items across questionnaires) were incomplete, the participant was excluded from the analysis. Additionally, as data from the Social Communication Questionnaire (SCQ; Berument, Rutter, Lord, Pickles & Bailey 1999) were critical to this study, if participants had incomplete total scores on the SCQ, they were removed from the analysis.

To ensure an accurate comparison of the prevalence and topography of self-injury, SCQ scores were used to screen the ASD group to ensure that all individuals in the ASD group displayed sufficient behaviours indicative of ASD. Therefore all individuals scoring below the ASD cut off on the SCQ were removed from the analysis.

2.3.3.1 Primary group analysis

For the primary between groups analysis, participants who scored the maximum possible score of nine on the self help subscale of the Wessex (Kushlick, Blunden & Cox, 1973) were excluded[1]. The Wessex was used as a proxy estimate of level of ability. This was done in order to increase control over the ability levels of the groups for between group analyses. Ability levels of those scoring nine would have included large, unidentifiable variance to a ceiling level of ability, whereas those scoring between zero and eight had a clearly identified level of lower ability.

---

[1] The self-help subscale is calculated by summing three items regarding independent washing, dressing and feeding ability. Each item is scored on a three point Likert scale ranging from one (not at all) to three (without help), resulting in total scores ranging from three to nine.

After applying the exclusion criteria detailed above, 321 (49.84%) individuals were included in the between group analyses. Participants were aged between 4 and 62 (mean = 12.92; SD = 8.45), 276 (86.0%) were male and 273 (85.0%) were able/partly able (score above six but below nine on the self help subscale of the Wessex Scale). The majority of participants were fully mobile (N = 305; 95.0%) and 270 (84.1) were verbal (more than thirty words/signs in their vocabulary). Table 2.1 describes the characteristics of each participant group. Significant differences were found between the groups for age, gender and total SCQ score.

Table 2.1 Mean age (standard deviation) and range, percentage of males, percentage of participants who were able, mobile and verbal, mean SCQ total score (standard deviation) and range for between group analyses.

		ASD	**Down Syndrome**	**Fragile X Syndrome**	**F/χ^2**	**Df**	***p* value**	**Post hoc analyses (Scheffe/chi square)**
N		149	49	123				
Age	Mean (*SD*) Range	9.98 (*4.86*) 4-39	15.84 (*12.59*) 4-62	15.32 (*8.74*) 6-47	18.76	2	<.001	DS, FraX > ASD
Gender	% male	88.6	42.9	100.0*	96.50	2	<.001	FraX > ASD > DS
Self Help	% partly able/able	83.2	85.7	87.0	0.773	2	.679	N/A
Mobility	% mobile	95.3	91.8	95.9	-**	-**	.506	N/A
Speech	% verbal	82.4	85.7	86.2	.794	2	.672	N/A
SCQ total score	Mean (*SD*) Range	26.46 *(5.46)* 15 - 38	12.80 *(7.85)* 2 - 33	21.36 *(6.32)* 1 – 38	92.41	2	<.001	ASD > FraX > DS

* Only male participants with Fragile X syndrome were recruited for the study
** Fishers exact T calculated as one cell had expected count < 5.

2.3.3.2 Secondary analysis: 'more able' individuals

Participants with ASD who scored the maximum score of nine on the Wessex, and were therefore excluded from the analysis above, were included in this secondary analysis of 'more able' individuals with ASD. In total, 83 individuals with ASD were identified as scoring nine on the Wessex self-help scale[2]. From within this group, 20 participants were identified as displaying self-injury. In order to test for differences between those who displayed self-injury and those who did not, a matched comparison group was selected from the remaining 63 participants who did not display self-injurious behaviour. This group were matched on age (+/-2 years) and gender, and by virtue of the selection criteria, was also comparable on Wessex self-help score. Importantly, the groups did not differ in level of ASD phenomenology as measured by the SCQ.

After matching, 40 individuals were included in the analysis. Participants were aged between 7 and 23 (mean = 13.45; SD = 4.36), 30 (75.0%) were male and 40 (100.0%) were able/partly able (score above six on the self help subscale of the Wessex Scale). All participants were fully mobile (N = 40; 100.0%) and 39 (97.5) were verbal (more than thirty words/signs in their vocabulary). Table 2.2 describes the characteristics of both groups. No significant differences were found between the groups.

[2] A score of 9 indicates that the individual can independently feed, wash and dress themselves.

Table 2.2 Mean age (standard deviation) and range, percentage of males, self help scores, percentage of participants who were mobile, verbal, could read, write and count and total SCQ scores for each group.

		ASD SIB	**ASD No SIB**	**t/χ^2**	**Df**	***p* value**
N		20	20			
Age	Mean	13.60	13.30	-.215	38	.831
	(*SD*)	*(4.36)*	*(4.46)*			
	Range	7 - 23	7 - 23			
Gender	% male	75.0	75.0	.000	1	1.00
Mobility	% mobile	100	100	N/A*	N/A*	N/A*
Speech	% verbal	95.0	100	-**	-**	1.00
Reading	% can read	80.0	95.0	-**	-**	.342
Writing	% can write	90.0	90.0	-**	-**	1.00
Counting	% can count	90.0	100	-**	-**	.487
SCQ total scores	Mean	25.86	25.00	-.550	38	.586
	(SD)	*(4.80)*	*(5.20)*			
	Range	15 – 33.4	17 - 34			

* no statistical value calculated as mobility is evident in all participants in both groups.
** Fishers exact t calculated as 50% of cells had expected count < 5

2.3.4 Measures

The questionnaire pack included the following informant based questionnaire measures which are all appropriate for children and adults with intellectual disabilities.

A demographic questionnaire that required information on date of birth, gender, mobility, verbal ability and diagnosis.

The Wessex (Kushlick *et al.,* 1973) was used to assess adaptive ability in children and adults with intellectual disabilities. It comprises five subscales including: continence, mobility, self help skills, speech and literacy. For the purpose of this study, the self help subscale was used as an estimate of degree of ability, and responses to items on mobility, speech, reading, writing and counting were used to further describe the groups. The Wessex Scale has good inter-rater reliability at subscale level for both children and adults (Kushlick *et al.,* 1973; Palmer & Jenkins, 1982).

The Mood Interest and Pleasure Questionnaire – Short form (MIPQ-S; Ross & Oliver, 2003) was included to assess affect. It comprises twelve items, forming two subscales: Mood, and Interest and Pleasure. The measure has good internal consistency (Cronbach's alpha coefficients: total = .88, Mood = .79, Interest and Pleasure = .87), test-retest (.97) and inter-rater reliability (.85).

The Activity Questionnaire (TAQ; Burbidge *et al.,* 2010) was included to assess behaviours indicative of overactivity and impulsivity. The measure has eighteen items which form three subscales of Overactivity, Impulsivity and Impulsive Speech. Item level inter-rater reliability ranges from .31 to .75 (mean .56) and test-retest reliability ranges from .60 to .90 (mean .75). Inter-rater and test-retest reliability indices for subscales and total score exceed .70.

The Social Communication Questionnaire – Lifetime Version (SCQ; Berument *et al.*, 1999) was included to assess ASD behaviours. The SCQ was developed as a tool for screening for ASD in children and adults and is based on the Autism Diagnostic Interview and asks questions about the individual's entire developmental history (Lord, Rutter & Lecouteur, 1994). The measure consists of 40 items which are grouped into three subscales: Communication; Social Interaction and Repetitive and Stereotyped patterns of Behaviours. The authors identify a cut off score of 15 as indicative of autistic spectrum disorder and a higher cut off of 22 to differentiate between individuals with autism and those with other Pervasive Developmental Disorders. The SCQ shows good concurrent validity with the Autism Diagnostic Interview and with the Autism Diagnostic Observation Schedule (Howlin & Karpf, 2004). Internal consistency is also good (α = .90 for the total scale). The Fragile X syndrome group completed an earlier version of the SCQ (Autism Screening Questionnaire; ASQ) which had the same items and subscales, calculated using the same procedure as the SCQ. Consequently, the ASQ data for the Fragile X syndrome group are comparable to the SCQ data for the ASD and Down syndrome groups.

In calculating the total score for the SCQ, item 17 was removed from analysis (‘has she/he ever injured her/himself deliberately, such as biting her/his arm or banging her/his head?’) to prevent confounds in the data. Therefore, all individuals in the ASD group who were included in the analysis, scored above the cut off for ASD *without* including their score on item 17.

The Repetitive Behaviour Questionnaire (RBQ; Moss, Oliver, Arron, Burbidge & Berg, 2009) and the Challenging Behaviour Questionnaire (CBQ; Hyman *et al.,* 2002) were also included. The RBQ comprises five subscales: Stereotyped Behaviour, Compulsive Behaviour, Insistence on Sameness, Restricted Preferences and Repetitive Speech. Previous examination of the psychometric properties of the RBQ (Moss *et al.,* 2009) reveals that it has good inter-rater reliability coefficients (range .46 - .80) and test-retest reliability (range .61 - .93; Moss *et al.,* 2009).

The CBQ (Hyman *et al.,* 2002) evaluates the presence of self-injury, physical aggression, destruction of property and stereotyped behaviour in the last month. The measure also examines eight topographies of self-injury that were adapted from Bodfish *et al* (1995). Items evaluating self-injury 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).

The orders of the measures in the questionnaire pack were counterbalanced across the group to reduce order effects.

2.3.5 Data analysis

Data were tested for normality using Kolmogorov–Smirnov tests. Where data were not normally distributed ($p<.05$), non-parametric techniques were employed.

To investigate the prevalence and topography of self-injury in ASD, the percentage of each group showing self-injury and specific topographies of self-injury were derived from the CBQ. Relative risk statistics were then calculated comparing the likelihood of individuals in each group showing self-injury and the specific topographies, compared to the likelihood of individuals in the other groups showing self-injury and the specific topographies. For all of these analyses, individuals who do not display self-injury were included, with scores of zero In order to compare the severity of self-injury across groups, a severity score was calculated from the CBQ by summing three items regarding the length of self-injurious behaviour bursts, the level of intervention required and the frequency of these bursts. A Kruskal-Wallis test was conducted in order to test for differences between the groups.

The differences between levels of ASD behaviour in those who engaged in self-injury and those who did not within each group was investigated through a series of Mann Whitney U tests upon the presence of self-injury and SCQ subscale and total scores.

To investigate variables associated with self-injury within ASD, participants with ASD showing self-injury were compared to participants with ASD who did not display the behaviour. The difference between variables contingent on the presence of self-injury was then examined. Chi-square statistics were applied to categorical data and Mann-Whitney U tests for ordinal data. To further examine the associations between person characteristics and self-injury, the association between the severity of self-injury score and significant person characteristics was evaluated within the ASD sample. Spearman's Rho correlations were applied to continuous data and Mann-Whitney U tests were employed for ordinal data.

To conduct the secondary analysis for the matched case-control ASD group, the 20 individuals showing self-injury were compared to the matched 20 individuals who did not display self-injury. The difference between variables contingent on the presence of self-injury was then examined using Mann Whitney U tests. Finally, a binary logistic regression was conducted to analyse whether the differing variables between these groups would predict the likelihood of an individual displaying self-injury.

2.4 Results

2.4.1 Prevalence, topography and severity of self-injurious behaviour

In order to test the first hypothesis of the study, prevalence data and relative risk statistics were calculated to compare the likelihood of each group displaying any form of self-injury and specific topographies of self-injury compared to the other groups. Additionally, the severity of self-injury score from the CBQ was compared across the three groups.

Table 2.3 displays the percentage of individuals showing self-injury in all groups and relative risk statistics of the likelihood of individuals in each group showing self-injury compared to the other groups. 50% of individuals in the ASD group had engaged in self-injury in the preceding month. The ASD group was 2.67 times more likely to show self-injury than the Down syndrome group, and the Fragile X syndrome group was 2.91 times more likely to show self-injury than the Down syndrome group. There were no significant differences in the likelihood of displaying self-injury between the ASD and Fragile X syndrome groups.

Table 2.3 Percentage of individuals showing self-injury in each group. Relative risk statistics and 99% confidence intervals are shown to demonstrate the likelihood of individuals in each group showing self-injury compared to the other groups. Bold text indicates a significant difference ($p<.01$)

Test Group	N	Percentage showing SIB	RR compared to ASD (99% CI's)	RR compared to Down syndrome (99% CI's)	RR compared to Fragile X syndrome (99% CI's)
ASD	148	50.0	**-**	**2.67 (1.19 – 5.96)**	0.92 (0.68 – 1.24)
Down syndrome	49	18.4	**0.38 (0.17 – 0.84)**	-	**0.34 (0.15 – 0.77)**
Fragile X syndrome	123	54.5	1.09 (0.81 – 1.47)	**2.91 (1.30 – 6.49)**	**-**

Table 2.4 displays the percentage of individuals showing each topography of self-injury in all groups. Additionally, it shows the relative risk statistics of the likelihood of individuals in each group showing each topography of self-injury compared to the other groups. Individuals in the ASD group were 4.79 times more likely to show self-injury that involved hitting their own body than the Down syndrome group. Individuals in the Fragile X syndrome group were 2.52 times more likely to show self biting behaviour that individuals in the ASD group, and 7.67 times more likely to show self-biting than individuals in the Down syndrome group.

A Kruskal-Wallis test was conducted in order to test for differences in severity of self-injury between individuals with ASD, Fragile X syndrome and Down syndrome. The test revealed no significant differences in severity between individuals who engaged in self-injury in any of the groups (Kruskal Wallis χ^2 (2) = .976).

In summary, individuals with ASD were more likely to engage in self-injury than the Down syndrome group, but are no more likely to engage in self-injury than the Fragile X syndrome group. Individuals with ASD were more likely to engage in hitting their own body than the Down syndrome group. The Fragile X syndrome group were more likely to engage in self-biting than the ASD and Down syndrome groups. Finally, there were no significant differences identified between the groups on measures of severity of self-injury.

Table 2.4 Prevalence of topographies of self-injury for each group. Relative risk statistics and 99% confidence intervals to demonstrate the likelihood of individuals in each group showing a specific topography of self-injury compared to the other groups. Bold text indicates a significant difference (p<.01).

	Hits self with body	Hits self against object	Hits self with object	Bites self	Pulls self	Rubs/ scratches self	Inserts
Prevalence of topographies							
ASD	29.5	15.4	10.7	18.8	11.4	16.8	6.0
Down syndrome	6.1	8.2	0.0	6.1	4.1	10.2	10.2
Fragile X syndrome	25.2	11.4	3.3	48.0	14.6	14.6	8.1
Relative Risk compared to Down syndrome group							
ASD	**4.79 (1.09 – 21.01)**	1.88 (0.50 – 7.10)	-	3.05 (0.67 – 13.76)	2.78 (0.42 – 18.20)	1.63 (0.50 – 5.36)	0.59 (0.15 – 2.32)
Fragile X syndrome	4.03 (0.90 – 18.02)	1.37 (0.34 – 5.51)	-	**7.67 (1.78 – 33.13)**	3.51 (0.54 – 22.84)	1.40 (0.41 – 4.80)	0.78 (0.20 – 2.99)
Relative Risk compared to Fragile X syndrome group							
ASD	1.19 (0.71 – 1.99)	1.37 (0.61 – 3.11)	3.35 (0.82 – 13.67)	**0.40 (0.24 – 0.66)**	0.79 (0.35 – 1.78)	1.16 (0.56 – 2.42)	0.75 (0.24 – 2.36)
Down syndrome	0.25 (0.06 – 1.11)	0.73 (0.18 – 2.95)	-	**0.13 (0.03 – 0.56)**	0.28 (0.04 – 1.85)	0.71 (0.21 – 2.43)	1.28 (0.33 – 4.91)
Relative Risk compared to ASD group							
Down syndrome	**0.21 (0.05 – 0.92)**	0.53 (0.14 – 2.01)	-	0.33 (0.07 – 1.48)	0.36 (0.05 – 2.36)	0.61 (0.19 – 2.01)	1.70 (0.43 – 6.72)
Fragile X syndrome	0.84 (0.50 – 1.41)	0.73 (0.32 – 1.64)	0.30 (0.07 – 1.22)	**2.52 (1.53 – 4.16)**	1.27 (0.56 – 2.85)	0.86 (0.41 – 1.79)	1.33 (0.42 – 4.16)

- = incalculable due to an empty cell

2.4.2 Difference in autism spectrum disorder behaviour between those who self-injure and those who do not

In order to test the second hypothesis of the study, median scores were calculated for all subscales of the SCQ for those who engaged in self-injury and those who did not, within each group[3]. Figure 2.1 displays the median, maximum and minimum scores and significant differences within groups.

Within all groups, those who engaged in self-injury had higher scores on all subscales and the total score of the SCQ, in comparison to those who did not engage in self-injury. At the total score level, this difference was significant for the Fragile X syndrome (Mann Whitney U = 2348.0, p = .008, one tailed, Cliff's *d* = .25[4]) and Down syndrome (Mann Whitney U = 276.0, p = .004, one tailed, *Cliff's d* = .57) groups. Additionally, within the Fragile X syndrome group, individuals who engaged in self-injury had significantly higher scores on the social interaction subscale (Mann Whitney U = 2363.5, p = .004, one tailed, Cliff's *d* = .26).

In summary, across all groups, those who engaged in self-injury had higher scores on the SCQ total score than those who did not engage in self-injury. This difference was significant for the Fragile X syndrome and Down syndrome groups.

[3] As above, item 17 (has she/he ever injured her/himself deliberately, such as biting her/his arm or banging her/his head?) was not included in any subscale or total score.

[4] Cliff's dominance (or *d*) statistic (1993) was used to calculate effect sizes for Mann Whitney U tests. Cliff's *d* statistic can be used to estimate the size of the differences between two distributions that have been shown to be significantly different with Mann Whitney U. A *d* value of +1 would indicate that every datum point in a series is greater than every other datum point in the other series. A *d* value of -1 would indicate that every datum point in a series is less than every other datum point in the other series. Arbitrary cut offs for effect strengths were assigned as follows: .0 - .4 = weak, .4 - .8 = moderate, .8 – 1.0 = strong.

Figure 2.1 Median, maximum, minimum and inter-quartile range of SCQ subscale and total scores indicating level of autistic behaviour for those who engage in self-injury and those who do not, for all groups.

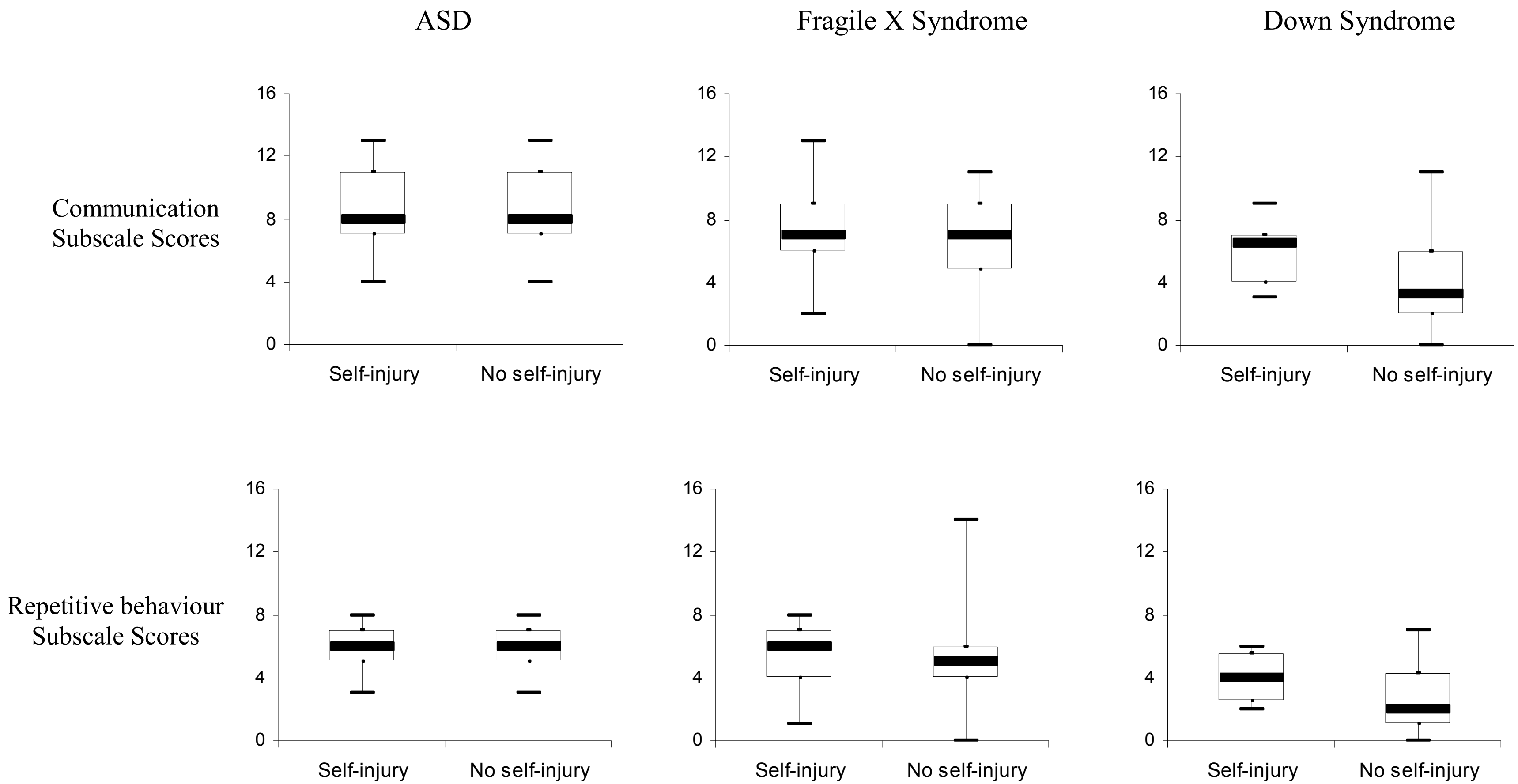
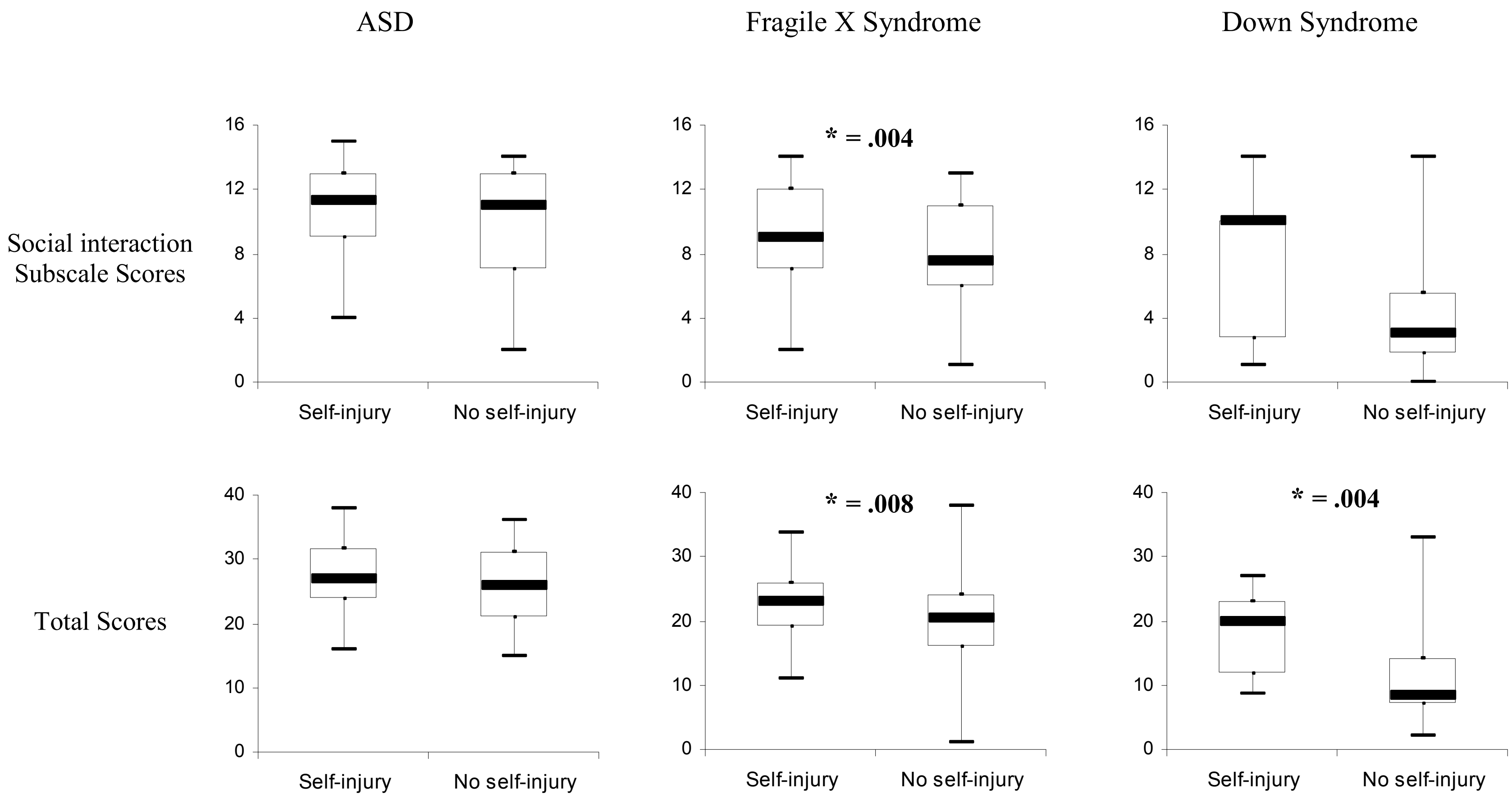


Figure 2.1 cont. Median, maximum, minimum and inter-quartile range of SCQ subscale and total scores indicating level of autistic behaviour for those who engage in self-injury and those who do not, for all groups. Significant difference in subscale score between those who engage in self-injury and those who do not are highlighted in bold (p < .01).



2.4.3 Differences in demographic and behavioural variables dependant on presence of self-injury in autism spectrum disorder

In order to test the third hypothesis, individuals with ASD who displayed self-injury were compared to individuals with ASD who did not display self-injury on a number of demographic and behavioural variables.

Table 2.5 shows the demographic variables for ASD participants who engaged in self-injury and revealed that there were no significant differences in gender, age, mobility, vision or hearing between those with self-injury and those without self-injury. However, significantly more individuals who engaged in self-injury were non-verbal. The difference in ability level between those with self-injury and those without self-injury approached significance.

Table 2.5 Demographic variables for ASD participants with and without self-injury. Chi square statistics for comparison on demographic variables for ASD participants with and without self-injury. Significant differences (p<.01) are highlighted in bold; all tests are two tailed apart from level of ability and speech).

		Percentage of individuals with self-injury *(N)*	**Percentage of individuals without self-injury *(N)***	**Chi-square**	***P* value**
Gender	Male	87.8 *(65)*	89.2 *(66)*	0.066	.797
	Female	12.2 *(9)*	10.8 *(8)*		
Age	≤11 years	63.0 *(46)*	74.0 *(54)*	-*	.218
	12-18 years	28.8 *(21)*	23.3 *(17)*		
	≥ 19 years	8.2 *(6)*	2.7 *(19)*		
Ability	Able/ Partly Able	77.0 *(55)*	89.2 *(66)*	3.899	.024
	Not Able	23.0 *(17)*	10.8 *(8)*		
Speech	Verbal/ Partly verbal	80.8 *(59)*	94.6 *(70)*	**6.487**	**.006**
	Non-Verbal	19.2 *(14)*	5.4 *(4)*		
Mobility	Mobile	87.7 *(64)*	94.6 *(70)*	2.185	.139
	Non-mobile/ Partly mobile	12.3 *(9)*	5.4 *(4)*		
Vision	Normal	93.2 *(69)*	98.6 *(73)*	-*	.209
	Poor Vision/ Blind	6.8 *(5)*	1.4 *(1)*		
Hearing	Normal	93.2 *(69)*	98.6 *(73)*	-*	.209
	Poor Hearing/ Deaf	6.8 *(5)*	1.4 *(1)*		

* Fishers exact T was calculated

Secondly, scores on measures of mood, repetitive behaviour and activity level were compared between those with ASD who engaged in self-injury and those who did not. Table 2.6 shows the median scores and Mann Whitney statistics for measures of affect, repetitive behaviour and activity level. Those who engaged in self-injury had significantly higher impulsive speech scores than those who did not engage in self-injury, although this difference has a weak effect size. Additionally, the difference between overactivity and impulsivity scores between those who engaged in self-injury and those who did not are approaching significance. There were no differences in affect or repetitive behaviour between the two groups, although the differences in mood and interest and pleasure also approached significance.

Table 2.6 Median scores and Mann-Whitney statistics for measures of affect, repetitive behaviour and activity levels for ASD participants with and without self-injury. Bold text indicates a significant difference (p<.01, one tailed).

Measure Subscale	**Median scores *(interquartile range)***		**U score**	***P* value**	**Cliff's *d***
	With self-injury (N = 74)	Without self-injury (N= 74)			
MIPQ-S					
Mood	18.50 *(16.00 – 21.00)*	19.00 *(18.00 – 21.00)*	2249.00	.030	-
Interest and Pleasure	14.00 *(11.00 – 16.00)*	15.00 *(12.00 – 18.00)*	2241.00	.028	-
RBQ					
Stereotyped behaviour	9.50 *(5.00 – 12.00)*	8.00 *(4.00 – 11.00)*	3061.00	.106	-
Compulsive behaviour	9.00 *(3.32 – 16.25)*	6.00 *(3.00 – 14.00)*	3076.50	.097	-
Insistence on sameness	4.50 *(3.00 – 7.00)*	4.00 *(2.00 – 7.00)*	2906.00	.133	-
Restricted preferences*	4.00 *(1.50 – 8.50)*	5.00 *(3.25 – 8.00)*	1710.00	.276	-
Repetitive language*	7.00 *(4.00 – 10.00)*	7.00 *(3.25 – 11.00)*	1886.50	.372	-
TAQ					
Overactivity	23.00 *(16.00 – 29.00)*	18.00 *(10.00 – 28.25)*	3331.50	.012	-
Impulsivity	20.00 *(16.00 – 22.00)*	18.00 *(11.50 – 21.00)*	3177.00	.022	-
Impulsive Speech*	7.00 *(4.00 -10.00)*	4.00 *(3.00 – 9.00)*	**2290.50**	**.008**	**.26**

* Subscales only calculated for verbal participants

In summary, those who engage in self-injury were significantly more likely to be non-verbal. Additionally, individuals who engaged in self-injury had significantly higher impulsive speech scores.

2.4.3.1 Association between severity of self-injury and demographic and behavioural variables in autism spectrum disorder

In order to test the final component of the third hypothesis of the study, tests were employed to evaluate an association between the self-injury severity score and identified person characteristics. Person characteristics which were significantly associated with the presence of self-injury and person characteristics which approached a significant association with the presence of self-injury were included in this analysis (see above). Table 2.7 reveals that self-injury severity scores were correlated significantly with lower mood and interest and pleasure and higher overactivity and impulsive speech, with weak correlation coefficients. The correlation between impulsivity and self-injury severity approaches significance.

Table 2.7 Spearman's Rho correlation coefficients for associations between self-injury severity scores, affect and activity levels. Bold text indicates a significant difference ($p<.01$, one tailed).

Measure Subscale	Spearman's Rho Correlation	*P* value
MIPQ-S		
Mood	**-.196**	**.008**
Interest and Pleasure	**-.194**	**.009**
TAQ		
Overactivity	**.211**	**.005**
Impulsivity	.146	.039
Impulsive Speech*	**.242**	**.004**

* Subscales only calculated for verbal participants

The Mann Whitney U tests revealed that individuals who were categorised as 'not able' evidenced significantly higher self-injury severity scores (Mann Whitney U =992.50, p =.001, Cliff's *d* =54.). Individuals who were categorised as 'non-verbal' evidenced significantly higher self-injury severity scores (Mann Whitney U =615.50, p <.001, Cliff's *d* =.66).

2.4.4 Matched case-control in 'more able' individuals with autism spectrum disorder

In order to evaluate the final aim of the study, a matched case-control analysis within a subsample of the ASD group who scored the maximum score of nine on the Wessex self-help scale was conducted. Within the total sample (N = 83) of those who scored at ceiling on the Wessex, 24.1% (N = 20) engaged in self-injury.

Table 2.8 shows the median scores and Mann Whitney statistics for measures of affect, repetitive behaviour and activity level. As with the total sample, there was no difference in affect between those who engaged in self-injury and those who did not, however the difference in mood approaches significance. Secondly, those who engaged in self-injury had significantly higher scores on measures of compulsive behaviour and insistence on sameness with moderate effect size. Finally, those who engaged in self-injury had significantly higher overactivity, impulsivity and impulsive speech scores than those who did not engage in self-injury. The differences in impulsivity and impulsive speech have a moderate effect size, whilst the difference in overactivity has a strong effect size.

Table 2.8 Median scores and Mann-Whitney statistics for measures of affect, repetitive behaviour and activity levels for ASD participants with and without self-injury. Bold text indicates a significant difference (p<.01, one tailed)

Measure Subscale	**Median scores *(interquartile range)***		**U score**	***P* value**	**Cliff's *d***
	With self-injury (N = 20)	Without self-injury (N = 20)			
MIPQ-S					
Mood	17.00 *(13.25 – 20.00)*	20.00 *(17.25 – 21.00)*	120.00	.015	-
Interest and Pleasure	11.50 *(7.00 – 15.00)*	13.50 *(8.25 – 16.50)*	167.50	.189	-
RBQ					
Stereotyped behaviour	5.00 *(2.00 – 9.00)*	2.00 *(0.00 – 6.75)*	255.50	.031	-
Compulsive behaviour	8.00 *(4.00 – 20.57)*	1.50 *(0.00 – 11.00)*	**283.50**	**.004**	**.44**
Insistence on sameness	4.00 *(3.00 – 7.00)*	2.00 *(0.00 – 5.50)*	**277.00**	**.007**	**.46**
Restricted preferences	7.00 *(3.75 – 9.25)*	3.50 *(1.25 – 7.00)*	234.50	.054	-
Repetitive language	6.00 *(3.00 – 7.75)*	2.50 *(0.00 – 6.00)*	249.50	.020	-
TAQ					
Overactivity	20.57 *(12.25 – 27.00)*	7.50 *(3.25 – 11.00)*	**342.00**	**<.001**	**.71**
Impulsivity	18.50 *(13.75 – 21.75)*	13.00 *(5.75 – 17.75)*	**297.00**	**.005**	**.49**
Impulsive Speech	9.00 *(5.00 – 11.00)*	4.00 *(3.00 – 6.75)*	**283.00**	**.005**	**.42**

In summary, 24.1% of the individuals scoring at ceiling on the Wessex engaged in self-injury. The individuals who engaged in self-injury had significantly higher compulsive behaviour, instance on sameness, overactivity and impulsivity scores.

2.4.4.1 Predictive value of variables associated with self-injurious behaviour

Finally, in order to ascertain the contribution of repetitive behaviour and activity level to self-injury within this 'more able' group, a logistic regression was conducted. Two composite scores were created: an Overactivity\Impulsivity composite (the sum of Overactivity, Impulsivity and Impulsive speech scores) and a Repetitive Behaviour composite (the sum of Compulsive behaviour and Insistence on sameness).

Binary logistic regression was performed to assess the impact of these two factors on the likelihood of participants displaying self-injury. The model contained two independent variables (Overactivity/Impulsivity composite and Repetitive Behaviour composite). The full model containing all predictors was statistically significant, (χ^2 (2, N = 39) = 18.66, $p < .001$), indicating that the model was able to distinguish between participants displaying self-injury and participants not displaying self-injury. The model as a whole explained between 38.0% (Cox and Snell R square) and 50.7% (Nagelkerke R squared) of the variance in self-injury status, and correctly classified 69.2% of cases.

As shown in Table 2.9, only one of the independent variables made a unique statistically significant contribution to the model (Overactivity/Impulsivity composite). The odds ratio for the Overactivity/Impulsivity composite was 1.11, suggesting that individuals with high levels of overactivity and impulsivity were 1.11 times more likely to show self-injurious behaviour, controlling for other factors in the model.

Table 2.9 Logistic regression predicting likelihood of displaying self-injury

	B	**S.E**	**Wald**	*df*	*p*	**Odds Ratio**	**95.0% C.I. for Odds Ratio**	
							Lower	Upper
Overactivity/Impulsivity	0.10	.04	8.11	1	**.004**	**1.11**	**1.01**	**1.22**
Repetitive Behaviour	0.04	.05	0.76	1	.383	1.04	0.92	1.19

In summary, the logistic regression revealed that, independently of other factors, high levels of overactivity and impulsivity significantly increase the likelihood of an individual displaying self-injurious behaviour.

2.5 Discussion

The prevalence, topography and severity of self-injury in a group of individuals with ASD were examined in this study in comparison to individuals with Down syndrome and Fragile X syndrome. The difference in ASD behaviour levels relative to the presence of self-injury was explored within each group and the characteristics of those with ASD who engaged in self-injury were also investigated. Finally, these associated characteristics were then examined in a matched ability, gender and age subsample of the ASD group. Importantly, the utilisation of relatively homogenous groups for both within and between group analyses strengthens the validity of this study. The inclusion of multiple comparison groups enables consideration of the specificity of findings to those with ASD. The employment of standardised measures across large groups allows the prevalence of self-injury in ASD to be considered in comparison to the prevalence of self-injury in the other groups. Additionally, the utilisation of an ASD screen for the ASD population increases the internal validity of the study, and ensures a robust estimate of the prevalence, topography and severity of self-injury in ASD. Finally, the addition of analysis within a 'more able', ability, age and gender matched group enables the findings to be considered without the confound of ability level and also contributes towards an understanding of self-injury in 'more able' individuals with ASD.

The primary between syndrome groups analysis revealed that the ASD group evidenced a higher prevalence and relative risk of self-injury than the Down syndrome group. Within all groups, those engaging in self-injury obtained higher scores on measures of autistic behaviour, with the self-injury Fragile X syndrome and Down syndrome groups scoring significantly higher on the total score for autistic behaviour. Those with ASD who engaged in self-injury had higher scores on measures of impulsive speech. Severity of self-injury was significantly associated with higher levels of overactivity and impulsive speech and lower levels of mood and interest and pleasure, suggesting that these factors may be associated with self-injury in ASD. When these factors were re-examined in the 'more able' subsample, those who engaged in self-injury obtained significantly higher overactivity and impulsivity scores, alongside higher repetitive behaviour scores. Finally, higher scores on a composite of the activity and impulsivity scores were revealed to independently predict an increase in the likelihood of an individual with ASD displaying self-injury.

<u>Primary between groups analysis:</u>

The results indicate a relatively high prevalence rate of 50.0% for self-injury in individuals with ASD, which is consistent with rates reported in other studies (Ando & Yoshimura, 1979a; Baghdadli *et al.,* 2003; Dominick, Davis, Lainhart, Tager-Flusberg & Folstein, 2007; Shattuck *et al.,* 2007). However, the prevalence rate is considerably lower than that reported by Bartak and Rutter (1976) perhaps because their data are from a subset of individuals with ASD and an average IQ of 45.7, and lower ability level has been consistently associated with higher prevalence of self-injury (McClintock *et al*., 2003; Collacott *et al.,* 1998; Schroeder, Schroeder, Smith & Dalldorf, 1978). The results of this study converge with previous research revealing self-injurious behaviour to occur in between 40-50% of individuals with ASD. Importantly, this study extends prior research by calculating the heightened risk of self-injury in ASD compared to individuals with intellectual disability who do not have ASD. The results of this study demonstrated that the ASD group were 2.67 times more likely to engage in self-injury than the Down syndrome group, supporting previous research indicating a heightened risk of self-injury in ASD (McClintock *et al,* 2003). Interestingly, the results also revealed that individuals with ASD were 4.79 times more likely to engage in self hitting with their own body than individuals with Down syndrome. However, there were no significant difference in this topography between the ASD group and the Fragile X syndrome group.

Therefore this finding requires further investigation, to delineate whether the difference is reflective of relative homogeneity of topography of self-injury in the ASD group, or of a low prevalence of this topography in the Down syndrome group. Consistent with previous research, 54.4% of individuals with Fragile X syndrome engaged in self-injury and these individuals were 2.91 times more likely to self-injure than individuals with Down syndrome. Converging with robust findings in other studies, individuals with Fragile X syndrome were significantly more likely to engage in self-biting than both the ASD and Down syndrome groups (Hall *et al.,* 2008), suggesting a genetic specificity to their self-injury. Finally, in line with previous research, the prevalence of self-injury in Down syndrome was substantially lower than the prevalence in either of the two other groups at 18.4%. This prevalence figure is comparable to that identified in the general intellectual disability population, and consequently reinforces the utility of making comparisons between these three groups.

In addition to the heightened risk of self-injury within the ASD group, the association between autistic characteristics and self-injury was explored across all three groups. Supporting findings from previous research, those who engaged in self-injury attained higher total scores on the SCQ (Baghdadli *et al.,* 2003; Bhaumik *et al.,* 1997; Matson and Rivet, 2008). However, this difference was only significant for the Fragile X syndrome and Down syndrome groups. The non-significant finding within the ASD group may be due to the careful diagnostic screening process employed within this study, comparative to other studies. Nonetheless, the trend for all groups was for higher total scores being associated with self-injurious behaviour. This result suggests that the concept of ASD *diagnosis* as a risk marker for self-injury needs to be broadened and re-conceptualised. The presence of high levels of ASD type behaviour, rather than a diagnosis of ASD per se, is associated with self-injury.

This distinction has clinical utility when considering risk for self-injury within non-ASD populations, such as those with genetic syndromes.

At a subscale level, the trend was for higher scores on all subscales to be associated with self-injury across all groups. However, the only significant results were found in the Fragile X syndrome group for the Social Interaction subscale. This result differs from the results reported by Matson and Rivet (2008), who demonstrated that restricted and repetitive behaviour was significantly associated with self-injury in an ASD population. This difference in findings may be due to the screening of the ASD population in this study, or the low inter-rater and test-retest reliability of the autistic behaviour measure used by Matson and Rivet. The results of this study suggest that for the ASD population, none of the triad of impairments is specifically associated with self-injury. This is particularly interesting in the area of repetitive behaviour, given the empirical evidence of a temporal association between stereotyped behaviours, proto-injurious behaviour and self-injurious behaviour (Petty *et al.,* 2009; Richman & Lindauer, 2005). The lack of difference in repetitive behaviour between those who self-injure and those who do not, may reflect a progression from stereotyped and repetitive behaviours, towards self-injurious behaviours that are more reliably reinforced and rewarded by caregivers (Guess & Carr, 1991). Thus, stereotyped and repetitive behaviours would reduce in frequency as self-injury became more functionally efficient within an individual's behavioural repertoire. This hypothetical model would predict the absence of difference in repetitive behaviour found in this study.

Additionally, it is of interest that for the Fragile X syndrome group, individuals who engaged in self-injury had significantly higher levels of impairment in social interaction but no significant differences in communication. A recent review of ASD phenomenology in genetic syndromes highlighted that deficits in social interaction contribute most strongly to an individual with Fragile X syndrome meeting criteria for ASD (Moss & Howlin, 2009). It could therefore be hypothesised, that for these individuals, ASD phenomenology is most apparent in the domain of social interaction, and consequently, it is these behaviours that are most strongly associated with self-injury.

A within group analysis between those with ASD who self-injure and those who do not was conducted, in order to reveal variables that may contribute to the high prevalence of self-injury in ASD. The results revealed that individuals with ASD who engaged in self-injury had significantly higher impulsive speech scores, and higher scores on measures of impulsivity and overactivity which approached significance. Additionally, individuals who engaged in self-injury evidenced lower levels of ability, mood and interest and pleasure; these differences all approached significance. Prior to a consideration of the statistically significant findings, the results that approached significance warrant further discussion.

Throughout the thesis, conservative alpha levels have been selected in order to avoid making Type 1 errors. However, given the important clinical implications of low mood and ADHD type behaviours, a pragmatic decision will be made to make less conservative inferences based on these data which approach statistical significance. Whilst the probability of making a Type 1 error is increased by this, this concern is outweighed by the concurrent decrease in the probability of making a Type 2 error. Whilst the over identification of differences in mood and ADHD type behaviours are not ideal, the under identification of this potential difference has far more significant clinical implications. Thus, the differences in mood, interest and pleasure, overactivity and impulsivity that approached significance will be treated as warranting further investigation and will be assessed in later Chapters. This decision is supported by the statistically significant finding that lower mood and interest and pleasure and higher levels of overactivity and impulsive speech are all significantly correlated with more severe self-injury. This suggests that these clusters of person characteristics are significantly associated with clinically severe self-injury.

Thus, the findings in this chapter can be seen to provide preliminary support to studies that associated overactivity, impulsivity and self-injury (Cooper *et al.,* 2009; Collacott *et al.,* 1998; Oliver *et al.,* 2009; Schneider *et al.,* 1996). This provides tentative evidence for an executive dysfunction account of self-injury where the elevated levels of impulsivity and overactivity constitute behavioural indicators of impaired inhibition. This may interact with an operant account of self-injurious behaviour, in which impairments in inhibition result in reinforcer delay being less tolerable. Consequently a greater proportion of antecedents will lead to self-injury being displayed.

The analyses also revealed an association between lower speech levels and self-injury within the ASD group. This association may be indicative of lower levels of ability in those who self-injure, and consequently it is plausible that the differences in overactivity and impulsivity are simply a reflection of differences in ability levels.

<u>'More Able' subsample analysis:</u>

In order to tease apart the relationship between self-injury, ability levels and overactivity/impulsivity, a final analysis was conducted upon individuals with ASD who scored at ceiling level on the Wessex. This matched case-control allowed differences in mood, repetitive behaviour and activity behaviours to be re-examined, whilst holding ability level constant within a 'more able' group. The results revealed significantly higher levels of compulsive behaviour, insistence on sameness, overactivity, impulsivity and impulsive speech in those who engaged in self-injury. There was also a trend towards lower levels of mood in those who engaged in self-injury. However, this difference did not reach significance, suggesting that low mood may not be as influential in effecting the presence of self-injury as, for instance, overactivity and impulsivity.

The significant differences in repetitive and overactive/impulsive behaviours support the findings in the main sample and confirm the pragmatic decision to view differences in overactivity and impulsivity as both clinically significant. This finding lends strength to the suggestion that impaired behavioural regulation, resulting in behavioural markers such as impulsivity and compulsivity, is associated with self-injury. The results in the 'more able' sample allow the findings to be disassociated from ability level, and specifically linked to the presence of self-injury. The logistic regression demonstrated that only the overactive/impulsive component of these behaviours is significantly, independently associated with the presence of self-injury. This model correctly predicted the self-injury status of almost 70% of the sample, suggesting that when ability level is controlled for, impulsivity and overactivity contribute significantly to the likelihood of self-injury being present. This suggests that impulsive behaviours may serve as putative risk markers for self-injurious behaviour in individuals with ASD. Further research should be conducted to test experimentally this perceived difference in behavioural regulation between those who self-injure and those who do not. The confirmation of this cluster of behaviours as a risk marker for self-injury would have great clinical utility for early intervention in self-injury.

Overall findings:

A caveat that must be considered when interpreting these findings is that all individuals were recruited via parent support groups. It is possible that this may have induced a recruitment bias, as those who are in contact with support groups may have more challenging children, and consequently be more in need of support. However, as all groups were recruited in the same way, the bias should be consistent across groups, and consequently comparisons remain valid. This validity is supported by the prevalence figures for self-injury in the ASD (Baghdadli *et al.,* 2003) and Fragile X syndrome (Hall *et al.,* 2008) groups being comparable to those previously reported in the literature. Secondly, key findings relating to overactivity and impulsivity relied upon the utilisation of the Wessex (a measure of adaptive behaviour) as a proxy measure of ability. However, the employment of the measure to match the 'more able' sample is supported by further comparisons on mobility, speech, reading, writing and counting revealing no differences between the two groups. Finally, the sample size for the logistic regression calculation was small, and therefore this finding should be interpreted with caution.

Taken as a whole, these results indicate that individuals with ASD are more likely to engage in self-injury than those with Down syndrome. The results also show that individuals who engage in self-injury show significantly more behaviours indicative of ASD. Finally, the results reveal that impaired impulse control and overactivity are associated with self-injury in ASD which leads to the possibility that impairments in executive functioning may contribute to the high prevalence of self-injury in ASD.

CHAPTER 3

Risk Markers for Self-Injurious Behaviour in Autism Spectrum Disorder: A Longitudinal Study

3.1 Preface

The study in Chapter 2 compared the prevalence, topography and severity of self-injury in individuals with ASD to individuals with Down and Fragile X syndromes. Additionally, demographic and behavioural variables associated with the presence of self-injury in ASD were delineated. In summary, it was found that self-injury was displayed by 50.0% of the ASD sample and that ASD phenomenology was significantly associated with the presence of self-injury in the Fragile X and Down syndrome groups. The results also demonstrated that lower levels of expressive language and higher levels of overactivity, impulsivity and repetitive behaviour were associated with the presence of self-injury in the ASD group. Using a longitudinal design, this study will extend the findings in Chapter 2 to describe the persistence of self injury in ASD over a three year period and evaluate whether demographic and behavioural characteristics are associated with persistence.

3.2 Introduction

Prevalence estimates for self-injury are significantly higher for individuals with ASD than for individuals with intellectual disability of heterogeneous aetiology (see Sections 1.3.2 & 1.4.1). Robust data from existing research, including the study in Chapter 2 estimate the prevalence of self-injury in ASD at between 40% and 50% (See Sections 1.4.1 & 2.4.1). The presence of self-injury leads to a higher risk of psychiatric hospitalisation (Mandell, 2008), reactive physical intervention (Allen, Lowe, Brophy & Moore, 2009) and lower quality of life (Beadle-Brown, Murphy & DiTerlizzi, 2009). Carers of those who display self-injury are reported to experience higher levels of stress (Hastings, 2003; Seltzer, *et al.,* 2010) and the presence of self-injury contributes to staff burnout in care settings (Hastings & Brown, 2002). In addition to the personal costs of self-injurious behaviour, there is also significant financial cost to services (Knapp, Comas-Herrera, Astin, Beecham, & Pendaries, 2005).

Behavioural interventions for self-injury are effective, but are often resource intensive and fragile (Harvey, Boer, Meyer & Evens, 2009; Meyer & Evens, 1993; Robertson *et al*., 2005). Consequently, research attention has begun to consider the viability of an early intervention strategy for self-injurious behaviour (Richman, 2008), which is predicated on the assumption that self-injury begins during childhood/early adulthood, becomes more severe with time and persists over time (Oliver, Hall & Murphy, 2005; Taylor, Murphy & Oliver, 2006, Emerson *et al.,* 2001b).

There is conflicting evidence regarding the persistence of self-injurious behaviour, both in individuals with intellectual disability and, more specifically, in individuals with ASD. The majority of studies conducted in populations with intellectual disability have demonstrated the persistent nature of self-injury; however there are also a number of studies which appear to show high levels of remission. Emerson *et al.,* (2001b) conducted a longitudinal study to investigate the persistence of self-injury in a cohort of individuals with intellectual disability. They demonstrated that over a seven year period, 71% of individuals continued to display self-injury. Similarly, in one of the longest follow up studies from a total population sample, Taylor, Oliver and Murphy (2011) demonstrated that 84% of a sample of people with intellectual disabilities continued to display self-injury over a 20 year period. However, in a 12 year follow up of a total population study, Murphy *et al*., (2005) demonstrated that the prevalence of self-injurious behaviour significantly decreased over time. Similarly, a recent longitudinal study also reported a moderate two year remission rate in adults with intellectual disability (38.2%; Cooper *et al.,* 2009), with the authors indicating that self-injurious behaviour may not be as persistent as initially thought. However, despite a remission rate of 38.2%, 21 individuals in Cooper *et al.'s* (2009) study continued to engage in self-injury over the two year period. As this study employed particularly stringent criteria for self-injury, this equates to 61.8% of the self-injury sample continuing to show self-injurious behaviour that causes tissue damage, is pervasive, presents significant risks to the health or safety of the person, and significantly impacts upon their own or other's quality of life. Whilst the persistence figure of 61.8% is lower than others reported in the literature (Emerson *et al.,* 2001b; Taylor *et al.,* 2011), these data still suggest that for the majority of individuals, self-injury continues to be a behaviour which significantly impacts upon their lives over time.

Despite the high prevalence of self-injury in ASD, few studies have examined the persistence of self-injury in this population. A recent literature review suggests that challenging behaviours may be more stable in individuals with ASD (Totsika & Hastings, 2009). Shattuck *et al.,* (2007) conducted a longitudinal study investigating the stability of ASD phenomenology and maladaptive behaviours in a cohort of adolescents and adults with ASD.

The study revealed that the prevalence of self-injury decreased significantly over four and half years. However, the study specifically recruited individuals through service agencies and clinics and consequently may have recruited individuals who would be most likely to receive intensive intervention to reduce self-injury. Baghdadli *et al.,* (2008) report similar results in a three year follow up of children with Pervasive Developmental Disorders (PDD). At the first time point, 49% of their sample engaged in self-injury, whereas at the second time point, only 32.7% did. However, the sample included in Baghdadli *et al.'s* study (2008) were very young (mean age at follow up = 8 years) and all of the children were receiving treatment in hospitals that provided services for children with psychiatric disorders. Therefore, as with the study presented by Shattuck *et al.* (2007), it is not possible to evaluate whether the remission in self-injury is the natural course of development or a result of intensive intervention. There is a need to evaluate the persistence of self-injurious behaviour in a population with ASD that have not been recruited from clinical services. If self-injury is indentified as persistent, then an early intervention strategy may be warranted.

Early intervention strategies have been effectively implemented in health settings (Blanks, Moss, McGahan, Quinn & Babb, 2000) and more broadly in autism (Cohen, Merine-Dickens & Smith, 2006; Eikeseth, Smith, Jahr & Eldevik, 2007; Lovaas, 1987; Remington *et al.,* 2007). It is hoped that by providing interventions for self-injury when individuals with ASD are young, that the interventions employed will be more successful as they are less difficult to implement with children who are smaller and easier to manage. Additionally, reinforcement history for self-injurious behaviour will be shorter and, consequently, it could be hypothesised that the behaviours will be less resistant to change (Oliver *et al.,* 2005). In order for early intervention strategies to be effective, it would be beneficial to identify those individuals with greatest risk of developing self-injury. Delineating risk markers which are associated with the presence of self-injurious behaviour in individuals with ASD could aid the early intervention process. The putative risk markers could then be utilised to identify those individuals who are most likely to develop self-injury, and therefore those individuals for whom early intervention would be most warranted.

There is emerging evidence of demographic characteristics that are associated with self-injury in ASD at a single time point. These putative risk markers include impairments in adaptive skills (Baghdadli, Pascal, Grisi & Aussilloux, 2003), higher degree of autism (Baghdadli *et al.,* 2003), younger age (Esbensen, Seltzer, Lam & Bodfish 2009) and perinatal conditions (Baghdadli *et al.,* 2003). The study reported in Chapter 2 extended these findings by investigating specific behavioural characteristics that could serve as putative risk markers. The results revealed that lower levels of expressive language and higher levels of impulsivity, overactivity and repetitive behaviours were associated with the presence of self-injury in ASD (Sections 2.4.3 & 2.4.4). These findings support data collected in individuals with genetic syndromes and intellectual disability of heterogeneous aetiology which found associations between repetitive, impulsive and compulsive behaviours and self-injury (Arron, Oliver, Moss, Berg & Burbidge, 2011; Hyman, Oliver & Hall, 2002; Davies, 2010). However, there are limited data on the capacity of these putative risk markers to predict onset, persistence and remission of self-injury over time. Therefore, a longitudinal study investigating the behavioural and demographic characteristics associated with self-injury in ASD at distal time points is required.

Studies of populations of individuals with intellectual disabilities have identified lower ability (Cooper *et al.,* 2009), lower verbal ability (Danquah *et al.,* 2009), attention deficit hyperactivity disorder (Cooper *et al.,* 2009), visual impairment (Cooper *et al.,* 2009) and the site of self-injury (Danquah *et al.,* 2009*;* Emerson *et al.,* 2001b) as variables which independently predict the persistence of self-injury over time. A recent review of research into the persistence of challenging behaviour noted that there has been relatively little work directly evidencing behavioural correlates of persistence (Totsika & Hastings, 2009). In line with this, only two studies have evaluated the predictive value of variables to identify persistent self-injury in ASD. In the first study, Shattuck *et al.* (2007) grouped self-injury into a subscale with repetitive, withdrawn and inattentive behaviours. Having an intellectual disability and being in an older age cohort significantly predicted the persistence of these 'internalised' behaviours over time. In the second study, Baghdadli *et al.,* (2008) demonstrated that speech deficits and autism severity significantly predicted a negative outcome in self-injury over three years. However, Baghdadli *et al.,* (2008) grouped both onset of self-injury and persistence of self-injury into a 'negative outcome' category. Consequently, it is not possible to isolate whether speech deficits and autism severity both contributed to the persistence of self-injury in ASD from their data. Thus far, no studies have investigated variables predictive of persistent self-injury specifically in isolation from other behaviours. Therefore, in order to identify those individuals for whom early intervention could be targeted, risk markers for the persistence of self-injury in ASD need to be delineated.

In summary, the prevalence of self-injury has been reliably demonstrated to be elevated in those with ASD, compared to those with intellectual disability of heterogeneous aetiology (Baghdadli *et al.,* 2003; Billstedt, Gillberg & Gillberg, 2005; McClintock, Hall & Oliver, 2003; Shattuck *et al.,* 2007). However, there is equivocal evidence for the persistent nature of self-injury, with some studies demonstrating persistence over time (Emerson *et al.,* 2001b) and other studies reporting significant decreases in self-injury over time (Cooper *et al.,* 2009; Shattuck *et al.,* 2007). Prior to a consideration of early intervention and putative risk markers for self-injurious behaviour in ASD, evidence should be gathered regarding the persistence of self-injury in ASD. In order to guard against threats to validity, this evidence must be established in an ASD population that has not been recruited from a clinical sample. There is evidence associating a range of demographic and behavioural characteristics with self-injury in ASD at a single time point (Baghdadli *et al.,* 2003; Esbensen *et al.,* 2009; Section 2.4.3), and predicting persistent self-injury in intellectual disability populations (Cooper *et al.,* 2009; Danquah *et al.,* 2009; Emerson *et al.,* 2001b). However, there is currently very limited evidence in ASD populations, demonstrating characteristics associated with self-injury at multiple time points and demonstrating the ability of these characteristics to predict persistent self-injury (Baghdali *et al.,* 2008; Shattuck *et al.,* 2007). These data are required in order to establish whether it is possible to identify those individuals with ASD for whom early intervention for self-injury may be warranted. Therefore this study will conduct a longitudinal follow up of the sample delineated in Chapter 2. Within this sample, the following research aims will be investigated:

i) The prevalence, topographies and severity of self-injury at one time point (T_1) will be compared to the prevalence, topographies and severity of self-injury at a second time point (T_2) three years later, in order to establish the persistence of self-injury over time.

ii) Behavioural and demographic variables associated with self-injury at T_2 will be investigated. It is predicted that the demographic and behavioural variables associated with self-injury at T_1 (Sections 2.4.3 & 2.4.4; poor speech, impulsivity, overactivity, repetitive behaviours) will also be associated with self-injury at T_2.

iii) The value of these behavioural and demographic variables at T_1 to differentiate between absent, transient and persistent self-injury at T_2 will be evaluated. Additionally, the value of these putative risk markers to predict the persistence of self-injury over three years will be established.

3.3 Methods

3.3.1 Recruitment

Participants with ASD who had taken part at T_1 (reported in Chapter 2) were contacted and invited to participate at T_2. In total, 68 carers of individuals with ASD (return rate 35.98%) completed the questionnaire pack.

3.3.2 Procedure

Three years after completing the questionnaire pack at T_1, carers received an information sheet, cover letter, consent form, demographic questionnaire and questionnaire pack (see Appendix B). To avoid priming, the study was described as investigating behaviours associated with ASD. Carers returned completed questionnaires and consent forms in a prepaid envelope.

Ethical approval for this study was obtained from the School of Psychology ethical review committee at the University of Birmingham.

3.3.3 Participants

Participants were included based upon inclusion criteria at T_1. All participants had a confirmed diagnosis of ASD from a professional at $T_{1.}$ Diagnoses included autism, Asperger syndrome, autism spectrum disorder and pervasive developmental disorder. These professionals included General Practitioner, Clinical Geneticist, Paediatrician, Psychiatrist, Clinical Psychologist and Educational Psychologist. At T_1, Social Communication Questionnaire (SCQ; Berument, Rutter, Lord, Pickles & Bailey, 1999) scores were used to screen the sample to ensure that all individuals in the ASD group scored at or above the cut-off for ASD on the SCQ. Individuals with incomplete SCQ scores, or SCQ scores which did not meet the SCQ cut off for ASD were removed from the analysis at T_1, and consequently were not included at T_2. In order to ensure a large enough follow up sample, both the ASD sample used for between groups analysis (see Section 2.3.3.1) and the 'more able' ASD sample used for within group analysis (see Section 2.3.3.2) were recruited to take part at T_2. For the purposes of this study, the two samples at T_1 are combined and referred to as 'T_1 sample'.

At T_2, if a large proportion of the data (25% or more of items across questionnaires) were incomplete the participant was excluded from the analysis. This resulted in one participant being excluded from the analysis, leaving a final sample of 67 at T_2.

To ensure that the T_2 sample was representative of the T_1 sample, and not biased by the loss of 122 participants, a series of Mann Whitney U and χ^2 analyses were conducted to detect possible significant differences between participants included at T_2 (67) and those from the T_1 sample who were not included. Table 3.1 describes the demographic and behavioural characteristics of those who took part at T_2 and those who declined to take part at T_2

Table 3.1 Median age (inter-quartile range) and range, percentage of males, percentage of participants who were able, mobile and verbal, median (interquartile range) MIPQ, TAQ, RBQ and SCQ scores for those who took part at T_2 and those who declined to take part at T_2. Significant differences are highlighted in bold (*p* <.05; two tailed)

		Took part at T_2	**Declined to take part at T_2**	**Mann Whitney U / χ^2**	**df**	***p* value**
N		67	122			
Age	Median *(IQ Range)*	10.00 *(7.00 – 14.00)*	10.00 *(7.00 – 14.00)*	3861.00	-	.529
Gender	% male	85.1	86.1	0.04	1	.852
Self Help	% partly able/able	89.6	85.2	0.70	1	.403
Mobility	% mobile	98.5	95.1	-*	-	.425
Speech	% verbal	89.6	82.8	1.30	1	.255
Self-injury	% with behaviour	**40.3**	**54.9**	**3.92**	**1**	**.048**
MIPQ Total Score	Median *(IQ Range)*	34.00 *(30.00 – 38.00)*	32.00 *(27.00 – 38.00)*	3766.00	-	.372
TAQ Total Score	Median *(IQ Range)*	43.00 *(24.63 – 53.00)*	43.00 *(30.00 – 56.00)*	3554.00	-	.220
RBQ Total Score	Median *(IQ Range)*	27.00 *(17.00 – 37.00)*	29.50 *(17.10 – 40.00)*	3709.50	-	.381
SCQ Total Score[1]	Median *(IQ Range)*	28.00 *(23.00 – 31.00)*	26.00 *(22.00 – 30.12)*	3722.5	-	.310

* Fishers exact T was calculated as 2 cells had an expected count < 5

The analysis revealed that significantly more individuals without self-injury took part at T_2 than individuals with self-injury. Apart from this, individuals who took part at T_2 did not differ on any other demographic or behavioural variable, to the individuals who declined to

[1] As in Chapter 2, all analysis conducted utilising the SCQ, excludes item 17 ('has she/he ever injured her/himself deliberately, such as biting her/his arm or banging her/his head?') to prevent confounds in self-injury analysis. See Section 2.3.4 for further detail.

take part at T_2. This suggests that the data sample collected at T_2 is broadly representative of the original sample collected at T_1.

3.3.4 Measures

The follow up study used predominantly the same measures as those reported in Chapter 2 to allow for a direct comparison of differences between T_1 and T_2. See Section 2.3.4 for the measures administered.

Whilst at T_1, the Lifetime Version of the Social Communication Questionnaire was employed (SCQ; Berument *et al.*, 1999), at T_2, the Current Version was administered as this version is recommended in order to evaluate measurement of change over time. Whilst this allows for comparisons of change over time between T_2 and all future longitudinal samples, this change does result in comparisons between T_1 and T_2 SCQ data being tentative.

3.3.5 Data analysis

Data were tested for normality using Kolmogorov–Smirnov tests. Where data were not normally distributed ($p<.05$), non-parametric techniques were employed. Where multiple tests were conducted, the alpha level was set to $p < .01$. McNemar tests were conducted in order to examine the persistence and topographies of self-injury. A self-injury severity score was calculated by summing items regarding the length of time self-injury was displayed for, the frequency of self-injury, and the level of intervention required for self-injury. Wilcoxen signed ranks test was used to evaluate differences in this score between T_1 and T_2.

Chi square and Mann Whitney tests were conducted in order to examine the difference between those who engaged in self-injury and those who did not on a variety of demographic and behavioural characteristics. Kruskal Wallis tests were employed to test for differences in T_1 putative risk markers between absent (self-injury absent at both T_1 and T_2), transient (self-injury present at *either* T_1 or T_2) and persistent (self-injury present at both T_1 and T_2) self-injury groups. Chi square and Fishers Exact T tests were used to test for these differences in categorical data. Finally, binary logistic regressions were conducted in order to evaluate the utility of the demographic and behavioural putative risk markers to predict persistent self-injury from absent self-injury.

3.4 Results

3.4.1 Changes in demographic and behavioural characteristics over time

Prior to analysis, the demographic and behavioural characteristics of the sample included at T_2 were compared to the demographic and behavioural characteristics of the same sample at T_1. This was done in order to evaluate whether any changes had occurred in demographic and behavioural characteristics that may interact with the persistence of self-injury. Table 3.2 presents the demographic and behavioural characteristics of the sample included at T_2 and $T_{1.}$ Wilcoxen signed ranks tests and McNemar analyses were conducted to test for differences between the two time points.

Table 3.2 Demographic and behavioural characteristics of the selected sample at T_1 and T_2. Significant differences between the two data collection points are highlighted in bold (p <.01; all tests are two tailed apart from age)

		$\mathbf{T_1}$	$\mathbf{T_2}$	***p* value**
N		67	67	
Age	Median *(IQ Range)*	10.00 *(7.00 – 14.00)*	13.50 *(10.00 – 17.00)*	**<.001**
Self Help	% partly able/able	89.6	88.1	1.00
Mobility	% mobile	95.5	97.0	1.00
Speech	% verbal	95.5	91.0	.50
Vision	% normal	97.0	86.6	.39
Hearing	% normal	98.5	98.5	1.00
MIPQ Total Score	Median *(IQ Range)*	34.00 *(30.00 – 38.00)*	34.00 *(29.00 – 40.00)*	.264
TAQ Total Score	Median *(IQ Range)*	43.00 *(24.63 – 53.00)*	41.00 *(21.00 – 50.00)*	.117
RBQ Total Score	Median *(IQ Range)*	28.00 *(17.00 – 37.00)*	26.00 *(18.00 – 33.00)*	.289
SCQ Total Score	Median *(IQ Range)*	28.00 *(15.00 – 37.00)*	21.36 *(6.00 – 34.00)*	**<.001**
SCQ Total Self-Injury	Median *(IQ Range)*	30.00 *(18.00 – 37.00)*	25.00 *(11.00 – 34.00)*	**<.001**
SCQ Total No Self-Injury	Median *(IQ Range)*	26.00 *(15.00 – 35.00)*	17.00 *(6.00 – 30.00)*	**<.001**

The results presented in Table 3.2 reveal that, as expected, there was a significant difference between age at T_1 and age at T_2. There were no significant differences revealed for any other demographic or behavioural characteristics between T_1 and T_2 when assessed using consistent measures. There was a significant difference between SCQ scores at T_1 and T_2, as measured by the SCQ – Lifetime Version at T_1 and the SCQ – Current Version at T_2. Scores on the Lifetime Version at T_1 indicated significantly higher levels of ASD phenomenology than Current Version scores at T_2. This difference was significant for both the self-injury and non-self-injury group at T_2.

3.4.2 Persistence of self-injury, topographies of self-injury and severity of self-injury

In order to examine the persistence, remission and incidence of self-injury, the percentage of the sample who showed self-injurious behaviour, and the various topographies of self-injury, at neither T_1 nor T_2, at $T_{1,}$ but not T_2, at $T_{2,}$ but not T_1, and at both $T_{1,}$ and T_2, respectively was calculated (see Table 3.3). McNemar analysis was employed to assess the persistence of self-injury.

Table 3.3. Percentage and number of participants (in parentheses) in remission, incidence and persistence and no behaviour groups and analysis examining the persistence of self-injury between T_1 and T_2 (left of the bold line). Remission and persistence of self-injurious behaviour in participants showing the behaviour at T_1 (right of the bold line).

Behaviour	Absent (Absent at T_1, Absent at T_2)	Remission (Present at T_1, Absent at T_2)	Incidence (Absent at T_1, Present at T_2)	Persistent (Present at T_1, Present at T_2	*P* (2 tailed)	Remission in participants with self-injury at T_1	Persistence in participants with self-injury at T_1
Self-injury	49.3 (33)	9.0 (6)	10.4 (7)	31.3 (21)	1.00	22.2 (6)	77.8 (21)
Hits self with body	79.1 (53)	1.5 (1)	10.4 (7)	9.0 (6)	.07	14.3 (1)	85.7 (6)
Hits self against object	73.1 (49)	7.5 (5)	10.4 (7)	9.0 (6)	.77	45.5 (5)	54.5 (6)
Hits self with object	92.5 (62)	1.5 (1)	3.0 (2)	3.0 (2)	1.00	33.3 (1)	66.7 (2)
Bites self	68.7 (46)	10.4 (7)	11.9 (8)	9.0 (6)	1.00	53.8 (7)	46.2 (6)
Pulls self	88.1 (59)	3.0 (2)	4.5 (3)	4.5 (3)	1.00	40.0 (2)	60.0 (3)
Rubs/ scratches self	79.1 (53)	9.0 (6)	10.4 (7)	1.5 (1)	1.00	85.7 (6)	14.3 (1)
Inserts	95.5 (64)	1.5 (1)	1.5 (1)	1.5 (1)	1.00	50.0 (1)	50.0 (1)

The results presented in Table 3.3 reveal that there were no significant differences in the presence or topography of self-injury displayed at T_1 and T_2, indicating that the behaviour is persistent and stable over time.

In order to evaluate the stability of the severity of self-injury, the self-injury severity score at T_1 (median = 6.00, IQR 4.00 – 8.00), and the self-injury severity score at T_2 (median = 5.00, IQR = 4.00 – 7.50) of those with persistent self-injury were compared using Wilcoxon signed ranks test. The results revealed that there was no significant difference between the self-injury severity scores at T_1 and T_2 (N = 21, $p = .374$).

In summary, the results revealed that the presence, topography and severity of self-injury were persistent and stable over time.

3.4.3 Demographic and behavioural characteristics associated with self-injury at T_2

In order to test the second hypothesis of the study, comparisons were made between those who displayed self-injury and those who did not at T_2 on a variety of demographic and behavioural characteristics. Table 3.4 reports the differences between those with self-injury, and those without on demographic measures.

The results reveal that individuals with self-injury were significantly more likely to be non verbal than those who did not engage in self-injury. Additionally, individuals with self-injury were significantly more likely to be 'not able' as evidenced through poorer self-help skills. There were no significant differences between those who engaged in self-injury and those who did not, on any other demographic items.

Table 3.4 Demographic variables for participants with self-injury and without self-injury at T_2. Significant differences (p<.01) are highlighted in bold; variables for which significant differences were obtained at T_1 are underlined (all tests are two tailed apart from level of ability and speech).

		Percentage of individuals with self-injury *(N)*	Percentage of individuals without self-injury *(N)*	Chi-square	*P* value
Gender	Male	82.1 (23)	89.7 (35)	-**	.474
	Female	17.9 (5)	10.3 (4)		
Age	≤11 years	39.3 (11)	38.5* (15)	.590	.745
	12-18 years	46.4 (13)	38.5 (15)		
	≥ 19 years	14.3 (4)	20.5 (8)		
Ability	Able/ Partly Able	75.0* (21)	97.4 (38)	-**	**.008**
	Not Able	21.4 (6)	2.6 (1)		
Speech	Verbal/ Partly verbal	78.6* (22)	100.0 (39)	<u>**-****</u>	<u>**.005**</u>
	Non-Verbal	17.9 (5)	0.0 (0)		
Mobility	Mobile	92.6* (26)	100.0 (39)	-**	.409
	Non-mobile/ Partly mobile	3.6 (1)	0.0 (0)		
Vision	Normal	89.3 (25)	84.6 (33)	-**	.724
	Poor Vision/ Blind	10.7 (3)	15.4 (6)		
Hearing	Normal	96.4 (27)	100.0 (39)	-**	.418
	Poor Hearing/ Deaf	3.6 (1)	0.0 (0)		

* One case of missing data
**Fishers exact T was calculated

Table 3.5 reports the differences between those with self-injury and those without, on measures of behavioural characteristics.

Table 3.5 Median scores and Mann-Whitney statistics for measures of affect, repetitive behaviour, activity level and autism phenomenology for ASD participants with and without self-injury at T_2. Bold text indicates a significant difference (p<.01, one tailed), variables for which significant differences were obtained at T_1 are underlined

Measure Subscale	Median scores *(interquartile range)* With self-injury (N = 28)	Without self-injury (N= 39)	U score	*P* value	Cliff's *d*
MIPQ-S					
Mood	18.91 *(15.25 – 21.00)*	21.00 *(18.00 – 23.00)*	401.00	.032	-
Interest and Pleasure	13.50 *(10.00 – 16.75)*	17.00 *(12.00 – 19.00)*	420.00	.054	-
RBQ					
Stereotyped behaviour	8.00 *(6.00 – 11.75)*	5.00 *(2.00 – 9.00)*	720.50	.013	-
Compulsive behaviour	8.50 *(4.50 – 12.75)*	4.00 *(1.00 – 8.00)*	**748.50**	**.005**	.37
Insistence on sameness	4.00 *(2.25 – 8.00)*	3.00 *(0.00 – 6.00)*	680.00	.043	-
Restricted preferences*	6.00 *(2.00 – 8.00)*	5.00 *(2.00 – 7.00)*	426.50	.331	-
Repetitive language*	7.00 *(3.50 – 9.00)*	4.00 *(4.00 – 8.00)*	496.50	.060	-
TAQ					
Overactivity	22.43 *(13.00 – 30.75)*	14.00 *(6.00 – 21.00)*	**759.00**	**.004**	.39
Impulsivity	20.50 *(16.00 – 22.00)*	16.00 *(9.00 – 19.00)*	**745.00**	**.006**	.36
Impulsive Speech*	3.00 *(1.00 – 8.50)*	5.00 *(2.00 – 6.00)*	383.50	.401	-
SCQ					
Communication	8.00 *(6.00 – 9.75)*	7.00 *(6.00 – 9.00)*	631.00	.097	-
Social Interaction	7.00 *(5.00 – 7.00)*	5.00 *(3.00 – 6.00)*	**814.50**	**.001**	.49
Repetitive Behaviour	9.50 *(7.00 – 12.00)*	5.00 *(3.00 – 9.00)*	**825.00**	**<.001**	.57

* Subscales only calculated for verbal participants

The results in Table 3.5 reveal that at T_2 individuals with self-injury evidenced significantly higher scores for measures of compulsive behaviour, overactivity and impulsivity, with moderate effect sizes obtained for all three behavioural characteristics. Additionally, individuals with self-injury evidenced significantly higher scores for measures of ASD phenomenology, specifically impairments in social interaction and repetitive behaviour. There were no significant differences identified for any other behavioural characteristics.

In summary, at T_2 individuals with self-injury were significantly more likely to be less able and non-verbal, and to show higher levels of compulsive behaviour, overactivity, impulsivity, repetitive behaviour and impairments in social interaction.

3.4.4 Comparison of persistent, transient and absent self-injury groups on T_1 behavioural and demographic variables

In order to evaluated the third aim of the study, participants at T_2 were categorised into three self-injury groups; absent (self-injury absent at both T_1 and T_2; N = 33), transient (self-injury present at *either* T_1 or T_2; N = 13) and persistent (self-injury present at both T_1 and T_2; N = 21). The small sample size in the transient group prevented an analysis of behavioural and demographic variables associated with the onset or remission of self-injury. In order to identify putative risk markers, comparisons were made between T_1 data for these three groups on any variables for which differences had been obtained between the self-injury and non self-injury samples at *either* T_1 or T_2.

Fisher's exact T tests revealed that there were no significant differences between the three groups at T_1 for speech ($p = .059$) or levels of ability ($p = .171$). Figure 3.1 displays the median, maximum and minimum scores and significant differences between groups on measures of behavioural characteristics.

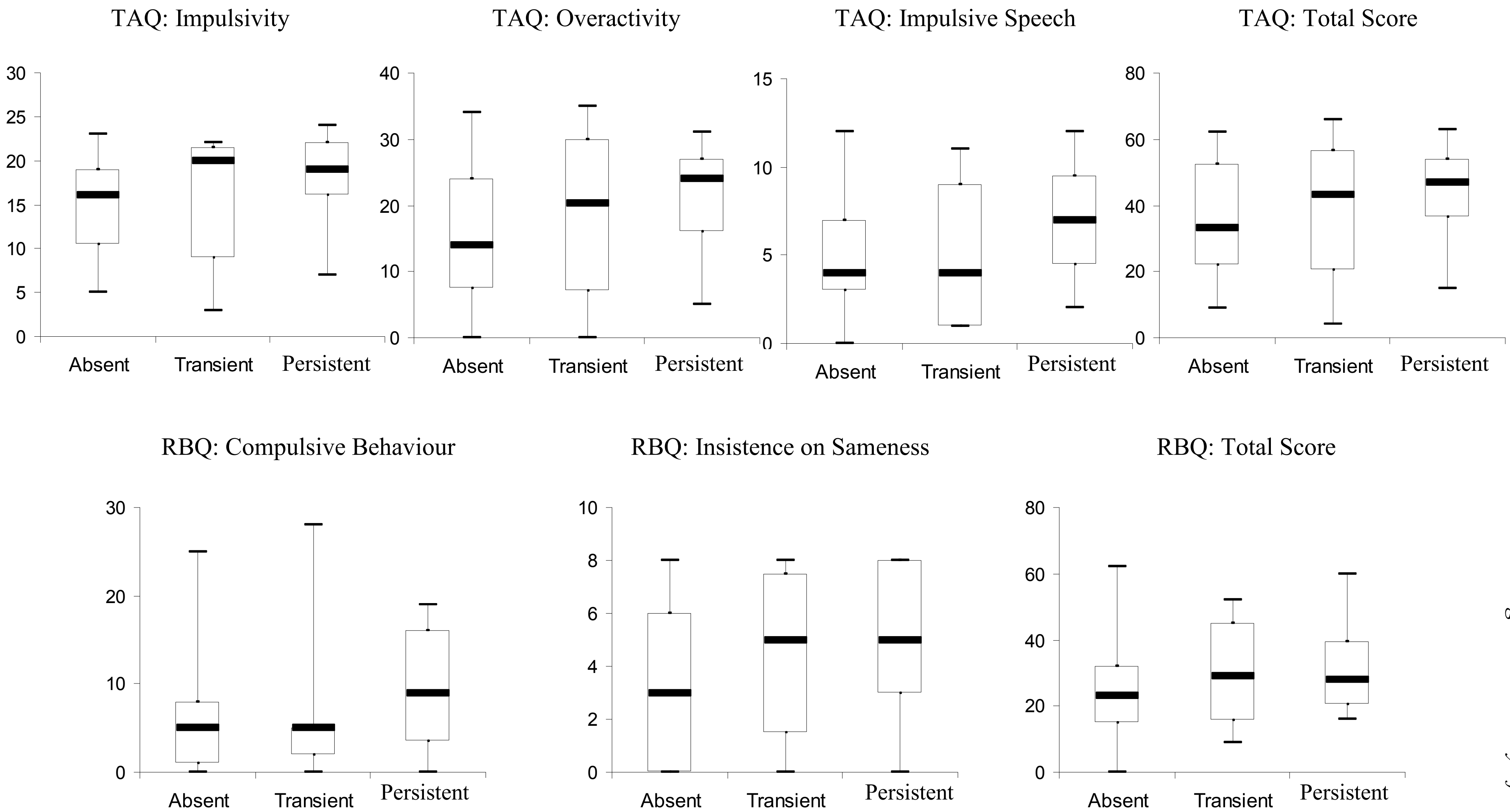


Figure 3.1 Median, maximum, minimum and inter-quartile range of TAQ, RBQ and SCQ subscale and total scores for absent, transient and persistent self-injury groups.

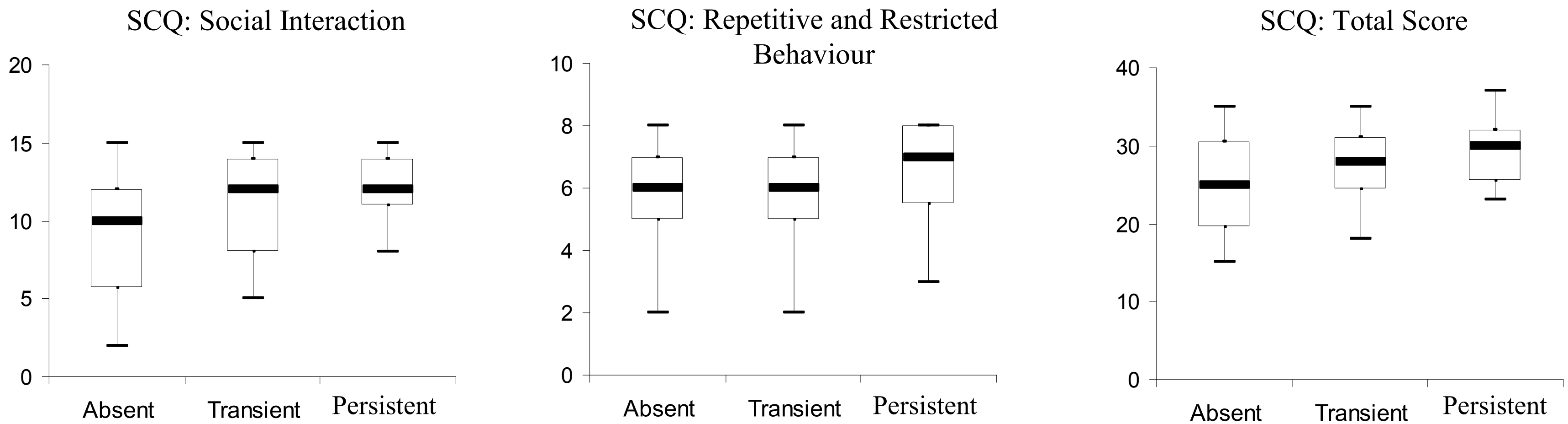


Figure 3.1 cont. Median, maximum, minimum and inter-quartile range of TAQ, RBQ and SCQ subscale and total scores for absent, transient and persistent self-injury groups.

The results reveal that across the majority of subscale and total scores, there was a broad trend towards the absent group having the lowest scores and the persistent group obtaining the highest scores, with the transient group falling between these two points. Whilst the differences between the groups approached significance for the SCQ subscale: Social Interaction (Kruskal Wallis χ^2 (2) = 7.49, p = .012) and the SCQ total score (Kruskal Wallis χ^2 (2) = 6.74, p = .017), no significant differences were identified between the groups on any measures.

3.4.5 Predictive value of T_1 demographic and behavioural characteristics

In order to evaluate the final aim of the study, to establish the predictive validity of the putative risk markers, a binary logistic regression was conducted. Due to the small sample of the ‘transient self-injury’ group (N = 13), and the heterogeneity of the self-injury in this group, the transient sample were not included in the analysis. Therefore, the binary logistic regression was conducted in order to evaluate the predictive value of the risk markers to predict persistent self-injury over and above absent self-injury. In order to establish robust risk markers, only subscale variables for which significant differences between those who engaged in self-injury at T_1 *and* T_2 were considered for this analysis.

Overactivity and impulsivity scores were found to be highly correlated (r_s = .69, p <.001), as were overactivity and compulsive behaviour scores (r_s = .35, p = .003) and impulsivity and compulsive behaviour scores (r_s = .38, p = .002). Therefore, in order to protect against multi-colinearity a composite score was created from T_1 scores (Comp/Ov/Imp) and entered into the logistic regression.

Binary logistic regression was performed to evaluate the effect of overactivity, impulsivity, compulsivity and the presence of speech on the likelihood that participants displayed persistent self-injury. The full model containing both predictors was statistically significant, (χ^2 (2, N = 54) = 10.51, p = .005), indicating that the model was able to distinguish between participants displaying persistent self-injury and participants not displaying self-injury. The model as a whole explained between 17.7% (Cox and Snell R square) and 24.0% (Nagelkerke R squared) of the variance in self-injury status, and correctly classified 63.0% of cases. As shown in Table 3.6, only one of the independent variables made a unique statistically significant contribution to the model (Comp/Ov/Imp composite). The odds ratio for the Compulsive/Overactivity/Impulsivity composite was 1.04, suggesting that individuals with high levels of compulsivity, overactivity and impulsivity were 1.04 times more likely to show self-injurious behaviour, controlling for other factors in the model.

Table 3.6 Logistic regression predicting likelihood of displaying persistent self-injury,from self-injury being absent (bold text indicates predictor variables where p <.05)

	B	**S.E**	**Wald**	***Df***	***p***	**Odds Ratio**	**95.0% C.I. for Odds Ratio**	
							Lower	Upper
Comp/Ov/Imp	0.04	.02	4.12	1	**.042**	**1.04**	**1.00**	**1.08**
Speech	-21.38	22937.34	0.00	1	.999	0.00	0.00	-

In summary, the logistic regression revealed that, independently of other factors, high levels of compulsive behaviour, overactivity and impulsivity significantly increase the likelihood of an individual displaying persistent self-injury.

3.5 Discussion

The persistence of the presence, topography and severity of self-injury in individuals with ASD was evaluated in this study. Additionally, behavioural and demographic variables that predicted the persistence of self-injury in ASD were delineated. Importantly, the recruitment of a demographically representative sample at T_2, and the utilisation of standardised measures, strengthens the validity of the study. The use of an ASD screen at T_1, increases the external validity of the study and ensures robust results were obtained from the sample. This was the first longitudinal study to assess behavioural differences between groups with absent, transient and persistent self-injury. Additionally, this was the first study to ascertain the characteristics that predicted the persistence of self-injury over and above the absence of self-injury in an ASD population, and therefore the findings have significant clinical utility. The results of the study revealed self-injury to be persistent in presence, topography and severity in individuals with ASD. Importantly the demographic and behavioural variables associated with the presence of self-injury at T_1 were revealed to be associated with self-injury at T_2. Broad trends were identified in these variables between the absent, transient and persistent self-injury groups. Finally, compulsive behaviour, impulsivity and overactivity at T_1 were demonstrated to predict persistent from absent self-injury at T_2. The results as a whole provide evidence to investigate early intervention for self-injury and begin to highlight those individuals with ASD for whom early intervention may be warranted.

The results of this study revealed that self-injury was persistent over three years in 77.8% of those who showed self-injury at T_1. This finding supports data collected in populations with intellectual disability, where the persistence of self-injury has been reported at between 71% and 84% (Emerson *et al.,* 2001b, Taylor *et al.,* 2011). The results in this study significantly differ from those reported in ASD populations where self-injurious behaviour was found to significantly decrease over time (Baghdadli *et al.,* 2008; Shattuck *et al.,* 2007). This difference is likely due to the fact that the ASD samples assessed in previous research were recruited from clinical services, and were therefore more likely to receive interventions to reduce self-injury. The results from this study indicate that self-injury in ASD is persistent and stable over time, suggesting that intervention with smaller, younger children, where self-injury has a shorter reinforcement history, may be beneficial as the behaviour is unlikely to stop or decrease with time. The results extended previous research in ASD populations by demonstrating that the topographies of self-injury were also persistent across time. Interestingly, the severity of self-injury was found to be stable across time, indicating that although self-injurious behaviour did not improve, it also did not increase in frequency, length or the level of intervention required. These results however, must be interpreted with caution. Although this sample was not drawn from a clinical population, the sample was recruited from a parent support group. It is therefore plausible that the families included may have been receiving greater levels of behavioural support and advice than families not enrolled in a support group. Additionally, these families may differ in socioeconomic status and therefore the stability in severity of self-injury may not be representative of the broader ASD population. Further research is required to investigate this, as changes in severity of self-injury over time would highlight the time points at which early intervention may be most effective and warranted.

The results also revealed that the majority of variables that were associated with the presence of self-injury at T_1 were also associated with self-injury at T_2 (Section 3.4.3). Being non-verbal, and having high levels of compulsive behaviour, overactivity and impulsivity were associated with the presence of self-injury. Importantly, there were no significant changes in the total sample over time in any of these variables. The stability of these variables and their consistent association with self-injury is important for both pragmatic and theoretical reasons. First, the results support the validity of delineating variables that help to separate those individuals with ASD for whom self-injury is likely to be persistent from those with absent self-injury. The preliminary results in this study indicate that there are stable variables, associated with self-injury over time which might be used as risk markers for persistent self-injury. Secondly, the consistent association between self-injury and ability (as evidenced through adaptive skills and speech) and self-injury and, arguably, behavioural inhibition (as evidenced through compulsive behaviour, overactivity and impulsivity) reveals further information about the nature of self-injurious behaviour, and identifies potential areas for future research. As discussed in Chapter 2 (Section 2.5), the association between impulsivity, overactivity, compulsive behaviour and self-injury lend strength to the suggestion that impaired behavioural regulation may contribute to the presence of self-injury in individuals with ASD, as has been highlighted in data from other syndrome groups (Arron *et al.,* 2011; Hyman *et al.,* 2002; Davies, 2010). Additionally, the association between self-injury and impulsivity and overactivity at T_2, supports the pragmatic decision taken at T_1 to be less conservative in the interpretation of weak or moderate statistical results which may have significant clinical implications. Therefore, there is a pressing need to investigate further the potential relationship between self-injury and behaviours indicative of compromised inhibition.

In addition to demographic and behavioural variables which were associated with self-injury at both T_1 and T_2, there were also variables which were associated with self-injury at only one time point. At T_2, but not T_1, ability level was associated with self-injury, however, at T_1 this association was approaching significance (Section 2.4.3). The emergence of this as a significant difference at T_2 may be reflective of an association between self-injury and lower ability which varies over time. Perhaps as chronological age increases, the impact of lower ability becomes more significant and therefore an association with self-injury emerges. Alternatively, parental estimation of their child's ability level may become more accurate over time as more chronological age comparisons are made with peers. This finding warrants further investigation in longitudinal samples with a broader spread of ages and ability levels.

Similarly, at T_2 social interaction and repetitive behaviour as measured by the current form of the SCQ were associated with self-injury, whereas at T_1 no subscales of the lifetime form of the SCQ evidenced an association with self-injurious behaviour. Tentative conclusions must be drawn about this result, given the slight change in measure used. Additionally, there was a significant decrease in total SCQ score between T_1 and T_2. Significant changes in ASD behaviour over time have also been robustly reported in other ASD populations, (Moss, Magiati, Charmin & Howlin, 2008), therefore it is possible the significant decrease in SCQ score reported in this study represents a true change in ASD phenomenology and an emergent association with self-injury. However, it is also possible that the change in the focus of the measure, to current ASD presentation rather than developmental ASD phenomenology, has revealed an association between ASD and self-injury. This preliminary result supports those reported in the literature, associating ASD behaviours and self-injury (Arron *et al.,* 2011; Baghdadli *et al.,* 2003; 2008) and may indicate that current ASD presentation is a stronger predictor of current self-injury than developmental ASD phenomenology or diagnosis. Again, further longitudinal data are required in order to investigate this hypothesis. Finally, although both impulsive speech and insistence on sameness were associated with self-injury at T_1, neither characteristic was associated with self-injury at T_2. Currently our understanding of variables associated with self-injurious behaviour broadly, and in ASD more specifically, is very limited (Totsika & Hastings, 2009). The differences in results between T_1 and T_2 warrant further investigation in order to delineate whether these are artefacts of a small data sample, or real differences indicative of variables that can and can not be utilised as longitudinal risk markers for self-injury.

The results of this study also revealed broad trends of difference in behavioural characteristics at T_1 between the absent, transient and persistent self-injury groups at T_2. For the majority of behavioural indicators, scores obtained at T_1 indicated higher and more severe levels of behaviour in the persistent group than for the transient and absent groups. However, although the results from the SCQ subscale and total score approached significance, no statistical differences were obtained on any measures. This is likely to be due to the small sample size at T_2, particularly in the transient self-injury group. Ideally, future research should replicate these analyses in larger samples with greater statistical power. Additionally, with a large enough sample, the transient self-injury group should be further subdivided to allow an analysis of remission and incidence subgroups. A longitudinal study of the behavioural differences between these four subgroups (absent, remission, incidence and persistence) would greatly add to a theoretical and clinical understanding of risk markers for self-injury in ASD populations.

Finally, the results revealed that it was possible to identify variables at T_1 which predicted the persistence of self-injurious behaviour over absent self-injury at T_2. Interestingly, the variables which predicted persistence, those of compulsive, overactive and impulsive behaviours, were again those which are associated with impairments in behavioural inhibition. Importantly, this analysis also controlled for the effect of ability evidenced through speech levels. The finding lends further support to the role of impaired inhibition in the presence and persistence of self-injury. Collectively, these findings represent a significant step forward, towards the possibility of early identification of those who will engage in self-injury. The study has demonstrated that self-injury is worryingly stable in both presence and severity, but that it is possible to identify variables which are consistently associated with the presence of self-injury, and are also able to predict those individuals with ASD who will have persistent self-injury. Further research is now required, in younger and larger samples, to identify whether these behavioural risk markers predict the onset and severity of self-injurious behaviour. If they do, then the plausibility of identifying those with ASD most at risk of developing self-injury, and consequently the evidence base from which to develop an early intervention strategy, are strengthened.

The study is limited by the relatively small sample size recruited at T_2. The small sample prevented investigation of variables associated with, and predictive of, incidence and remission of self-injury. Additionally, the small sample size limits the validity of the regression analysis conducted to identify variables predictive of persistent self-injury. However, the validity of the results is strengthened by the utilisation of an ASD screening measure at T_1 to ensure a homogenous sample. An additional limitation of the study is the under-representation of individuals with self-injury at the T_2 data collection. Whilst no other behavioural or demographic variables differed between the two samples, this does slightly limit the external validity of the findings. However, at both time points the identified prevalence of self-injurious behaviour was in line with other robust estimates in the literature, suggesting that the sample is still representative of the wider ASD population (e.g., Baghdadli *et al.,* 2003; Billstedt *et al.,* 2005; Murphy, Healy & Leader, 2009).

In summary, the results have revealed that self-injury is a persistent and stable behaviour in individuals with ASD. The study has also demonstrated that compulsive behaviour, overactivity and impulsivity remain stable over time, and are consistently associated with the presence of self-injury. Importantly, the results revealed that these behavioural risk markers predict the persistence of self-injury in individuals with ASD. These findings support the necessity and plausibility of an early intervention strategy for self-injury in ASD.

CHAPTER 4

Self-Injurious Behaviour and Self-Restraint in Autism Spectrum Disorder

4.1 Preface

The studies presented in Chapters 2 and 3 described the results of a prevalence study and an assessment of the persistence of self-injury in a sample with ASD. The results indicated that self-injury was persistent in 77.8% of individuals over three years. The study also demonstrated that overactive, impulsive and compulsive characteristics were associated with, and predictive of, persistent self-injury. The current study aims to support and extend these findings by replicating the identification of risk markers in a different and larger sample of children and adults with ASD. In addition to investigating the associated behavioural characteristics highlighted in Chapters 2 and 3, this study also aims to investigate the phenomenology of self-restraint in relation to self-injury. Through the delineation of the associations between behavioural characteristics, self-injury and self-restraint, a theoretical model will be constructed in order to guide research in self-injury in ASD populations.

4.2 Introduction

The prevalence of self-injury in individuals with ASD has been robustly estimated at between 40% and 50% (See Sections 1.4.1, 2.4.1 & 3.4.2), which is significantly higher than prevalence estimates in individuals with intellectual disability of heterogeneous aetiology (see Section 1.3.2). However, despite the elevated prevalence of self-injury in ASD and the identification of ASD as a risk maker for self-injury (Arron, Oliver, Moss, Berg & Burbidge, 2011; Collacott, Cooper, Branford & McGrother, 1998; McClintock, Hall & Oliver, 2003), there has been limited research delineating characteristics and behaviours associated with self-injury in this population (Totsika & Hastings, 2009).

Cross sectional research has identified putative risk markers for self-injury in ASD of impairments in adaptive skills (Baghdadli, Pascal, Grisi & Aussilloux, 2003), higher degree of autism (Baghdadli *et al.,* 2003), younger age (Esbensen, Seltzer, Lam & Bodfish 2009) and perinatal conditions (Baghdadli *et al.,* 2003). Additionally, longitudinal studies have identified having an intellectual disability and being in an older age cohort as predictive of 'internalised behaviours' (Shattuck *et al.,* 2007). In a similar longitudinal study, Baghdadli *et al.,* (2008) reported speech deficits and autism severity as being predictive of a longitudinal negative outcome in self-injury. These preliminary results are useful in delineating characteristics that may identify those individuals with ASD for whom an early intervention approach for self-injury may be warranted (Sections 3.2 & 3.5). However, the majority of published research identifying correlates of self-injury in ASD has focused upon demographic rather than behavioural characteristics. This is despite the evidence that behavioural variables such as overactivity, impulsivity and repetitive behaviour have been identified as risk markers for self-injury in other populations with intellectual disability (Arron *et al.,* 2011; Cooper *et* *al.,* 2009; Hyman, Oliver & Hall, 2002; Davies, 2010). The results of the study presented in Chapters 3 extended the existing research in ASD populations by utilising a longitudinal design to investigate both behavioural and demographic characteristics associated with self-injury. This study and the prevalence study described in Chapter 2 revealed that self-injury at one time point was associated with being non-verbal and having higher levels of impulsivity, overactivity and repetitive behaviour (Sections 2.4.3 & 2.4.4). The longitudinal results demonstrated that high levels of compulsive, overactive and impulsive behaviour predicted persistent self-injury over absent self-injury (Section 3.4.5). Whilst these results are promising, they are preliminary and were identified in relatively small sample sizes. Therefore, it is critical to replicate these initial findings using another ASD population, in order to assess the reliability of these behavioural correlates and the potential for generalisation to clinical samples.

In addition to identifying samples for whom early intervention for self-injury may be warranted, identifying correlates of self-injury also affords the opportunity to test hypotheses regarding the potential causes of self-injury. For example, the co-occurrence of health problems, such as otitis media, gastro-oesophageal reflux or constipation, with self-injury has resulted in a tentative causal association between pain and self-injury (Berg, Arron, Burbidge, Moss & Oliver, 2007; Carr & Owen-DeSchryver, 2007; Carr, Smith, Giacin, Whelan & Pancari, 2003; Christensen *et al*., 2009; Luzzani, Macchini, Valade, Milani & Selicorni, 2003; O' Reilly, 1997). Existing pain is conceptualised in the literature as a setting event for challenging behaviour (Carr *et al.,* 2003; Carr & Blakeley-Smith, 2006; Carr & Smith, 1995), and chronic pain has been hypothesised to lead to self-injury as an attempt to 'gate' the painful experience (Melzack & Wall, 1965; Section 1.3.3.3). Whilst the association of pain and self-injury has face validity, there is currently no evidence associating pain or painful health conditions with self-injury in a cohort of individuals with ASD (de Winter, Jansen & Evenhuis, 2011). Without this initial evidence, it is not possible to evaluate the plausibility of a causal role for pain and painful conditions in the development and maintenance of self-injurious behaviour in ASD.

In addition to evaluating a model of pain and self-injury, the associations between self-injury and impulsive and overactive behaviours identified in Chapters 2 and 3 warrant further investigation. Attention deficit hyperactivity disorder (ADHD), and ADHD type behaviours such as overactivity and impulsivity are evidenced to be independently associated with self-injury in other populations with intellectual disability (Arron *et al.,* 2011; Cooper *et al.,* 2009; Collacott *et al.,* 1998; Oliver, Sloneem, Hall & Oliver, 2009; Schneider, Bijam-Schulte, Janssen, & Stolk, 1996). Importantly, ADHD is thought to be underpinned by a delayed development of inhibition, which, amongst other deficits, comprises both an inability to prevent the initiation of a prepotent response to a stimulus, and the inability to terminate an ongoing response (Barkley, 1997). Similarly, repetitive behaviours have been conceptualised as a deficit in executive function and response inhibition which has an impact on the individuals' ability to generate and control behaviour (Sayers, Oliver, Ruddick & Wallis, 2011; Turner, 1999). Thus, the identified associations between self-injury, repetitive behaviour and impulsivity\overactivity suggest a hypothetical model in which the co-occurrence of overactivity, impulsivity and repetitive behaviour indicates a fundamental deficit in behavioural control. Drawing upon Barkley's theory of ADHD (1997), this model would predict more severe self-injury that is either unrelated to environmental triggers, initiated at a low threshold in the presence of discriminative stimuli, or difficult for the individual to terminate.

Key behaviours which might indicate that self-injury is difficult to control are self-restraint and the preference for imposed restraint. Self-restraint behaviours 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 has been found to be 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 has also been associated with the presence of self-injury (Fovel *et al.,* 1989; Hyman *et al.,* 2002). The prevalence of self-restraint in those who engage in self-injury has been estimated at between 46 and 76 % (Oliver *et al.,* 2003, Powell, Bodfish, Parker, Crawford & Lewis, 1996). Self-injury significantly decreases when self-restraint occurs (Forman, Hall & Oliver, 2002; Marzullo, Progar, Morales, 2009; Rojahn, Mulick, McCoy & Schroeder, 1978; Smith, Iwata, Vollmer & Pace, 1992) and consequently, self-restraint is conceptualised as a behaviour exhibited by an individual in order to inhibit their self-injury.

A recent single case study demonstrated that a heart rate measure of anxiety is elevated when restraint is unavailable (Jennett *et al.,* 2011), suggesting that the drive to engage in self-restraint has physiological correlates. King (1993) argues that the associations between self-injury and self-restraint provide evidence of a 'compulsive' nature of self-injurious behaviour; that attempts to refrain from self-injury by a sub-group of self-injurers suggests that for these individuals, the self-injurious behaviour has no adaptive function. In other words, the self-injurious behaviour is not under the individual's control, leading once again to a hypothesis of impaired behavioural inhibition.

Evidence for an association between self-injury, self-restraint and 'compulsive' behaviour has been found in individuals with Cornelia de Lange Syndrome (CdLS; Hyman *et al.,* 2002). CdLS is a rare genetic syndrome associated with autism spectrum behaviours and severe to profound levels of intellectual disability (Jackson, Kline, Barr & Koch*,* 1993; Moss *et al.,* 2008; Oliver, Arron, Sloneem & Hall, 2008). Hyman *et al.,* (2002) found that those individuals with CdLS who engaged in both self-injury and self-restraint displayed significantly more compulsive behaviours than those who did not display self-injury or self-restraint. In support of this, they also demonstrated that individuals with no compulsive behaviour had no significant association between self-restraint and self-injury. This suggests that it is the presence of 'compulsive' behaviours which moderates the relationship between self-restraint and self-injury. Similarly, in a sample of individuals with intellectual disability who all displayed self-injury, Powell *et al.,* (1996) demonstrated that 'compulsive' behaviour occurred significantly more frequently in those with self-restraint *and* self-injury than in those with self-injury, but no self-restraint.

In these preliminary studies and the model proposed by King (1993), 'compulsive' behaviour was not defined in terms of the classical compulsions seen in clinical populations with Obsessive Compulsive Disorder where the compulsions function to relieve anxiety or prevent a feared consequence (Burns, Keortge, Formea & Sternberger, 1996; Grant & Potenza, 2006; Mcelroy, Phillips, Keck, 1994; Salkovskis, 1999). Rather, 'compulsive' behaviour was used as a term to describe behaviours such as lining up, ritualistic behaviour and spotless behaviours or behaviours which could not be inhibited by the individual with no clear cognitive component. These therefore may be better conceptualised as 'impulsive like' repetitive behaviours. Thus, the associations between self-injury, self-restraint and 'compulsive' behaviour further support a model of compromised inhibition in those who engage in self-injury. However, thus far, there has been no investigation of the prevalence and topographies of self-restraint, or its interaction with self-injury in individuals with ASD. In addition, there have been no investigations of the relationship between self-injury, self-restraint and variables indicative of impaired behavioural control (e.g., overactivity, impulsivity) which controlled for age or lower ability level.

In summary, there is a need to replicate associations identified in Chapters 2 and 3 between behavioural and demographic behaviours and self-injury. Additionally, there is a need to investigate the association between self-injury and self-restraint in individuals with ASD. In order to investigate these factors, a sample of children and adults with ASD will be assessed. A large sample with a broad age range will allow for replication of the risk markers for self-injury identified in Chapters 2 and 3, and will also provide opportunities to model build the behaviours of self-injury, self-restraint, painful health conditions, repetitive behaviour and overactivity and impulsivity. Through analysing the data, a number of areas will be investigated:

i) The prevalence, topographies and severity of self-injury and the prevalence and topographies of self-restraint will be described within this population, comparing results from child and adult subsamples.

ii) It is hypothesised that greater degree of intellectual disability, the presence of health problems, repetitive behaviour, overactivity and impulsivity will be associated with the presence of self-injury.

iii) The validity of the variables identified to *predict* the presence and severity of self-injurious behaviour will be replicated. Based upon the findings in Chapters 2 and 3, it is predicted that presence of overactivity and impulsivity will predict self-injury.

iv) Variables associated with the presence of self-restraint will be delineated. Given the suggested model presented above, it is hypothesised that self-injurious behaviour and behaviours indicative of impaired impulse control will be associated with the presence of self-restraint.

v) The validity of these variables to *predict* the presence of self-restraint will also be assessed. Again, it is predicted that self-injury and behaviours indicative of impaired impulse control will predict the presence of self-restraint.

4.3 Methods

4.3.1 Recruitment

All National Autistic Society (NAS) schools and adult services were contacted and invited to participate using an opt-out consent procedure. Opt-out consent was selected in order to obtain a larger and more representative sample (for a more extended rationale on opt-out consent and a discussion of the ethical issues, see Appendix C). This study was part of an audit of service need for provision for self-injury within the 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.

4.3.2 Procedure

All carers of individuals in the NAS adult services and schools received an information sheet detailing the study (see Appendix D). The information sheet explained the opt-out procedure and gave parents and carers three weeks to contact the school, service or lead researcher if they did not wish data to be collected about the individual they cared for. Following this, the questionnaire packs were distributed to the schools and services (see Appendix E for questionnaire pack) with a letter detailing any children or adults to be excluded from the data collection. Services and schools were instructed to complete a questionnaire pack for all other adults and children. Questionnaire packs were completed by teachers or keyworkers who knew the individual well. To avoid priming, the study was described as research into the behaviour 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.

4.3.3 Participants

Data were collected on 515 individuals with ASD attending 12 NAS adult services and 6 NAS schools. The return rate of the questionnaires was estimated at 60%[1]. Questionnaires with 25% or more of the total items incomplete were excluded from the study (N = 32). Two further individuals were excluded from the study, as the individuals were under the age of six. For these individuals an alternative measure of ability had been used, and comparison to the total sample was therefore not possible. Finally, an additional 57 participants were excluded from analysis, due to missing age data. This left a total of 424 individuals (82.3% of original sample; 208 < 18 years, 216 ≥ 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%) individuals 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%). As in Chapter 3, the Wessex self-help score was used to estimate ability, and was used to form a lower ability group - those with some or substantial impairments in self-help skills (Wessex score = 3 - 8) and a higher ability group - those without impairments in self-help skills (Wessex 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.

4.3.4 Measures

The questionnaire pack comprised items regarding demographic information, the Challenging Behaviour Screening Questionnaire (CBSQ; Davies, 2010), the Self-Restraint Checklist

[1] Services were contacted and asked how many questionnaires they required to allow completion of one per service user. It is plausible that some services asked for more or less questionnaires than service users, however, return rate is calculated based upon this figure as an estimate of number of service users in each service.

(Powell *et al.*, 1996) and the Challenging Behaviour Questionnaire (CBQ; Hyman *et al.,* 2002).

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

The CBSQ (Davies, 2010) was developed as a screening measure to assess putative risk markers for challenging behaviour. 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 questionnaires from which items were drawn included The Wessex Behaviour Scale (Kushlick, Blunden & Cox, 1973), Health Questionnaire (Hall, Arron, Sloneem & Oliver, 2008), Self-Help and Behaviour Rating Scale (Petty, 2006), Activity Questionnaire (Burbidge *et al.,* 2010), Challenging Behaviour Questionnaire (CBQ; Hyman *et al.,* 2002) and the Challenging Behaviour Interview (Part II) (CBI; Oliver *et al.,* 2003). Consequently, the CBSQ provides a measure of ability, health problems, activity levels, repetitive behaviours levels and challenging behaviour levels. Inter-rater reliability for the CBSQ is fair to good with Spearman's Rho correlations ranging from .06 to .81[2], with strong concurrent and convergent validity demonstrated (Davies, 2010).

[2] The correlation coefficient for overactivity/impulsivity was low at .06. However, the construct of overactivity and impulsivity is often reported with low levels of inter-rater reliability (Amador-Campos, Forns-Santacana, Guardia-Olmos & Pero-Cebollero, 2006; Charach, Chen, Hogg-Johnson & Schachar, 2009; Papageorgiou, Kalyva, Dafoulis & Vostanis, 2008).

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 behaviours. The scale has good inter-rater reliability of 91% (Powell *et al.,* 1996).

The CBQ (Hyman *et al.,* 2002) evaluates the presence of self-injury, physical aggression, destruction of property and stereotyped behaviour over the last month. The measure also examines eight topographies of self-injurious behaviour that were adapted from Bodfish *et al.* (1995). Items evaluating self-injury 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).

4.3.5 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 self-injury and self-restraint, the percentage of the sample showing each behaviour was derived from the CBSQ, CBQ and Self-Restraint Checklist. Severity of self-injury was derived from three items from the CBSQ. These items rate the frequency, management difficulties and concern caused by the self-injurious behaviour 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 self-injury correlated very strongly with the scores for frequency of self-injury ($r_s = .92$, $p <.001$), as did the scores for management difficulties ($r_s = .92$, $p <.001$). Therefore, as frequency of self-injury was a more easily defined and concrete 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 self-injurious behaviour.

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

In order to investigate variables associated with the presence and severity of self-injury, a number of categorical groups were created for key variables. For health problems, the sample was categorised into those displaying no health problems, and those displaying one or more health problems. For ability, a median split was conducted on the Wessex self-help score forming two groups; those with lower ability (score < 9 on Wessex self-help scale) and those with higher ability (score $= 9$ on Wessex self-help scale). A repetitive and restricted behaviours and interests (Repetitive/Restricted) composite was formed by summing two items in the CBSQ 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 behaviours and interest (score < 4 on Repetitive/Restricted composite) and those with high levels of repetitive and restricted behaviours and interest (score ≥ 4 on Repetitive/Restricted composite). An overactivity and impulsivity (Overactive/Impulsive) composite was formed by summing four items in the CBSQ referring to overactive and impulsive behaviours. 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 behaviours and interested (score < 5 on Overactive/Impulsive composite) and those with high levels of repetitive and restricted behaviours and interests (score $\geq$ 5 on Overactive/Impulsive composite).

Relative risk analyses (with 99.9% confidence intervals), were conducted to measure the associations between each variable and the presence and severity of self-injury and the associations between each variable 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 self-injury, a series of Chi Square tests were utilised 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 theoretical predictive models for the presence and severity of self-injury and the presence of self-restraint, binary logistic regressions were also conducted.

4.4 Results

4.4.1 Sample characteristics and age differences

Prior to investigating the aims and testing the hypotheses, prevalence data for characteristics of the sample were generated for the two age groups. Table 4.1 displays the prevalence of males, those with lower ability, those with one or more health problems, those with high Repetitive/Restricted behaviours and those with high Overactive/Impulsive behaviours for both groups. Additional data are presented on the frequency of scores for individuals in each group.

Table 4.1 Demographic and behavioural 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)[3]	**16.81**	**<.001**
Lower ability	36.1 (75)	52.3 (113)[4]	**12.30**	**<.001**
Health problems	38.5 (80)[5]	61.1 (132)[6]	**20.19**	**<.001**
0 health problems	60.6 (123)	38.6 (83)	-	-
1 – 2 health problems	27.6 (56)	36.4 (78)	-	-
3 – 4 health problems	8.9 (18)	18.2 (39)	-	-
5 – 6 health problems	2.5 (5)	3.8 (8)	-	-
7 – 8 health problems	0.5 (1)	3.2 (7)	-	-
High Repetitive/ Restricted	42.8 (89)[7]	60.6 (131)[8]	**14.48**	**<.001**
Score 0 on Repetitive/Restricted	25.7 (53)	7.6 (16)	-	-
Score 1 – 2 on Repetitive/Restricted	20.9 (43)	20.3 (43)	-	-
Score 3 – 4 on Repetitive/Restricted	26.7 (55)	26.9 (57)	-	-
Score 5 – 6 on Repetitive/Restricted	11.6 (24)	21.2 (45)	-	-
Score 7 – 8 on Repetitive/Restricted	15.1 (31)	24.1 (51)	-	-

[3] Missing data, N =1, 0.5%
[4] Missing data, N =3, 1.4%
[5] Missing data, N =5, 2.4%
[6] Missing data, N =1, 0.5%
[7] Missing data, N=2, 1.0%
[8] Missing data, N =4, 1.9%

Table 4.1 cont Prevalence of demographic and behavioural characteristics for child and adult samples. Significant differences are highlighted in bold (p <.01; 2 tailed)

Characteristic	% (N) Child (N = 208)	% (N) Adult (N = 216)	Chi Square	P value
High Overactive/Impulsive	39.4 (82)	50.0 (108)	4.79	.029
Score 0 on Overactive/Impulsive	24.0 (50)	17.6 (38)	-	-
Score 1 – 4 on Overactive/Impulsive	36.6 (76)	32.4 (70)	-	-
Score 5 – 8 on Overactive/Impulsive	22.6 (47)	28.3 (61)	-	-
Score 9 – 12 on Overactive/Impulsive	12.5 (26)	14.9 (32)	-	-
Score 13 – 16 on Overactive/Impulsive	4.3 (9)	7.0 (15)	-	-

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 behaviour in the adult sample. There were no significant differences between the groups in numbers of individuals with high levels of Overactive/Impulsive behaviour.

4.4.2 Prevalence and topographies of self-injurious behaviour

In order to investigate the first aim of the study, prevalence data were generated for the child sample and adult sample for self-injury, severity of self-injury and topographies of self-injury. Table 4.2 reveals that 45.7% of the child sample and 49.1% of the adult sample engaged in self-injury; 18% of the child sample and 19.9% of the adult sample engaged in severe self-injury. For both groups, the most frequent topography of self-injury 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 self-injury. The number of topographies of self-injury displayed by both groups was broadly similar.

Table 4.2 Prevalence, severity and topographies of self-injury for the child and adult samples.

	Behaviour	**% (N)**		**Chi Square**	**P value**
		Child (N = 208)	**Adult (N = 216)**		
Presence of SIB	All self-injury	45.7 (95)[9]	49.1 (106)	0.43	.513
Severity of SIB	Severe self-injury	18.8 (39)[10]	19.9 (43)[11]	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

[9] Missing data, N =1, 0.5%
[10] Missing data, N =9, 4.3%
[11] Missing data, N =6, 2.8%

Table 4.2 cont. Prevalence, severity and topographies of self-injury for the child and adult samples.

	Behaviour	% (N) Child (N = 208)	% (N) Adult (N = 216)	Chi Square	P value
Numbers of Topographies of SIB	0 topographies of self-injury	53.8 (112)[12]	50.9 (110)[13]	-	-
	1 topography of self-injury	13.9 (29)	13.9 (30)	-	-
	2 topographies of self-injury	13.0 (27)	13.0 (28)	-	-
	3 topographies of self-injury	5.8 (12)	5.6 (12)	-	-
	4 topographies of self-injury	3.8 (8)	4.6 (10)	-	-
	5 topographies of self-injury	0.5 (1)	3.2 (7)	-	-
	6 topographies of self-injury	1.4 (3)	0.9 (2)	-	-
	7 topographies of self-injury	1.4 (3)	0.0 (0)	-	-

In summary, self-injurious behaviour was displayed by 45.7% of the child sample and 49.1% of the adult sample. Between 15 and 20% of both samples engaged in more severe self-injurious behaviour. There were no differences in the topographies of self-injury displayed between the adult and child samples.

4.4.3 Prevalence and topographies of self-restraint

In order to investigate the second aim of the study, prevalence data were generated for child and adult samples for topographies of self-restraint behaviour. Chi square tests were conducted to test for differences between the two groups. Table 4.3 reveals that 40.9 % of the child sample and 42.6 % of the adult sample engaged in self-restraint behaviour. Additional

[12] Missing data, N =13, 6.3%
[13] Missing data, N =15, 6.9%

analysis revealed that significantly more individuals with self-injury engaged in self-restraint than those without self-injury in both the child ($\chi^2 = 19.97$, $p < .001$) and adult ($\chi^2 = 21.55$, $p < .001$) samples. Significantly more children than adults engaged in a particular topography of self-restraint: holding onto others or holding onto others' clothing. 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.

Table 4.3 Prevalence and topographies of self-restraint behaviour for the child and adult samples. Significant differences are highlighted in bold (p <.01; 2 tailed)

	Behaviour	% (N) Child (N = 208)	% (N) Adult (N = 216)	Chi Square	P value
Presence of self-restraint	All self- restraint	40.9 (85)[14]	42.6 (92)[15]	0.03	.959
	Self-restraint if self-injury occurs	*56.8 (54)*	*57.5 (61)*	-	-
	Self-restraint if self-injury 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
Number of Topographies of self-restraint	0 topographies of self-restraint	53.8 (112)	55.6 (120)	-	-
	1 topography of self-restraint	21.2 (44)	23.1 (50)	-	-
	2 topographies of self-restraint	10.6 (22)	11.6 (25)	-	-
	3 topographies of self-restraint	6.3 (13)	6.5 (14)	-	-
	4 topographies of self-restraint	1.0 (2)	1.4 (3)	-	-
	5 topographies of self-restraint	1.0 (2)	0.0 (0)	-	-
	6 topographies of self-restraint	1.0 (2)	0.0 (0)	-	-

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

[14] Missing data, N =11, 5.3%

[15] Missing data, N =4, 1.9%

In summary self-restraint behaviour was displayed by approximately 40% of both the child and adult samples and was significantly associated with the presence of self-injury. The prevalence of one topography of self-restraint differed between the two age groups. Interestingly, this was the only topography of self-restraint that involved others.

4.4.4 Variables associated with self-injurious behaviour

In order to investigate the third hypothesis of the study, a series of relative risk statistics were calculated to assess the association between demographic and behavioural variables and the presence and severity of self-injury. Table 4.4 displays the relative risk statistics for the child and adult samples for each variable and the presence and severity of self-injury. As can be seen in the table, being male did not increase the risk for self-injurious behaviour. Lower ability was associated with an increased risk of self-injury for the adult sample, and with an increased risk of severe self-injury for the child sample. Health problems were associated with an increased risk of self-injury and severe self-injury for the child sample. High Repetitive/Restricted behaviour was associated with an increased risk of self-injury for the child sample and with severe self-injury for all both samples. High Overactive/Impulsive behaviour was associated with an increased risk of self-injury, and severe self-injury for both samples.

Table 4.4 Relative risk statistics for variables associated with the presence of self-injury and severe self-injury. Significant relative risk statistics are highlighted in bold (p <.01)

Risk Variable	Behaviour	Relative Risk Statistics (99% CI, p <.01) Child (N = 208)	Adult (N = 216)
Male	SIB	0.71 (0.44 – 1.13)	0.87 (0.60 – 1.26)
	Severe SIB	0.66 (0.26 – 1.66)	1.06 (0.49 – 2.33)
Lower Ability	SIB	1.46 (1.00 – 2.13)	**1.50 (1.02 – 2.19)**
	Severe SIB	**3.44 (1.57 – 7.58)**	2.03 (0.94 – 4.43)
Health Problems	SIB	**1.66 (1.13 – 2.43)**	1.16 (0.79 – 1.70)
	Severe SIB	**2.54 (1.19 – 5.42)**	1.23 (0.58 – 2.62)
High Repetitive/Restricted	SIB	**2.50 (1.64 – 3.80)**	1.41 (0.94 – 2.12)
	Severe SIB	**4.48 (1.81 – 11.11)**	**3.07 (1.13 – 8.34)**
High Overactive/Impulsive	SIB	**2.23 (1.51 – 3.31)**	**2.12 (1.41 – 3.18)**
	Severe SIB	**6.14 (2.37 – 15.90)**	**3.78 (1.54 – 9.28)**

In summary, the relative risk statistics revealed that gender did not increase the risk of self-injurious behaviour. Lower ability, health problems and high Repetitive/Restricted behaviour increased the risk of self-injury and severe self-injury in some cases. High Overactive/Impulsive behaviour consistently increased the risk of self-injury and severe self-injury in all samples.

4.4.4.1 Investigation of health variables associated with self-injurious behaviour

In order to further evaluate the relationship between health problems and self-injurious behaviour in the child population, a series of Chi square tests were calculated to assess the association between specific types of health problems and the presence of self-injury. Table 4.4 displays the prevalence of health problems and Chi square tests for the child sample for each health problem and the presence of self-injury.

Table 4.5 Prevalence and Chi square statistics for specific forms of health problems associated with the presence of self-injury in the child sample. Significant Chi square statistics are highlighted in bold (p <.01; one tailed)

Health Problem	**% (N)**		**Chi Square**	**P value**
	Self-Injury (N = 95)	**No Self-Injury (N = 112)**		
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[16]	8.4 (8)	9.8 (11)	0.15	.352

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

The results in Table 4.5 reveal that there were significantly more skin and digestive problems in the sample of children who engaged in self-injury. Additionally, the difference between

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

levels of ear problems between the sample who engaged in self-injury and the sample who did not approaches significance.

4.4.5 Variables associated with self-restraint

In order to test the fifth hypothesis of the study, a series of relative risk calculations were conducted; first investigating the relative risk of self-restraint given differing severity of self-injury and second the relative risk of self-restraint given behavioural markers associated with self-injury (Ability, Repetitive/Restricted and Overactive/Impulsive). A third series of relative risk calculations were made to assess the risk of self-restraint given the presence of *both* self-injury *and* the behavioural markers of Ability, Repetitive/Restricted behaviour and Overactive/Impulsive behaviour. These can be found in Appendix F. As can be seen in Table 4.6, all variables significantly increased the risk of self-restraint except for the presence of low ability in the adult sample.

Table 4.6 Relative risk statistics for variables associated with the presence of self-restraint. Significant relative risk statistics are highlighted in bold (p <.01)

	Relative Risk Statistics (99% CI, p <.01)	
Variable	**Child (N = 208)**	**Adult (N = 216)**
SIB	**2.12 (1.34 – 3.35)**	**2.12 (1.36 – 3.31)**
Severe self-injury	**1.77 (1.14 – 2.74)**	**2.17 (1.51 – 3.11)**
Low ability	**1.54 (1.02 – 2.32)**	1.55 (1.00 – 2.41)
High Repetitive/Restricted	**2.38 (1.51 – 3.74)**	**1.62 (1.01 – 2.59)**
High Overactive/Impulsive	**2.35 (1.52 – 3.64)**	**1.88 (1.21 – 2.91)**

In summary, the presence of self-injury, severe self-injury, high Repetitive/Restricted behaviour and high Overactive/Impulsive behaviour significantly increased the risk of self-restraint in both child and adult samples. Low ability only increased the risk of self-restraint in the child sample, although this approached significance in the adult sample.

4.4.6 Logistic regression for predictors of self-injury and self-restraint

In order to control for the overlap between variables, to produce predictive models of self-injury and self-restraint, a series of binary logistic regressions were conducted. If the relative risk of self-injury 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 conducted 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 self-injury and those not, those displaying severe self-injury and those not, and those displaying self-restraint and those not. Table 4.7 reveals the results of the logistic regressions, and indicates the variables which made a significant independent contribution to each of the models.

Table 4.7 Logistic regression predicting likelihood of displaying self-injury, severe self-injury 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	Nagelkerke R squared	Correct Classification of cases (%)	Predictor Variables	*B*	S.E	Wald	*Df*	*p*	Odds Ratio	95.0% C.I. for Odds Ratio	
														Lower	Upper
Presence of self-injury															
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 self-injury															
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

Table 4.7 cont. Logistic regression predicting likelihood of displaying self-injury, severe self-injury 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
Self-restraint															
Child (N = 194)	43.09	4	<.001	.20	.27	73.7	**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
							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
Adult (N = 205)	29.89	3	<.001	.13	.18	67.3	**SIB**	1.09	.31	12.21	1	**<.001**	2.97	1.61	5.47
							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

Figure 4.1 outlines the significant findings of these models. The logistic regressions revealed that, independently of other factors, children and adults with high Overactive/Impulsive behaviour were significantly more likely to show self-injury. Children with high Repetitive/Restricted behaviour were more likely to show self-injury but high Repetitive/Restricted behaviour did not contribute to the adult model for presence of self-injury. In addition to contributing to the presence of self-injury, high Overactive/Impulsive behaviour scores also increased the likelihood of severe self-injury in both the child and adult samples. For the child sample, the presence of health problems significantly increased the likelihood of self-injury and severe self-injury. Low ability only contributed to the presence of severe self-injury in the child sample. Finally, children with high Overactive/Impulsive behaviour scores, high Repetitive/Restricted behaviour scores and self-injury were more likely to display self-restraint whereas only the presence of self-injury contributed to self-restraint in the adult sample.

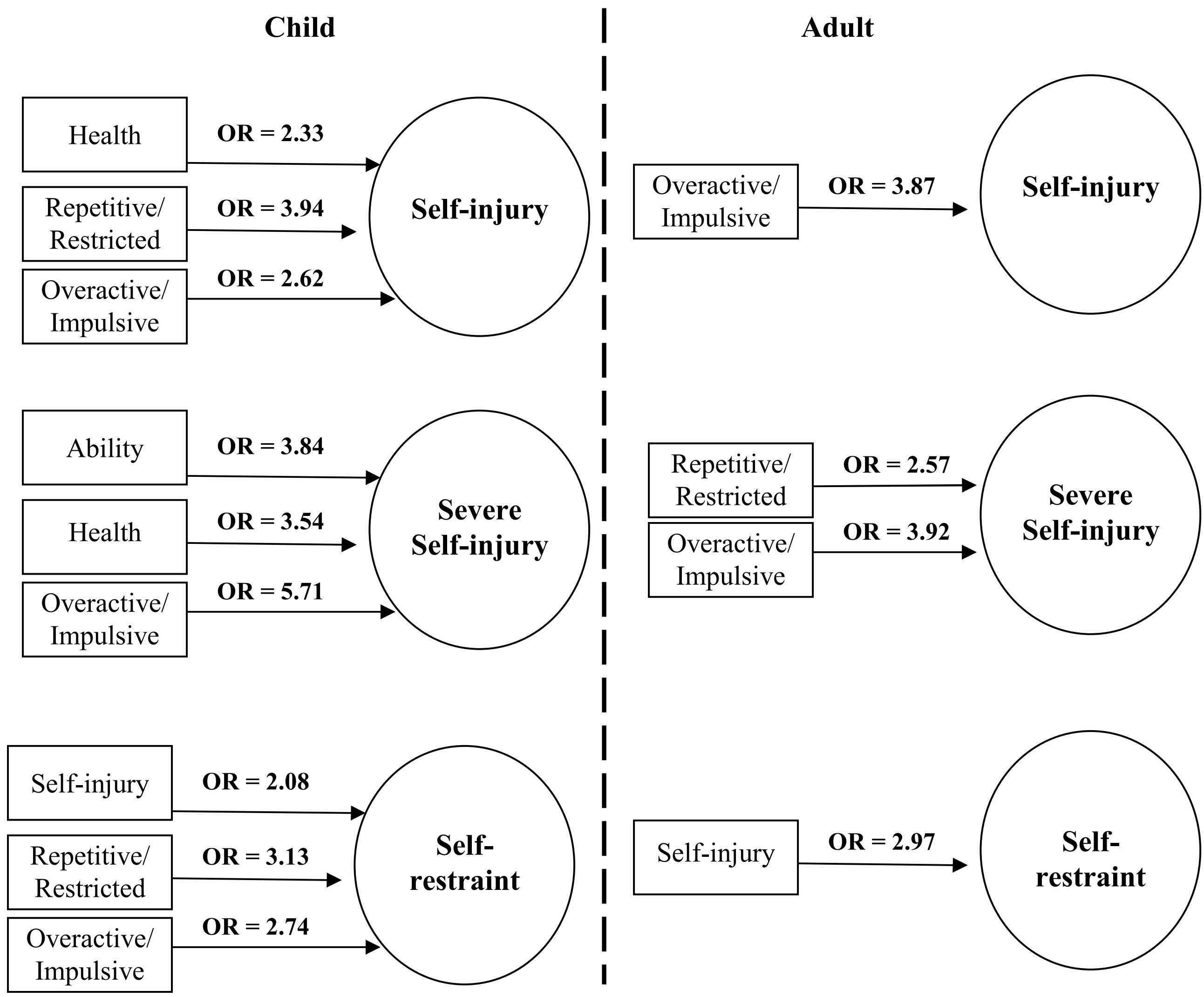


Figure 4.1 Significant independent variables predictive of self-injury, severe self-injury and self-restraint for the child and adult samples

In conclusion, repetitive, overactive and impulsive behaviours, lower ability and the presence of health conditions contributed to self-injury and severe self-injury. The presence of self-injury, repetitive, overactive and impulsive behaviours contributed to the presence of self-restraint.

4.5 Discussion

The prevalence and topographies of self-injury and self-restraint in a group of children and adults with ASD were delineated in this study. The demographic and behavioural characteristics associated with self-injury and self-restraint were then investigated in both groups. Finally, the independent predictive value of these characteristics was examined in order to evaluate a model of behavioural inhibition combined with self-injury predicting the presence of self-restraint. Drawing a large sample from the NAS ensured a relatively homogenous group where ASD diagnosis has been previously confirmed. Conducting the data analysis separately for the child and adult samples, and including ability level as a variable within analysis, ensured that the potential confounds of age and ability were controlled within the study. The utilisation of logistic regression allowed the independent contribution of each variable to be assessed within a large sample. The study is novel in its investigation of health conditions and self-injury and self-restraint in an ASD population. Additionally, the study has a novel focus to model build theories of self-injury, self-restraint and compromised behavioural control.

The results indicated a high prevalence of self-injury of 45.7% in the child sample and 49.1% in the adult sample. This figure supports the prevalence data reported in Chapters 2 and 3 (50.0% - Section 2.4.1; 41.7% - Section 3.4.2) and adds further confirmation to a growing body of research identifying the prevalence of self-injury in ASD as between 40 and 50% (Ando & Yoshimura, 1979a; Baghdadli *et al.,* 2003; Shattuck *et al.,* 2007). There were no significant differences in the prevalence of self-injury for the child and adult samples, providing cross-sectional data supporting the longitudinal evidence for persistence of self-injury identified in Chapter 3 (Section 3.4.2). There were also no differences between the two samples in the numbers of individuals engaging in severe self-injury. Again, although limited conclusions can be drawn, this may indicate that whilst self-injurious behaviour is persistent, levels of severity do not differ across age groups. Finally, there were no differences between the child and adult samples in the topographies of self-injury displayed, and supporting findings in Chapter 2 (Section 2.4.1), the most prevalent form of self-injury was hitting self with a body part. The prevalence figures for this topography (24.5 % of children; 28.2 % of adults) are very similar to the prevalence of 29.5% reported in Chapter 2. The overall similarities in prevalence data identified in this sample, and the sample surveyed in Chapters 2 and 3 suggest that these findings are robust and reliable[11].

When investigating variables associated with self-injury in ASD, the results revealed that gender did not increase the risk of self-injury or severe self-injury. This result supports findings reported in Chapter 2 and 3 (Sections 2.4.3 & 3.4.3) and in previous research (Bagdadli *et al.,* 2003; McClintock *et al.,* 2003) and indicates that for individuals with ASD, being male does not increase the risk of self-injury. The presence of lower ability significantly increased the risk of self-injury in the adult sample, but not in the child sample. However, the logistic regression revealed that lower ability was not independently predictive of self-injury for the adult sample. The relative risk analysis revealed that lower ability did increase the risk of severe self-injury in the child sample, and the logistic regression confirmed an independent effect of lower ability for severe self-injury. These results partially support previous findings associating lower ability and self-injury in ASD (Bagdadli *et al.,* 2003; 2008; Shattuk *et al.,* 2007). However the results from this study are mixed, with differing results dependant on the

[11] It is assumed that the sample assessed in Chapters 2 & 3 is independent of the sample assessed in Chapter 4. However, as the sample in Chapter 4 were recruited through the NAS schools/adult services, it is plausible that there may be a limited number of individual's who took part in both survey studies. As the data collected in Chapter 4 were often anonymised by the schools/services, it is not possible to verify this. However, given the significantly larger sample collected in Chapter 4, it is not felt that this warrants significant concern.

age group and severity of self-injury investigated. Further research utilising robust cognitive measures of ability could usefully augment the results presented in this study.

The relative risk analysis also revealed that the presence of health problems significantly increased the risk of self-injury and severe self-injury in the child sample. The logistic regression supported these findings, demonstrating that the presence of health problems significantly predicted the presence of self-injury and the presence of severe self-injury in the child sample. This finding supports previous research associating painful health conditions and self-injurious behaviour (Christensen *et al*., 2009; Luzzani *et al.,* 2003) and indicates a key area for clinical intervention. Extending previous research, this study revealed associations between specific types of health problems and self-injury, demonstrating that levels of skin and digestive problems were significantly elevated in the self-injury sample. Additionally, the difference in ear problems approached significance. These data provide valuable information regarding causal hypotheses of pain and self-injury. Whilst skin problems may be a result of self-injurious behaviour (e.g., skin picking, scratching or biting), it is less likely that digestive problems and ear problems are a direct consequence of self-injury. Therefore, it can be hypothesised that these specific health problems, which have also been identified in other populations as associating with self-injury (Digestive problems: Luzzani *et al.,* 2003; Ear problems: O' Reilly, 1997) may be causally implicated in the development and maintenance of self-injury. This study provides preliminary evidence of such an association, however significant further research is required in order to test experimentally the relationship between pain and self-injury in individuals with ASD.

Although health problems were significantly predictive of self-injury in the child sample, they did not increase the risk of self-injury in the adult sample. The difference in influence of health problems upon self-injury between the child and adult samples may be due to the significantly higher overall levels of health problems in the adult sample. Alternatively, the results may indicate that health problems are implicated in the early development of self-injury in childhood, but not in the maintenance of self-injury in adulthood. Longitudinal data are required in order to test this hypothesis. Taken together, these novel findings associating painful health conditions and self-injury in individuals with ASD suggest that pain may be causally implicated in self-injury in this population. Therefore, clinical evaluations of self-injurious behaviour should include a full health assessment in order to rule out or treat the influence of pain upon self-injury.

High levels of repetitive behaviour, and overactive and impulsive behaviour significantly increased the risk of self-injury in both samples. This increase was most consistent for overactive and impulsive behaviours, which significantly increased the risk of self-injury and severe self-injury for both groups. This supports the pragmatic assertion in Chapter 2 to treat differences in overactive and impulsive behaviour as clinically significant. The logistic regression revealed that high levels of repetitive behaviour only predicted the presence of self-injury in the child sample, whereas high levels of overactive and impulsive behaviour independently predicted the self injury in both groups and severe self-injury in both groups. Taken together, these findings support those reported in Chapter 2 (Sections 2.4.3 & 2.4.4), Chapter 3 (Sections 3.4.3 & 3.4.5) and previous research (Cooper *et al.,* 2009; Collacott *et al.,* 1998; Oliver *et al.,* 2009; Schneider *et al.,* 1996). This body of evidence consistently associating ADHD like behaviours of overactivity and impulsivity with self-injurious behaviour lends tentative support to a model in which impairments in inhibition may be implicated in the presence of self-injury. These data and those presented in Chapter 2 (see Section 2.4.3.1) also provide evidence that the presence of overactive and impulsive behaviour is associated with more severe self-injury. This provides preliminary support to the assertion that impairments in inhibition may contribute to both the presence and increasing severity of self-injury.

The results of the self-restraint analysis revealed that 40.9% of the total child sample, and 42.6% of the total adult sample engaged in self-restraint behaviours. Importantly, 56.8% of children who engaged in self-injury, also engaged in self-restraint and 57.5% of adults who engaged in self-injury also engaged in self-restraint, evidencing a significant association between the presence of self-injury and self-restraint. The prevalence figures for self-restraint reported in this study fall between those reported by Oliver *et al.* (2003; 46%) and Powell *et al.* (1996; 76%) indicating that individuals with ASD engage in a similar level of self-restraint as individuals with intellectual disability. There were no significant differences in the prevalence of self-restraint in the child and adult samples, which contrasts with previous findings indicating lower age was associated with self-restraint (Fovel *et al.,* 1989). 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 utilisation of mechanical 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 behaviours are more dependant on external support than in adulthood. It is possible that gaining support from others provides the mechanism by which independent self-restraint enters a behavioural repertoire. It may be that parents and carers initially provide physical support and restraint for a child engaging in self-injury, and through a process of learning the child eventually begins to engage in self-restraint. This warrants further investigation.

In line with previous research, the presence of self-injury significantly increased the risk of self-restraint in both child and adult samples (Fovel *et al.,* 1989; Hyman *et al.,* 2002). The results extend previous findings by also demonstrating that severe self-injury increases the risk of self-restraint, suggesting an interaction between severity of self-injury and self-restraint. These findings provide support for the theory that self-restraint is displayed by those who engage in self-injury in order to reduce their self-injurious behaviour, particularly those showing severe self-injury (Forman, Hall & Oliver, 2002; Marzullo *et al.,* 2009; Rojahn, *et al.,* 1978; Smith *et al.,* 1992). However, an interesting finding which is often overlooked in the self-restraint literature, is that 26.8% of children and 28.2% of adults who *did not* engage in self-injury still engaged in self-restraint. If self-restraint was simply a learned behaviour, negatively reinforced by the absence of self-injury, then individuals who do not engage in self-injury, should not engage in self-restraint either. The presence of self-restraint in those who do not self-injure suggests that although inhibition of self-injury may be one function of self-restraint, it may not be the only function. An alternative although relatively implausible explanation for these data that must be considered is that the self-restraint displayed by these individuals is so effective at reducing self-injury, that no self-injury is displayed.

The results also revealed that lower ability significantly increased the risk of 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 behaviours significantly increased the risk of 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 self-injury; self-injury and self-restraint, ability and repetitive behaviour).

Finally, high levels of overactive and impulsive behaviour significantly increased the risk of self-restraint in both populations. This finding builds upon the reported association between compulsive behaviour and self-restraint (Hyman *et al.,* 2002; King, 1993; Powell *et al.,* 1996), and indicates that self-restraint is associated with the hypothesised impairments in behavioural control which contribute to the presence of self-injury. The logistic regression revealed that overactivity and impulsivity independently predicted the presence of self-restraint for the child sample. Overactive and impulsive behaviours did not predict self-restraint in the adult model; however the presence of self-injury did. In interpreting these differences in findings, it is important to highlight that there were no significant differences in the presence of self-injury, self-restraint or overactive and impulsive behaviours 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 behavioural control, as evidenced through impulsive and overactive behaviour, leads to the early development of self-restraint during childhood. This supports the assertion made earlier that self-restraint may not only function to inhibit self-injurious behaviour. It may be that for a group of children with poor behavioural inhibition, self-restraint develops during childhood in order to help control a variety of behaviours and impulses. This theory would explain why self-injurious behaviour was not predictive of self-restraint in the child sample; it may that during childhood, self-restraint occurs to control poor behavioural inhibition and is displayed in order to inhibit a range of behaviours, not specifically self-injury. However, in adulthood, it is merely the presence of self-injury, and not the presence of overactive and impulsive behaviours, that predicts the presence of self-restraint. It would seem that with development, self-restraint comes to function primarily to reduce self-injurious behaviour, rather than as a response to general impairments in behavioural control. This may happen through a process of operant learning, as over time self-restraint behaviour is negatively reinforced by the avoidance of self-injury, and develops into a functional behaviour within an individual's repertoire. This tentative model requires significant further research, specifically detailing the development of self-injury, self-restraint and behavioural inhibition. Ideally, this research would contain a strong cognitive component or a behavioural test of inhibition in order to delineate the precise nature of impairment in inhibition. Results from this future research may open the possibility of cognitive interventions to improve behavioural control. If this were possible, it is plausible that an intervention for behavioural inhibition may result in a decrease in self-injurious behaviour, self-restraint behaviour and overactive and impulsive behaviours.

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

A number of caveats to these findings must be considered. Firstly, no independent verification of ASD diagnosis was conducted, and this was highlighted in Chapter 1 as a key threat to validity in prevalence studies (Section 1.4.1). However, the NAS require a diagnosis of ASD from a professional for every child and adult in their service. Given the costs of service provision, it is unlikely that the NAS would provide services for individuals without ASD. Additionally, the prevalence rates for self-injury reported in this study are very similar to those reported in studies which used independently confirmed ASD diagnoses (Baghdadli *et al.,* 2003; Dominick *et al.,* 2007; Shattuck *et al.,* 2007) suggesting that the sample characteristics can be assumed to be similar. Secondly, there may be a sample bias given that all children and adults were recruited through the NAS. However, given that the NAS is the largest specialist service provider for individuals with ASD, and the purpose of this study was to model build self-injury and self-restraint in ASD, the sample choice seems sensible. The results however, must be interpreted with this possible bias in mind.

In summary, the results indicate that lower ability, health problems, overactive, impulsive, and repetitive behaviour significantly independently predict self-injury and severe self-injury. The presence of overactivity and impulsivity independently predicted self-restraint for children, and the presence of self-injury independently predicted self-restraint for adults. A developmental model of self-injury and self-restraint, underpinned by impairments in behavioural control is hypothesised to account for these findings.

CHAPTER 5

Experimental Functional Analysis of Self-Injurious Behaviour in Children with Autism Spectrum Disorder

5.1 Preface

The study in Chapter 4 used survey methodology to identify variables associated with the presence and severity of self-injury and the presence of self-restraint. The value of these variables to predict self-injury and self-restraint was then evaluated, in order to build a model of self-injury and self-restraint in ASD. The findings demonstrated that key risk markers of lower ability, health problems, overactive, impulsive, and repetitive behaviours predicted self-injury. Additionally, overactivity, impulsivity and self-injury were associated with the presence of self-restraint. These findings were used to build a preliminary developmental model of self-injury and self-restraint in ASD, which allows for the generation of further hypothesis driven research.

Thus far, the thesis has focused on risk and model building using indirect assessments of large samples. These studies have contributed to a theoretical understanding of self-injury in ASD and have provided useful epidemiological evidence for service provision. The review in Chapter 1 highlighted the utility of operant theory to a comprehensive understanding of self-injurious behaviour (Section 1.3.3). Therefore, this study will utilise direct assessment methods to conducting fine grained analyses of the operant function of self-injury in a sample of children with ASD within the context of experimental functional analysis.

5.2 Introduction

Previous research and the results of this thesis have demonstrated that self-injury is more prevalent in individuals with ASD than in individuals with intellectual disability of heterogeneous aetiology (See Sections 1.4.1, 2.4.1, 3.4.2 & 4.4.2). The elevated prevalence in individuals with ASD may suggest biological difference in this population which leads to an increase in self-injury. However, a theory that reflects the empirical literature suggests that self-injurious behaviour may result from an interaction between person characteristics, that might be associated with or definitive of a neurodevelopmental disorder, and operant influences in the environment (Langthorne, McGill & O'Reilly, 2007; Langthorne & McGill, 2008; Oliver, 1993; Tunnicliffe & Oliver, 2011). Whilst these 'gene-environment' interactions are beginning to be explored in individuals with rare genetic syndromes (e.g., Rett syndrome: Oliver, Murphy, Crayton & Corbett, 1993; Williams syndrome: O' Reilly, Lacey & Lancioni, 2000), this theory has not been applied and investigated in individuals with ASD. This is perhaps due to ASD being defined by behavioural rather than genetic criteria. Nonetheless, a coherent account of the elevated levels of self-injury in ASD may usefully borrow from this literature, substituting a behaviourally defined phenotype for a genetic one, and investigating the interactions between this ASD phenotype and operant influences upon self-injurious behaviour.

In single-case experimental design and cohort studies it has been demonstrated that self-injury can be an operant behaviour sensitive to reinforcement contingencies (Oliver, 1995; Oliver, Hall & Murphy, 2005). Experimental functional analyses, such as those described by Iwata, Dorsey, Slifer, Bauman & Richman (1994) and Carr and Durand (1985) are proven, robust techniques for identifying the antecedents and consequences involved in evoking and maintaining self-injury in individuals who are non-verbal or who have a severe to profound intellectual disability. However, despite robust evidence supporting the operant model of self-injury (Section 1.3.3.1) and the heightened prevalence of self-injury in ASD, (Sections 2.4.1, 3.4.2 & 4.4.2) there is a paucity of research investigating reinforcement contingencies maintaining self-injury in individuals with ASD. A recent review urged researchers to pay more attention to this clinically relevant area (Matson & LuVollo, 2008), with another study suggesting that:

> "there is a pressing need to examine what makes so many contexts aversive...for children with ASD and how these antecedent events can be changed..." (Blakeley-Smith, Carr, Cale & Owen-DeSchryver, 2009, p.132).

Initial evidence from experimental functional analysis in individuals with intellectual disability, suggested that this method was able to identify social reinforcers for self-injury in 64.4% of cases (Iwata *et al.,* 1994). These commonly utilised experimental functional analysis techniques typically employed a 'control' or 'play' condition, a low attention condition and task demand condition. Therefore, the conditions assess typical social functions of escaping from demands and gaining access to attention. Through the use of 'alone' conditions, automatically reinforced functions can also be delineated. These typical functional analysis conditions have been used to investigate self-injury in individuals with ASD, but with fewer conclusive results (Borrero & Borrero 2008; Hagopian, Bruzek, Bowman & Jennett, 2007; Hausman, Kahng, Farrell & Mongeon 2009; Healey, Ahearn, Graff & Libby, 2001; McKerchar, Kahng, Casioppo & Wilson, 2001; O'Reilly, Sigafoos, Lanioni, Edrisinha & Andrews, 2005). The majority of studies have used single case experimental designs with functional analytic methods based on those described by Carr and Durand (1985) or Iwata *et* *al.* (1994). Some studies have demonstrated social functions for self-injury in individuals with ASD (Borrero & Borrero 2008; O'Reilly *et al.,* 2005). However, other studies have failed to demonstrate social function, often due to very low levels of self-injury being displayed (Hagopian *et al.,* 2007; Hausman et al., 2009) or due to high, undifferentiated levels of self-injury occurring across all conditions (Healey *et al.,* 2001; McKerchar *et al.,* 2001). The results of these studies suggest variable results from the most commonly employed functional analyses regarding the function of self-injury in individuals with ASD.

To date, no cohort studies have been conducted using functional analysis to assess self-injury specifically in individuals with ASD. However, a cohort study investigating the function of challenging behaviour was conducted by Love, Carr and LeBlanc (2008) for 32 children with ASD. The results demonstrated that challenging behaviour was maintained predominantly by access to social reinforcement and the authors conclude that function for challenging behaviour in ASD can be conceptualised and assessed similarly to challenging behaviour in intellectual disability populations. In contrast, O'Reilly *et al.* (2010) conducted functional analyses for challenging behaviour across ten children with ASD, and did not identify a social function of escape from demand or gaining access to attention, for eight of the children. Therefore, O'Reilly and colleagues suggest that individuals with ASD are less likely to have social functions for their challenging behaviour and are more likely than other groups to display challenging behaviour maintained by automatic reinforcement. As both studies analysed all challenging behaviours together, it is difficult to assess these results for self-injury specifically. Additionally, both cohort studies, and all of the single case studies, relied exclusively upon visual inspection of data to ascertain function. Finally, all studies, except for that conducted by O'Reilly *et al.* (2010), did not assess ASD diagnosis in participants.

Consequently, the results of all of these studies are compromised by threats to internal, external and statistical validity, although the most robust studies do appear to suggest that commonly employed functional analysis may not reveal a social function for challenging behaviour in children with ASD (O'Reilly *et al.,* 2010). In order to assess the function of self-injury in individuals with ASD accurately, a cohort study utilising robust assessment of ASD diagnosis, statistical analysis, and assessment of specific topographies of self-injury is required.

Whilst there are limited data from cohort studies investigating self-injury in ASD, several single case experimental studies have been conducted. However, alongside the methodological problems highlighted above, many of these have evidenced inconclusive results. Further investigation of these results reveals two key patterns in the data; firstly, undifferentiated responding or high levels of challenging behaviour occurring across all conditions was evident in a number of the studies (Healey *et al.,* 2001; McKerchar *et al.,* 2001). In these cases, a social function was not obtained as levels of responding in the 'test' conditions could not be differentiated from levels of responding in the 'control' condition. It is therefore often assumed, by default, that the challenging behaviour is automatically reinforced (O'Reilly *et al.* 2010). However, an alternative explanation for the lack of differentiation between the test and control conditions is the employment of a social interaction condition as a control. Impairments in social interaction are a necessary diagnostic component of ASD using DSM-IV criteria, and these impairments in social interaction may present in a number of different ways. Some individuals with ASD may lack the social motivation to engage in interactions, and may perhaps therefore present as 'aloof', actively avoiding social contact, or 'passive', allowing social contact to occur but not initiating or engaging in it (Wing & Gould, 1979). Alternatively, individuals with ASD may evidence impairments in social skills whilst maintaining preserved social motivation, resulting in markedly different or 'odd' social interactions (Wing & Gould, 1979). In all cases, it is plausible that deficits in social skills and communication may result in enforced social interactions, such as those utilised in the commonly employed 'play' or control conditions of experimental functional analysis, being aversive for individuals with ASD.

The hypothesis that enforced social interaction may be aversive to individuals with ASD, has good face validity. The interaction between a 'play' condition and the given social impairments in ASD may result in participants having to engage in an interaction for which they have limited motivation or limited skills through which to exercise any control of the interaction. Support for the proposed aversive nature of social interaction for individuals with ASD may be evidenced by the presence of social anxiety in those with ASD (Simonoff *et al.* 2008) and in those with genetic syndromes associated with ASD (Cornelia de Lange syndrome, Richards, Oliver, Moss, Kaur & O'Farrell, 2009; Fragile X syndrome, Dykens & Volkmar, 1997). Taken together, this hypothesis would suggest that enforced social interaction in the standard 'play' condition may be uncomfortable, demanding or aversive to those with ASD. Therefore, the 'control' condition may actually comprise a setting event for challenging behaviour in individuals with ASD, resulting in elevated levels of challenging behaviour in the 'control' condition.

In line with a hypothesis of social interaction as a setting event, Hagopian, Wilson & Wilder (2001) reported that the highest levels of challenging behaviour, including self-injury, occurred in the play condition for a young boy with autism. They confirmed subsequently a hypothesis of social escape by conducting a modified play condition, in which a break from social interaction was provided contingent upon challenging behaviour. Similarly, Tiger, Fisher, Toussaint and Kodak (2009) report the successful utilisation of a social escape condition to identify the function of aggressive behaviour for a boy with ASD. Interestingly, the authors note their surprise that social escape has rarely been identified as a function for challenging behaviour in individuals with ASD. These case studies provide a robust method for assessing social escape function. However, the experimental evidence is limited to single case studies with the same threats to validity detailed above. These studies may have been strengthened by the inclusion of assessments to detail person characteristics such as ASD diagnoses and intellectual disability status, which is highlighted in the introduction as a critical component of robust research in ASD (Section 1.4.5). Additionally, the results of the study rely upon visual inspection of data that may be open to bias. Consequently, further investigation of social escape as a function for self-injurious behaviour specifically, is required, in parallel with assessments to ascertain ASD diagnosis, and statistical analysis to confirm function.

In addition to the undifferentiated responding or high levels of challenging behaviour across all conditions, a second data pattern resulting in inconclusive functional analysis results was identified; that of consistently low levels of responding across all conditions (Hagopian, *et al.,* 2007; Hausman *et al.,* 2009). Several single case studies have attempted to progress beyond these initially undifferentiated results, by identifying alternative establishing operations that have occasioned the behaviour, or alternative reinforcement contingencies which maintain the behaviour. Hagopian *et al.* (2007) identified that interruptions to free operant behaviour occasioned challenging behaviour, including self-injury, in two boys with ASD. Maintaining access to a repetitive behaviour or items involved in a repetitive behaviour and avoiding the termination of a complex ritual, have been identified as functions for challenging behaviour in individuals with ASD (Hausman *et al.,* 2009; Murphy, McDonald, Hall & Oliver 2000; White *et al.,* 2011). Additionally, several single case studies have identified noise as an antecedent for challenging behaviour in individuals with ASD (Buckley & Newchock, 2006; Devlin, Healy, Leader & Reed, 2008). Taken together, these studies suggest that there may be 'ASD related'[1] functions to challenging behaviour, including self-injury, in individuals with ASD. In these cases a phenotype x environment interaction may be in operation; whereby ASD characteristics result in certain environmental conditions being more aversive or reinforcing to individuals with ASD. These studies demonstrate the utility of building unique functional analysis conditions to identify these ASD related functions that could not be identified through the most commonly employed functional analysis assessments. However, as with the single case studies identifying social escape function, none of these studies confirmed ASD diagnosis or used statistical analysis to support their visual inspection of function. Further investigation of these ASD related functions using statistical analyses, and including rigorous assessment of person characteristics is therefore necessary.

Broadly speaking, many of the identified functions reported above can be hypothesised to be influenced by ASD symptomatology. Individuals with ASD are reported to exhibit high levels of sensory sensitivity (Kientz & Dunn, 1997; Ornitz, Guthrie, & Farley, 1977; Rogers, Hepburn & Wehner, 2003). Therefore, it could be hypothesised that they will have a lower tolerance for aversive noises *and* find a higher proportion of typically tolerable noises aversive (O'Reilly *et al.,* 2000). Consequently, individuals with ASD would be expected to

[1] The term 'ASD related' functions will be used throughout to refer to operant functions for challenging behaviour which are associated with ASD or person specific characteristics such as preference for routine, repetitive behaviours or specific objects and sensory sensitivity.

show more challenging behaviour occasioned by noise, if the behaviour is reinforced by escape from noise, than individuals with intellectual disability of heterogeneous aetiology. This interaction between ASD characteristic and environment is also plausible in the case of repetitive behaviours. Again, individuals with ASD exhibit unusually high levels of repetitive and stereotyped behaviours, and these behaviours are often assumed to be 'compulsive' and\or highly preferred (Estes *et al.,* 2011; Richler, Bishop, Kleinke & Lord, 2007; Turner, 1999). Therefore, given the elevated presence of repetitive behaviours in this population, it is more likely that these behaviours will naturally be interrupted and therefore more likely for this social function to emerge. Additionally, although less plausible, interruption to repetitive behaviours displayed by those with ASD could be hypothesised to create a more significant deprivation as the behaviour may be more preferred. Therefore, interruptions to repetitive behaviour in individuals with ASD would be more likely to occasion challenging behaviour than interruptions to behaviour in other individuals.

The reinstatement of repetitive behaviours as a function of challenging behaviour for individuals with ASD has been investigated using interview methodology (Reese, Richman, Zarcone & Zarcone, 2003; Reese, Richman, Belmont & Morse, 2005). These studies demonstrated that gaining access to repetitive activities and escaping demands whilst engaged in these repetitive activities frequently contributed to challenging behaviour for children with ASD. The results also revealed that these functions for challenging behaviour were significantly more common in individuals with ASD than in matched individuals without ASD, supporting a hypothesis that this function develops in individuals with ASD due to the heightened prevalence of repetitive behaviours. Importantly, both studies employed robust assessment of ASD diagnosis, enabling the findings to be generalised to the broader ASD population. However, the utilisation of indirect assessment does not allow conclusions regarding cause to be drawn (Section 1.5.2). The results of these studies indicate that there may be ASD related functions to challenging behaviour, which could be predicted by a consideration of the characteristics of those with ASD. To date, no study has investigated ASD related functions of self-injury in a cohort of individuals with ASD using direct experimental assessments.

In summary, research has demonstrated variable results for the most commonly employed functional analyses in identifying social function for self-injury in individuals with ASD. However, to date, there have been no cohort studies investigating the function of self-injury using commonly employed functional analysis, with appropriately confirmed ASD diagnosis, intellectual disability *and* assessed function at the level of topography, using statistical methods. Evidence is emerging for the consideration of functions for challenging behaviour that are directly influenced by ASD symptomology, for instance, the avoidance of social interaction or interruptions to repetitive behaviours. However, again the evidence is limited to single case studies with poor validation of person characteristics, and interview based assessments of function. Therefore this study will assess function of self-injury in a cohort of children with ASD, utilising the most commonly employed functional analysis conditions, allowing comparison to previous research. Progressing from previous research, a hypothesised ASD related function of social escape will be assessed in addition to the more commonly employed functional analysis conditions. Finally, the study will allow for the functional assessment of ASD related reinforcement contingencies. In order to guard against threats to validity, this study will also conduct independent assessments of ASD phenomenology and intellectual disability for each participant. Final, novel statistical analysis of the functional analysis results will be implemented to ensure statistical validity.

The following predictions are made:

i) The most commonly employed[2] functional analyses will not reveal function for self-injury for the *majority* of participants with ASD.

ii) The inclusion of a social escape condition in the standard functional analysis will reveal a function for self-injury for *some* participants.

iii) Function will be identified for the *majority* of participants through the utilisation of ASD related experimental functional analysis[3].

[2] For brevity, the term 'standard' will be used to refer to the most commonly employed functional analysis conditions throughout the methods and results, as described by Iwata *et al.,* (1994) and Carr and Durand (1985).
[3] These assessments will be broadly termed 'ASD modified functional analyses'; indicating that the functional analysis has been tailored to investigate an ASD related function for a given child.

5.3 Methods

5.3.1 Recruitment

Participants were recruited from the National Autistic Society (NAS) schools identified and surveyed in Chapter 4. Data from the questionnaires were examined and children were selected initially if they were under 16 years of age, showed self-injury weekly and were non-verbal[4]. For practical reasons, the two NAS schools in London were selected for the first wave of recruitment, in order to conduct research visits as close to Birmingham as possible. If insufficient numbers were recruited from these schools, further recruitment from the other three NAS schools was planned. Using these initial selection criteria, 16 children were identified across the two schools.

Following this selection, letters were sent to the Head Teachers of the two schools, explaining the study, and identifying the children who had met the initial selection criteria. Head Teachers were asked to confirm which of the children met the inclusion criteria for the study. The inclusion criteria necessitated that the child was under 16 years of age, had a severe to profound intellectual disability and a diagnosis of ASD, was non verbal (less than 30 functional words), did not have a diagnosis of a genetic syndrome and exhibited daily self-injurious behaviour. Additionally, Head Teachers were asked to identify any other children who also met the inclusion criteria, who had not been identified in the questionnaire screen due to joining the school recently or due to exhibiting self-injury which had started or become more severe after the screen was conducted. Through this process, two children were screened out of the study, and an additional eight children were identified that met the inclusion criteria. This left a total sample of 22 children.

[4] Previous research has predominantly been conducted in individuals who are non-verbal. Therefore, in order to allow comparison to previous data, non-verbal participants were recruited for this study.

A brief letter outlining the study was then sent to parents/carers of the 22 children (see Appendix G). This letter summarised the study and asked parents to contact the lead investigator to discuss further the study if they were interested in taking part. When parents/carers contacted the lead investigator, a final screening interview was conducted to ensure the child currently met the inclusion criteria. Following this, an extended information pack with consent forms was sent to parents/carers with a freepost return envelope (see Appendix H).

Five families did not reply to the initial information sent to them and one family decided not to take part following the screening interview. A further four families were excluded after the screening interview due to low levels of self-injury, or high levels of ability in the child. A final two families expressed initial interest, but did not complete and return the consent forms within the 12 month period of the study. This left a final sample of 10 children who met the inclusion criteria and whose parents/carers had completed and returned the consent forms.

5.3.2 Participants

Table 5.1 describes the participant characteristics of the 10 children. As can be seen in the table, all participants were under 16 (Mean age = 11.85; SD = 2.94), six were male and all participants met diagnostic criteria for autism or ASD on both the Autism Diagnostic Observation Schedule (ADOS; Mean total score = 17.20; SD = 2.20) and the Social Communication Questionnaire[5] (SCQ; Mean total score = 20.35; SD = 5.45). Additionally, all participants had a severe to profound degree of intellectual disability (relative IQ score <50)

[5] Excluding Participant 3 whose SCQ total score is below the threshold for ASD. The SCQ was completed by a teacher for this participant as the parent/carer was unable to do so. The lifetime SCQ includes items regarding the behaviour of the child between ages four and five and the teacher was unable to complete this accurately for the participant. Therefore, the participant was included in the study on the basis of their ADOS score.

as demonstrated by the full scale ratio IQ calculated from the Mullen Scales of Early Learning[6] (Mullen; Mean ratio IQ = 13.33; SD = 4.37). Participants had a low level of adaptive functioning (Adaptive Behavior Composite, ABC, score <70) as measured by the Vineland Adaptive Behavior Scales (Mean = 40.33; SD = 6.54). The topographies of self-injury demonstrated by participants were varied and are described in the table.

[6] The Mullen Scales of Early Learning do not produce an IQ score, however a relative IQ was calculated for each participant using a methodology demonstrated by Richler *et al.,* 2007. For more detail see Measure section 5.3.3.1. Whilst it is acknowledged that confidence in measurement of IQ scores <50 is limited, these data were included in order to allow for future comparisons.

Table 5.1 Age, gender, Mullen full scale ratio IQ score, VABS adaptive composite standard score, ADOS total score, SCQ total score and description of self-injurious behaviour displayed for each participant.

			Ability		Autism		
Participant	**Age**[7]	**Gender**	**Mullen Ratio IQ**	**VABS ABC score**	**ADOS total score**[8]	**SCQ total score**[9]	**Self-injurious behaviour**
One	15:06	Male	14.25	42	16	23	Hand to object hit
Two	12:07	Female	17.38	-[10]	13	11.5	Scratch self, Leg slap Head hit
Three	15:07	Male	9.23	36	19	22	Mouth hit, Finger bite
Four	7:00	Male	10.42	49	20	26	Head bang, Teeth hit Bite self, Bite hard object Hand to object hit
Five	11:05	Male	15.69	38	18	24	Bite self, Bite hard object, Hit self, Hand to object hit, Head bang
Six	13:05	Female	11.96	40	19	29	Hand bite, Head bang Hit self, Hand to object hit
Seven	14:05	Male	14.31	38	16	17	Skin pick, Hit self Hand to object hit
Eight	14:06	Male	13.51	37	15	16	Hand to hand hit, Wrist hit, Hit self, Eye press
Nine	8.11	Female	21.03	52	19	15	Bite self, Head bang, Hit self, Scratch self
Ten	13:03	Female	5.50	31	17	20	Bite self, Cheek press

[7] Age in years: months; calculated from the date of the first observational assessment
[8] Cut off for ASD = 7; cut off for autism = 12
[9] Cut off for ASD = 15 ; cut off for autism = 22
[10] It was not possible to complete the Vineland Adaptive Behavior Scales with the parent/carer of this participant.

5.3.3 Measures

5.3.3.1 Measures of ability

The following two measures were utilised to estimate cognitive and adaptive behaviour levels in participants.

The Mullen Scales of Early Learning (Mullen, 1995)

The Mullen Scales of Early Learning is a standardised developmental assessment. It is intended to assess children with a mental age from birth through to 68 months. The Mullen Scales of Early Learning yields scores on five scales: Visual Reception, Fine Motor, Receptive Language, Expressive Language and Gross Motor (the Gross Motor scale is not used to calculate a child's IQ). Typically, each scale results in a T-score, and the sum of the primary four T-scores creates an early learning composite score. The Mullen Scales of Early Learning has very good internal consistency (coefficients range from .75 to .83), test-retest reliability (coefficients range from .71 to .96) and inter-rater reliability (coefficients range .93 to .99). The measure also has good construct and concurrent validity (Mullen, 1995).

As the population tested in this study were older than the 68 month upper limit for the T scores in the Mullen Scales of Early Learning, an alternative method was utilised to calculate Ratio IQ scores (this method was first demonstrated by Richler *et al.,* 2007). All four non-verbal and verbal subtest age equivalents were averaged to create a full total age equivalent score. This full scale age equivalent was then divided by the chronological age, and multiplied by 100. This resulted in a full scale ratio IQ.

Vineland Adaptive Behavior Scales (Sparrow, David & Cicchetti, 1985)

The Vineland Adaptive Behavior Scale (VABS) is a semi-structured interview which is suitable for use with individuals with or without intellectual disability. It assesses personal and social adaptive behaviour levels and level of intellectual disability. Items comprise four domains: Communication Skills, Daily Living Skills, Socialisation Skills and Motor Skills. An overall Adaptive Behavior Composite is calculated from this. It has good internal consistency, test-retest reliability, inter-rater reliability, and validity.

5.3.3.2 Measures of autism

In order for each child to receive a place at an NAS school, they were required to have received a diagnosis of autism or ASD from a qualified health professional, Psychologist or Speech and Language Therapist. However, in order to examine the homogeneity of the sample and to operationalise the inclusion criteria, the following measures were administered.

*Autism Diagnostic Observation Schedule (*Lord *et al.,* 2000)

The Autism Diagnostic Observation Schedule is a semi-structured, standardised observational assessment of communication and social interaction skills, play and imaginative skills and repetitive behaviour. The assessment incorporates the use of clear planned social 'presses' that provide the opportunity for the participant to display certain behaviours or responses; these behaviours and responses are then recorded. The ADOS consists of four modules, and selection of a particular module is based on the individuals expressive language. Module 1 was used for all participants in this study, as it is selected for individuals with no speech or simple phrases only. Sensitivity, specificity and inter-rater reliability are reported to be robust (Lord *et al.,* 2000). Concurrent validity between the ADOS and the Autism Diagnostic

Interview (*rs* = .57) and the Social Communication Questionnaire (*rs* = .55) is good (Howlin & Karpf, 2004; Rutter, Bailey & Lord, 2003).

Social Communication Questionnaire – Current Version (SCQ; Berument, Rutter, Lord, Pickles & Bailey, 1999)

The SCQ consists of 40 items related to behaviours and characteristics associated with ASD demonstrated during the previous three months, the questionnaire provides scores for three subscales: social interaction, communication and repetitive behaviour. All items require a yes/no response and are scored as 0 or 1 respectively. Total scores range from 0 to 39 (excluding items regarding language). A cut off of 15 and 22 for ASD and autism respectively has been proposed (Rutter, Bailey & Lord, 2003). Good concurrent validity has been found with other measures of ASD (Bishop & Norbury, 2002; Howlin & Karpf, 2004).

5.3.3.3 <u>Assessment of self-injurious behaviour</u>

The Challenging Behaviour Interview (CBI; Oliver *et al.,* 2003)

The CBI was used to provide a detailed description of the participants' challenging behaviour through teacher and parent report. Conducted in two parts, the respondent is asked whether the participant has shown one of the following three types of behaviour within the last month: self injury, physical aggression and disruption of the environment. Each behaviour is operationally defined and examples given. For the purposes of this study, data were only collected on self-injury. The second part of the interview assesses the severity of each topography of behaviour identified in part one through the summation of fourteen questions. Each of these items is based on a 4 or 5 point Likert scale, the description of each point depends on the specific question. The authors reported good inter-rater and test-retest reliability kappa indices for the behaviours in part 1 and 2 of the interview: part 1 (.67, range: .50-.80 and .86, range: .70-.91 respectively) and part 2 (.48, range: .02-.77 and .76, range: .66-.85). See Appendix I for a copy of the Challenging Behaviour Interview.

The Functional Analytic Interview

A semi-structured interview was used in order to identify common triggers for self-injury for each participant. The Functional Analytic Interview asked parents/carers and teachers about events/situations which always or almost always lead to an episode of self-injury (see Appendix J for a copy of the Functional Analytic Interview).

5.3.3.4 Observational measures

The following observational measures were used to assess self-injury.

Standard experimental functional analysis

For each participant, experimental functional analyses based on those by Iwata *et al.* (1994) and Carr and Durand (1985) were employed. Three test conditions were paired with a control condition using an alternating treatment design (ABACAD), this combination was repeated four times to evaluate the influence of social reinforcement on self-injurious behaviour. Each condition lasted two and a half minutes which is shorter than many condition lengths in the published literature. However, a recent review of experimental functional analysis revealed that brief session durations are as effective as longer sessions (Hanley, Iwata & McCord, 2003). Given the breadth of assessments conducted in this study and the reported frequency of self-injury in participants, two and a half minute conditions were utilised.

In order to comply with the requirements of the schools where the assessments were being conducted, a visual timetable was constructed with a different symbol for each condition. For all children this timetable was explained prior to the start of the experimental functional analysis. At the end of each condition, the symbol was removed from the visual timetable, and replaced with the symbol for the next condition. The inclusion of the visual timetable encouraged children to discriminate more accurately between conditions and thus improved the validity of the results (see Conners *et al.,* 2000). The visual timetable was used for the standard, extended and ASD modified experimental functional analyses.

The conditions in the standard experimental functional analysis were as follows:
Condition A: High Attention. This condition was analogous to Carr and Durand's (1985) 'Easy 100' condition and involved the researcher maintaining high levels of verbal attention with the participant without issuing any demands. This condition acted as a control condition for the presence of the researcher and the presence of attention. The researcher maintained proximity to the participant throughout the condition and this included moving around the room with the participant. If the participant spoke to or approached the other researcher in the room the high level of verbal attention was maintained. The rooms in which the sessions were carried out were always as distraction-free as possible. However, there were times when participants would comment on or pick up objects in the room. At these times, the researcher would remark briefly on the object without introducing the object into the session. Any challenging or inappropriate behaviours shown by the participant were not responded to in any way.

Condition B: Low Attention. This condition was analogous to Carr and Durand's (1985) 'Easy 33' condition and began with the researcher in close proximity to the participant. The researcher then removed attention from the participant by saying "I'm going to talk to X now" and then talking to the other researcher in the room. In this condition it was not necessary for the researcher to maintain proximity with the participant and so mobile participants were free to move around the room. This was to ensure that any approaches directed toward the researcher could be observed. All behaviours except for self-injury were ignored; this included any attempts by the participant to initiate interaction. Verbal and physical attention were given contingent on self-injurious behaviour. The attention was a standard verbal statement of concern – "Don't do that, you'll hurt yourself" accompanied by physical attention, such as rubbing the participant's arm or removing their hand. After five seconds of attention, the researcher withdrew their attention again. This condition allows the positive reinforcement through attention delivery hypothesis to be tested when compared with the results from Condition A. Higher levels of self-injurious behaviour in this condition would be indicative of behaviour occasioned by low levels of adult attention and maintained by contingent attention.

Condition C: Task Demand. This condition was analogous to Carr and Durand's (1985) 'Difficult 100' condition and involved the researcher prompting the participant through a task that their teacher or parent had identified as being difficult. Tasks varied considerably depending on the ability of the participant; commonly used tasks included shape sorters, jigsaw puzzles and matching tasks. A three-point prompting procedures of verbal, verbal and gestural (or model) and physical prompt was used throughout. Each stage of the prompt occurred approximately three seconds after the previous prompt if the participant had not completed the task. Completion of the task, for both independent and prompted responding, was consequenced with verbal praise and physical attention (mostly rubbing of the participant's arm or back). Contingent on self-injury, the task and the researcher's attention were removed for ten seconds but all other behaviours were ignored. When the researcher removed their attention, they said to the child ‘Ok, we don’t have to do it’. If the self-injurious behaviour continued longer than the ten second ‘time-out’ period, the task was not reinstated until there had been five seconds with no self-injury. This condition allowed a negative reinforcement through escape from demands hypothesis to be tested. High levels of target behaviour in this condition compared to Condition A would suggest behaviour occasioned by an aversive task and maintained by contingent removal (escape) of that task.

Condition D: Social Escape. This condition was again analogous to Carr and Durand’s (1985) ‘Easy 100’ condition and involved the researcher maintaining high levels of verbal attention with the participant without issuing any demands. The researcher maintained proximity to the participant throughout the condition and this included moving around the room with the participant. If the participant spoke to or approached the other researcher in the room the high level of verbal attention was maintained. However, in this condition the researcher’s attention was removed for ten seconds contingent upon self-injurious behaviour, but all other behaviours were ignored. When the researcher removed their attention, they said to the child ‘Ok, I’ll leave you alone’. If the self-injurious behaviour continued longer than the ten second ‘time-out’ period, the task was not reinstated until there had been five seconds with no self-injury. In order to help the participant discriminate between this condition, and Condition A (high attention), the Social Escape condition was always run by the second researcher so that the second researcher would act as a discriminative stimuli (S^D) to the child for the availability of negative reinforcement through the removal of social attention (Conners *et al.,* 2000). High levels of target behaviour in this condition compared to Condition A would suggest that the behaviour was occasioned by aversive social contact and maintained by escape from that contact.

Extended standard experimental functional analysis

Extended standard experimental functional analysis conditions were conducted for either of the following reasons. First, if self-injurious behaviour had occurred in standard experimental function analysis condition but differentiation between the condition and the control had not been achieved. Secondly, if reports from both parents/carers and teachers on the Functional Analytic Interview indicated an attention maintained/escape or demand escape function. For example, if a participant had displayed low levels of self-injury in the demand condition, but differentiation from the control condition was not achieved, an extended demand condition with control would be conducted. Similarly, if both parent/carer and teacher had reported a demand escape function to the participant's self-injury, an extended demand condition with control would be conducted, regardless of the levels of self-injury seen in the initial standard functional analyses. This was done in order to ensure that the brief time of the initial standard functional analysis was not obscuring the function of the self-injury.

Extended standard experimental functional analysis conditions were identical to those described above (Condition A: high attention, Condition B: low attention, Condition C: task demand, Condition D: social escape), utilising the same contingencies for self-injurious behaviour. The condition lengths were five minutes, rather than two and a half minutes. The condition in which the self-injurious behaviour had occurred at low levels was paired with an appropriate control i.e. high attention or in some cases low attention. In order to further enhance differentiation between the conditions, the test and control conditions were run by the first and second researcher respectively. The test and control conditions were paired in an alternating treatment design (ABABABAB) and each condition was repeated four times. As with the standard experimental functional analysis, a visual timetable was utilised for each participant.

ASD modified functional analysis

This assessment was conducted if neither the standard experimental functional analysis nor the extended experimental functional analysis had revealed a social function to the self-injurious behaviour.

This assessment used a similar procedure to that of Structured Descriptive Assessments to build ASD related conditions to assess potential ASD and person specific functions to self-injury. Structured Descriptive Assessments (e.g. Anderson & Long, 2002) programme antecedents hypothesised to be related to the behaviour into a natural observation period. However, in order to maintain experimental control, the antecedents in this case were built into functional analysis conditions, run by the researchers with standardised contingencies and timings. The test conditions were derived from answers within the Functional Analytic Interview. Parents/carers and teachers were independently asked about ‘high risk’ situations in which the child’s self-injury would almost certainly occur. Where parents/carers and teachers both reported the same situations, a test condition was designed to reflect this, e.g., where changes to routines were reported, a test condition was constructed in which a set routine was periodically changed. Additionally, a control condition was also constructed e.g., a condition in which the routine is allowed to run as expected with no changes. The test and control conditions were run by the first and second researcher respectively in order to increase discrimination between conditions. The test and control conditions were paired in an alternating treatment design (ABABAB) and the test and control conditions were repeated the same number of times. As with the standard experimental functional analysis, a visual timetable was utilised for each participant. Table 5.3 describes the extended and ASD modified functional analysis conditions conducted for each participant.

Table 5.2. Extended and ASD modified conditions, including class of condition, explanation of condition, control and test descriptions and condition lengths for each participant. For all conditions, four repeats were conducted unless otherwise indicated.

Participant	Class of Condition	Condition	Control *(Condition length)*	Test *(Condition length)*
One	Extended	Extended social escape	Low attention *(5 minutes)*	Social escape *(5 minutes)*
	ASD modified: Repetitive behaviour	Change to routine of visual timetable	Timetable of two activities completed *(2 minutes per activity)**	Test 1: Addition of activity to timetable; no consequence for self-injury Test 2: Removal of activity from timetable; no consequence for self-injury *(2 minutes per activity)**
	ASD modified: Repetitive behaviour	Interruption to repetitive behaviour of colour sorting blocks	Free access allowing colour sorting *(2.5 minutes)*	30s of free access, examiner disruption to colour sorting; removal of examiner disruption for 10 seconds contingent upon self-injury *(2.5 minutes)*
Two	Extended	Extended social escape	Low attention *(5 minutes)*	Social escape *(5 minutes)*
	ASD modified: Repetitive behaviour	Access to tangible involved in repetitive behaviour (box of blocks)	Free access to tangible* *(30 seconds)*	Tangible removed; access granted for 10 seconds contingent upon self-injury *(1 minute)**
Three	ASD modified: Sensory	Sensory escape of singing	Verbal high attention *(1 minute)*	Singing high attention; removal of singing for 10s contingent upon self-injury *(1 minute)*

Table 5.2 cont. Extended and ASD modified conditions, including class of condition, explanation of condition, control and test descriptions and condition lengths for each participant. For all conditions, four repeats were conducted unless otherwise indicated.

Participant	Class of Condition	Condition	Control *(Condition length)*	Test *(Condition length)*
Four	Extended	Extended demand	High attention *(5 minutes)*	Task demand *(5 minutes)*
Five	ASD modified: Behaviour interruption	Interruption to any free operant behaviour	Free access to tangibles and examiner attention *(5 minutes)*	30s free access, then child behaviours and access to tangible/attention interrupted; access granted for 10 seconds contingent upon self-injury *(5 minutes)*
Six	ASD modified: Repetitive behaviour	Repetitive questioning ignored	High attention with repetitive questions answered *(2.5 minutes)***	High attention with repetitive questions ignored; repetitive questions answered when occurred with self-injury *(2.5 minutes)***
Seven	Extended	Extended social escape	Low attention *(5 minutes)*	Social escape *(5 minutes)*
	Extended	Extended demand	Low attention *(5 minutes)*	Task demand *(5 minutes)*
	ASD modified: Restraint availability	Restraint availability	Control 1: Low attention *(2.5 minutes)* Control 2: Social escape *(2.5 minutes)*	Social escape condition with self-restraint blocked; social attention removed and restraint allowed for 10s contingent upon self-injury *(2.5 minutes)*

Table 5.2 cont. Extended and ASD modified conditions, including class of condition, explanation of condition, control and test descriptions and condition lengths for each participant. For all conditions, four repeats were conducted unless otherwise indicated.

Participant	Class of Condition	Condition	Control *(Condition length)*	Test *(Condition length)*
Eight	Extended	Extended demand	Low attention *(5 minutes)*	Task demand *(5 minutes)*
	ASD modified: Tangible	Access to tangible (ipod with preferred music)	Free access to tangible *(2.5 minutes)*	30s free access, tangible removed; access granted for 10 seconds contingent upon self-injury *(2.5 minutes)*
Nine	Extended	Extended demand	High attention *(5 minutes)*	Task demand *(5 minutes)*
	ASD modified: Sensory	Sensory escape	Quiet environment maintained in classroom *(1 minute)******	30s quiet, loud whistle blown from outside classroom; no consequence for self-injury *(1 minute)******
Ten	ASD modified: Tangible	Transitions to alternative rooms	Transition to preferred room *(Transition+ 2 minutes of free time in preferred room)*	Transition to standard analogue room; no consequence for self-injury *(Yoked to time taken to complete each control condition)*
	Repeated standard	Repeat of standard analogues in alternative room	Full set of standard analogues conducted in alternative neutral room *(2.5 minute conditions of all)*	

* = Three repeats of conditions conducted.
** = Eight repeats of conditions conducted due to low frequency of repetitive questions.
*** = Eight repeats of conditions conducted due to low frequency of sudden noise.

5.3.4 Procedure

5.3.4.1 Protocol

Following successful recruitment (detailed above, Section 5.3.1), parents/carers were contacted to conduct the initial interviews necessary before data collection visits could begin. This included a risk assessment and the Functional Analytic Interview. Data collection visits were then organised with the respective school and parents/carers, and questionnaire measures including the Social Communication Questionnaire were distributed to parents/carers and schools. Data collection visits were conducted over a period of 12 months. During the first set of visits for each child, interview data were collected from schools (Functional Analytic Interview and Challenging Behaviour Interview) and further interviews were conducted with parents/carers (Challenging Behaviour Interview and Vineland Adaptive Behavior Scales).

Results from the Functional Analytic Interview item were used to construct the ASD modified functional analyses. Where *both* parents/carers and school teachers reported a common trigger for self-injury, an ASD modified functional analyses was conducted. Table 5.3 displays the common triggers that were independently identified by both parents/carer and school teachers for each participant.

Table 5.3 Events/situations leading to self-injurious behaviour identified by teachers and parents/carers identified through the Functional Analytic Interview for each participant

Participant	Events/situations leading to self-injurious behaviour identified by teachers and parents/carers
One	Changes/interruptions to routines
Two	Removal of preferred items
Three	Sudden, unexpected noises; removal of preferred items (music, computer)
Four	No common events/situations identified
Five	Being told 'no', access to preferred items
Six	Adults not providing answers, or providing incorrect answers, to repetitive questions
Seven	No common events/situations identified
Eight	Being asked to do something he doesn't want to do; removing preferred items
Nine	Sudden, unexpected noises e.g., babies and children, being asked to do something she doesn't want to do
Ten	Transitions to certain places/rooms

The results of the Functional Analytic Interview revealed events/situations independently reported by both parents/carers and teachers for eight of the ten participants. For these participants, the results of the Functional Analytic Interview were used to build ASD modified functional analysis conditions where appropriate. No common events/situations leading to self-injurious behaviour were reported for participants 4 and 7. Therefore, for these participants, extended standard experimental functional analysis and ASD modified functional analysis were based upon results of the initial standard experimental functional analysis.

During initial visits for each child, the standard experimental functional analysis, Mullen Scales of Early Learning and Autism Diagnostic Observation Schedule were conducted. Data from the standard experimental functional analysis were then briefly analysed to ascertain whether function for self-injury had been obtained. If it had not, dependant on the results of the standard experimental functional analysis and the Functional Analytic Interview (above), extended experimental functional analysis or ASD modified functional analysis were conducted. ASD modified functional analyses were conducted for as many identified events/situations reported jointly by parents/carers and teachers, or until function for self-injurious behaviour had been obtained.

If children became excessively distressed or self-injury levels were significantly higher than that typically seen, functional analysis conditions were terminated. This decision was made if either the first or second researcher, or the member of observing staff from the school felt the child was becoming excessively distressed. If a condition was terminated, the same condition was repeated at a later time. To prevent loss of data, the data from the terminated condition were utilised up to the point at which the condition was terminated. The data from the repeat of the terminated condition were then appended to the original data ensuring that all session lengths were uniform (e.g., if a standard experimental functional analysis High Attention condition was terminated at one minute, then this one minute would be coded, and the first one and a half minutes of the repeated High Attention condition would also be coded and appended to the original condition, resulting in a two and half minute condition). Finally, although data collection took place over 12 months, a paired control and test condition were always conducted on the same day to control for participant and environment variability.

All conditions were video-recorded and coded subsequently in real time. Ethical approval for this study was obtained from the School of Psychology ethical review committee at the University of Birmingham.

5.3.4.2 Data coding and reliability

Adult and child behaviours were coded using real time Obswin software (Martin, Oliver & Hall, 2000). Some self-injury topographies were common across children, and some topographies were specific to individual children. Verbal and physical contact from researchers was coded to assess integrity of the experimental conditions. Behaviours were either coded as duration variables (e.g., examiner communication), where the behaviour onset and offset was recorded, or as event variables, where only the behaviour occurrence was recorded (e.g., head hit). An operational definition for each behaviour and a list of participants who were recorded to show this behaviour can be seen in Table 5.4. Inter-observer reliability using 3 second time intervals was calculated for 25% of all video recorded sessions. Mean Cohen's Kappa values was .87 (range 0.47 – 1.00) illustrating good agreement (Fleiss, 1981).

Table 5.4. Coded behaviours, operational definitions, participants who displayed this behaviour and Kappa values.

Code	Definition	Participant	Kappa
Adult codes			
Examiner comment	Examiner speaks to child, includes rhetorical questions	All	.88
Examiner demand	Examiner makes verbal/physical request of child ('do')	All	.91
Examiner denial	Examiner makes verbal/physical request to stop or not do something ('don't). May include physically moving child, removing objects, blocking door.	All	.83
Physical contact	Examiner touches the child	All	.68
Child Codes			
Bite hard object	Child clenches jaw and teeth around a solid object (e.g., wood, chair, table). Must use top and bottom teeth and definite biting movement.	4,5	.82
Bite self	Child clenches jaw and teeth around own body part	4,5,9,10	.87
Cheek press	Child forcefully pushes thumbs into cheek	10	.92
Eye press	Child pushes fingers on eye, pulls or pokes eyelashes	8	.97
Finger bite	Child clenches jaw and teeth on side of finger	3	1.00
Hand bite	Child clenches jaw and teeth on hands or arms	6	.67
Hand to hand hit	Child forcefully brings hands together, palm to palm	8	.81
Hand to object hit	Child forcefully makes contact with hard surface with palm of hand	1,4,5,6,7	.47
Head bang	Child forcefully makes contact with head against hard surface	1,4,5,6,9	.80
Head hit	Child forcefully makes contact with fist and/or open palm on their head	2	1.00
Hit self	Child forcefully makes contact with fist and/or open palm on own body part	5,6,7,8,9	.80
Leg slap	Child forcefully makes contact on leg with palm of hand	2	.79
Mouth hit	Child forcefully makes contact on mouth/around mouth with palm of hand	3	1.00
Scratch self	Child drags nails across or pinches their own body part	2,9	1.00
Skin pick	Child scrapes skin using forefinger at right angles to the surface of skin	7	.89
Teeth hit	Child forcefully makes contact with top teeth against object	4	1.00
Wrist hit	Child forcefully makes wrist to wrist contact	8	1.00

5.3.5 Data analysis

The analysis to explore social function in self-injury for each participant was conducted at two levels. First, the percentage duration of self-injury across the standard experimental functional analysis conditions and ASD modified functional analysis conditions were plotted in multi element graphs. Each datum point on the graph represents the percentage of time the participant spent engaging in self-injurious behaviour during that trial of the standard experimental functional analysis or ASD modified functional analysis condition.

Second, the dominance statistic (*d;* Cliff, 1993) was calculated for each participant's self-injurious behaviour across the experimental functional analysis or ASD modified functional analysis conditions. The *d* statistic is a measure of the extent to which one sample distribution lies above another and this is determined by comparing each score in one condition to all scores in another condition using a dominance matrix. In order to calculate whether self-injury had an attention maintained function, each datum point (datum point = percentage of intervals self-injury occurred in) from the low attention condition was compared to each datum point from the high attention condition. Demand escape function was determined by comparing each task demand condition datum point to each datum point from the paired high attention condition. Social escape function was determined by comparing each social escape condition datum point to each datum point from the paired high attention condition. Finally, function from ASD modified functional analysis was determined by comparing each test condition datum point to each datum point from the paired control condition. The sample value *d* is the mean value of the elements in the dominance matrix. A *d* value of +1 would indicate that every datum point in a series is greater than every other datum point in another series. A *d* value of -1 would indicate that every datum point in a series is less than every other datum point in another series. In order to compare the *d* statistic for function at a categorical level, a conservative cut-off value was arbitrarily nominated. A *d* value of between 0 and .70 indicates no function, whilst any *d* value over .70 would suggest functional behaviour[11].

5.3.5.1 Assessment of topography

Most participants displayed more than one topography of self-injury. If the frequency of occurrences of each individual topography of self-injury was less than 20, function was analysed for all self-injury topographies combined. If the frequency of occurrences of each individual topography was equal to or greater than 20, then function was determined for each topography separately. If the *d* statistic revealed the same function for the different topographies, then overall function of self-injury is presented for brevity. If the *d* statistic revealed different functions for the different topographies of self-injury, then the function of each topography is presented separately. Table 5.5 displays the frequencies for each topography of self-injury for each participant.

The results in Table 5.5 reveal that Participant One only displayed one topography of self-injury. Participants Two, Three, Four, Five and Nine displayed one topography of self-injury more than 20 times, however all other topographies of self-injury had a frequency of less than 20. Therefore, for these participants, due to the low frequency of the majority of their topographies of self-injury, the data were collapsed, and analysed for self-injurious behaviour as a whole. Similarly, Participant Six did not display any topography of self-injury with more than 20 occurrences and therefore the data were collapsed.

[11] This value was selected as it indicates that the individual was displaying more self-injury in approximately three of the four test conditions compared to the control conditions. This was felt to be a clinically significant difference between the test and control conditions.

Participants Seven, Eight and Ten displayed multiple topographies of self-injury with frequencies of onset of equal to or greater than 20[12]. Consequently, for each individual topography, *d* statistics were calculated comparing the control and test conditions in the standard, extended and ASD modified functional analyses. For Participants Seven and Eight, none of the individual topographies obtained *d* statistic values over 0.7 for any condition, indicating that there was no difference in function across topographies. Therefore, for Participants Seven and Eight, all of the topographies were combined. For Participant Ten, in the ASD modified functional analysis, one topography of self-injury (Bite Self) gained a *d* statistic value over 0.7, and the second topography of self-injury displayed by Participant Ten (Cheek Press) did not. This indicates that there was a difference in function across the topographies, and consequently, for Participant Ten, the function for each topography is presented separately. For all of the *d* statistic results evaluating differences in topography, see Appendix K

[12] Whilst one topography of self-injury for Participant 7 only had a frequency of 15 (hit self), the other topographies of self-injury displayed by this participant evidenced a frequency greater than 20. Therefore, the *d* statistic calculations for individual topographies were made for this participant.

Table 5.5. Frequency of each topography of self-injury (frequency per minute in which behaviour was displayed in parentheses) displayed by each participant, summed across all standard, extended and modified experimental functional analyses

Participant	Bite hard object	Bite self	Cheek press	Eye press	Finger bite	Hand bite	Hand to hand hit	Hand to object hit	Head bang	Head hit	Hit self	Leg slap	Mouth hit	Scratch self	Skin pick	Teeth hit	Wrist hit
One	-	-	-	-	-	-	-	32 (0.19)	-	-	-	-	-	-	-	-	-
Two	-	-	-	-	-	-	-	-	-	4 (0.04)	-	60 (0.54)	-	6 (0.05)	-	-	-
Three	-	-	-	-	6 (0.14)	-	-	-	-	-	-	-	13 (0.31)	-	-	-	-
Four	4 (0.27)	5 (0.22)	-	-	-	-	-	12 (0.10)	56 (0.48)	-	-	-	-	-	-	8 (0.07)	-
Five	4 (0.16)	50 (3.06)	-	-	-	-	-	1 (0.01)	9 (0.09)	-	11 (0.11)	-	-	-	-	-	-
Six	-	-	-	-	-	9 (0.44)	-	1 (0.01)	1 (0.01)	-	16 (0.16)	-	-	-	-	-	-
Seven	-	-	-	-	-	-	-	40 (0.23)	-	-	15 (0.09)	-	-	-	31 (1.78)	-	-
Eight	-	-	-	99 (23.71)	-	-	377 (3.16)	-	-	-	21 (0.18)	-	-	-	-	-	29 (0.24)
Nine	-	35 (1.20)	-	-	-	-	-	-	3 (0.03)	-	1 (0.01)	-	-	1 (0.03)	-	-	-
Ten	-	205 (10.34)	37 (1.49)	-	-	-	-	-	-	-	-	-	-	-	-	-	-

5.4 Results

5.4.1 Results from experimental functional analyses

In order to test the hypotheses of the study, the percentage of intervals in which self-injury occurred were plotted for the standard, extended and ASD modified functional analyses, and *d* statistic results were calculated. Figure 5.1 presents these results for each participant.

As can be seen in the figures, the results of the standard and extended functional analyses did not reveal function for any of the participants. These findings support the first hypothesis which predicted that function would not be obtained using standard functional analysis conditions for the majority of participants. However, the findings do not support the second hypothesis, which predicted that for some participants, function would be obtained through the addition of the social escape condition.

Finally, the results of the ASD modified functional analyses support the final hypothesis, that ASD modified functional analyses would reveal function for the majority of participants. The *d* statistic comparing the control and ASD related test conditions was greater than 0.7 for six of the nine participants for whom ASD modified functional analyses were conducted. The flow chart in Figure 5.2 summarises the results from all the participants.

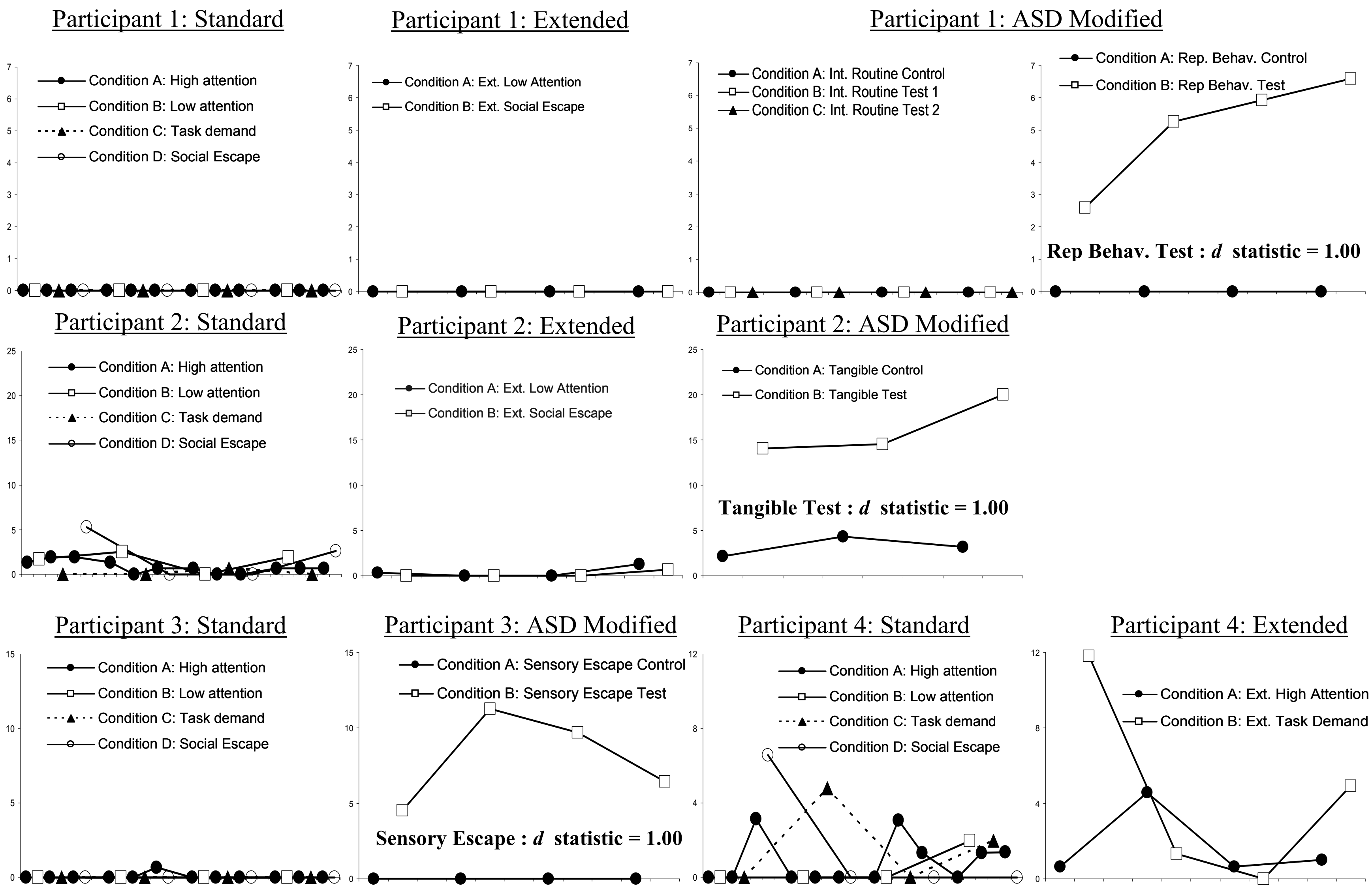


Figure 5.1. Percentage of intervals self-injury occurred in for Participants 1, 2, 3 and 4 in Standard, Extended and ASD modified experimental functional analysis. *d* statistic values > .07 are reported in bold.

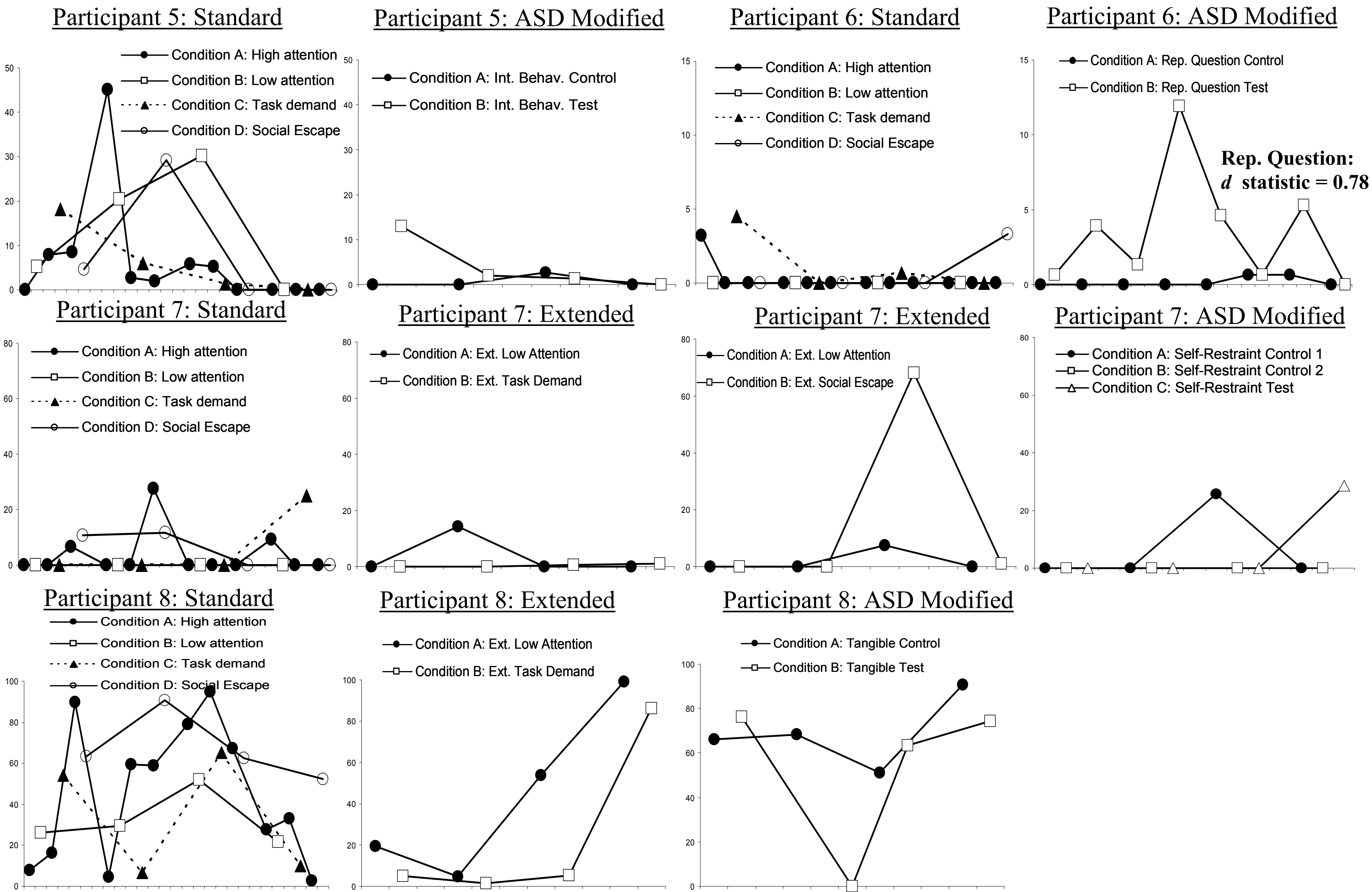


Figure 5.1 cont. Percentage of intervals self-injury occurred in for Participants 5, 6, 7 and 8 in Standard, Extended and ASD modified experimental functional analysis. *d* statistic values > .07 are reported in bold.

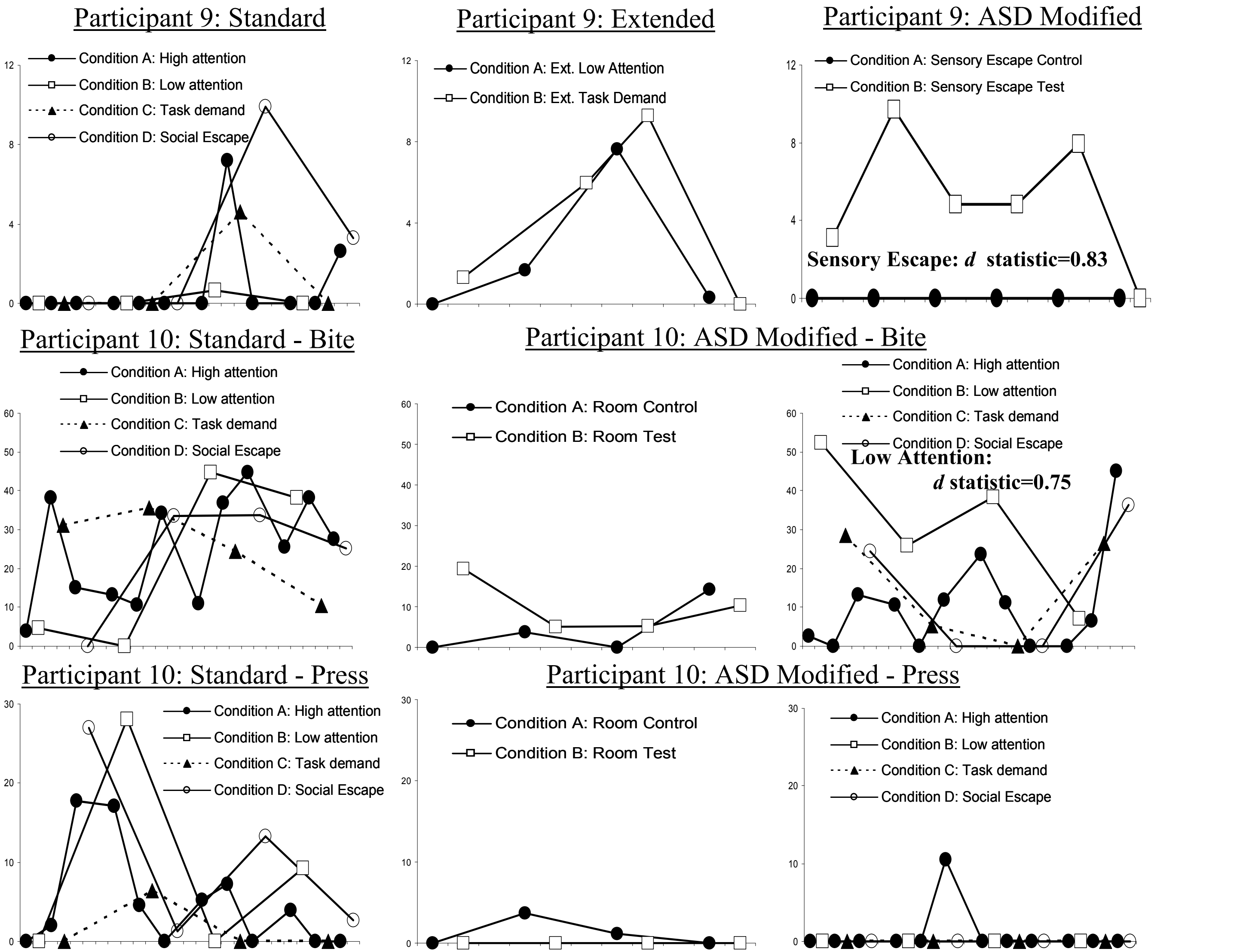


Figure 5.1 cont. Percentage of intervals self-injury occurred in for Participants 9 and 10 in Standard, Extended and ASD modified experimental functional analysis. *d* statistic values > .07 are reported in bold.

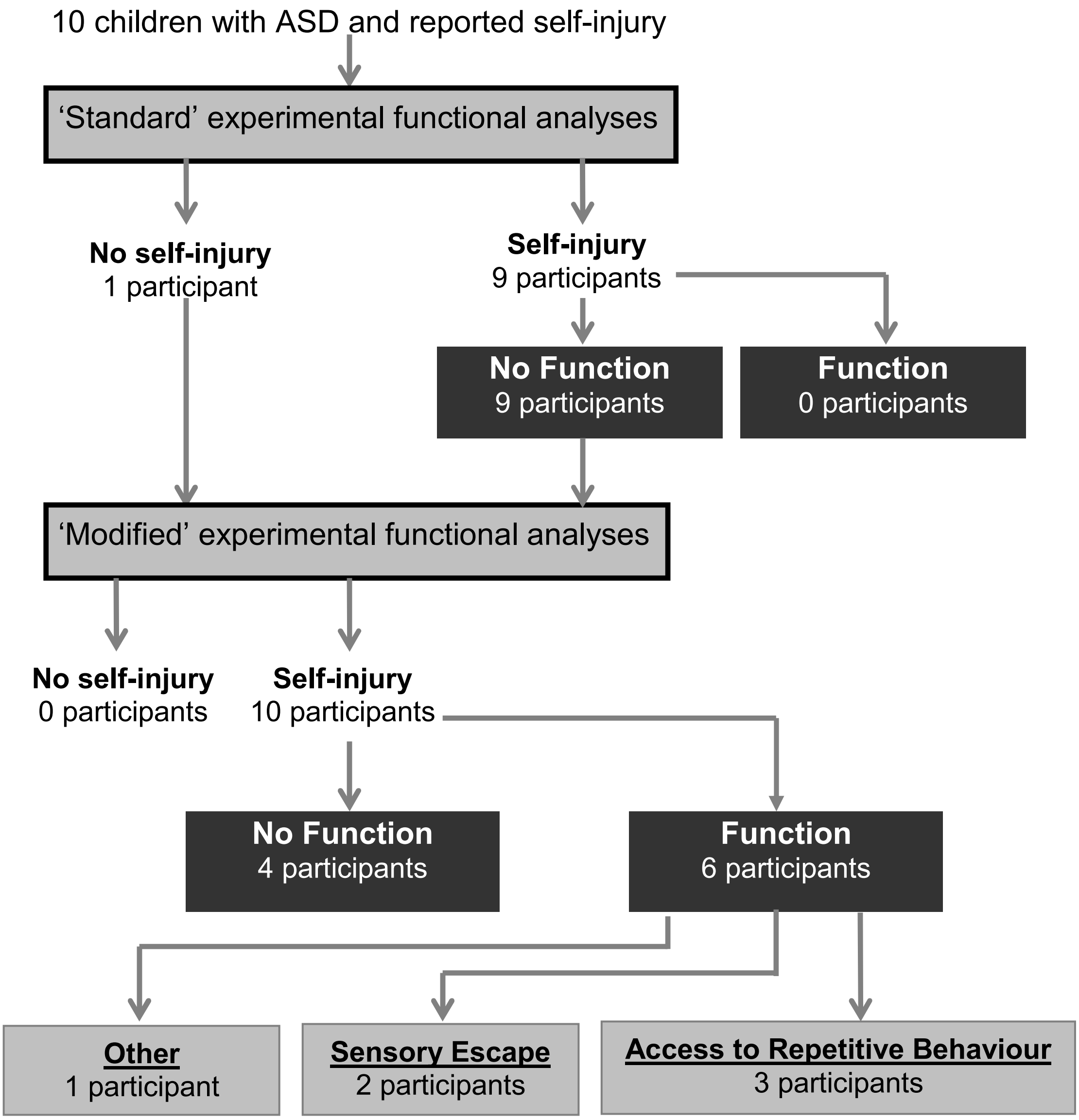


Figure 5.2. Flow chart summarising the outcome of 'standard' and 'modified' experimental functional analyses for all participants.

Figure 5.2 demonstrates that during the 'standard' experimental functional analyses, self-injurious behaviour was displayed by nine participants; however an operant function was not identified for any behaviour. During the 'modified' experimental functional analysis, self-injurious behaviour was displayed by all ten participants, with an operant function identified for the self-injurious behaviour displayed by six of the participants. For two of these participants, the operant function of the self-injury could be broadly categorised as 'sensory escape'. For a further three participants, the operant function could be described as maintaining 'access to repetitive behaviour'.

5.5 **Discussion**

This is the first empirical study to evaluate the function of self-injury in a cohort of children with ASD. The study used robust experimental functional analyses, based on those described by Carr & Durand (1985) and Iwata *et al.* (1994) and extended these by including a social escape condition and by constructing individualised ASD modified functional analyses. All experimental functional analyses were carried out in controlled circumstances, with systematic manipulations of environmental antecedents and consequences and appropriate control conditions. Behaviours were operationally defined, and a high level of inter-rater reliability was achieved. The study improved upon previous research by independently detailing ASD phenomenology and intellectual disability status with instruments with good reliability and validity. This study was the first to employ statistical methods to evaluate functional analysis results for individuals with ASD, and applied this statistical approach to delineate results for individual topographies of self-injury. The results of the study indicate that the most commonly employed functional analyses did not reveal typical social functions for self-injury in children with ASD. However, ASD modified functional analyses did reveal a socially mediated function to self-injury for the majority of the sample.

In line with previous findings, commonly employed functional analysis did not reveal typical demand escape or attention maintained social functions for self-injury for any of the ten children (Hagopian *et al.*, 2007; Hausman *et al.,* 2009; Healey *et al.,* 2001; McKerchar *et al.,* 2001). This finding contrasts significantly with the original results reported by Iwata and colleagues (1994), who identified demand escape and attention maintained functions for 64.4% of their participants with intellectual disability. It is possible that this difference is due to a positive submission and publishing bias whereby cases are only reported and published if functional analyses identify an operant function (see Section 1.3.3.1). The results from the current study indicate that self-injurious behaviour in children with ASD may be less frequently occasioned and reinforced by the antecedents and consequences tested in the commonly employed functional analyses than in the general intellectual disability population. This finding has significant clinical implications, as these commonly employed experimental functional analyses are often viewed as the 'gold standard' assessment for challenging behaviour. This study is now the second study to demonstrate that in a cohort of children with validated ASD diagnosis, these commonly employed assessments do not necessarily reveal a behavioural function to challenging behaviour (O'Reilly *et al.,* 2010). It is plausible that the findings in the current study are due to the shortened condition lengths employed (two and a half minutes, compared to the standard five or ten minute conditions). However, the extended commonly employed functional analysis conditions utilised for six of the participants, also failed to reveal a typical social function. Additionally, the majority of the ASD modified conditions which did reveal social functions were the same length as, or shorter than, the commonly employed functional analyses, suggesting that length of time was not a causal factor in these findings. Given these considerations, further research using a larger sample is required to investigate the possibility that challenging behaviour and, more specifically, self-injury, is associated with escaping demands and gaining attention less frequently in children with ASD than in individuals with intellectual disability of heterogeneous aetiology.

Surprisingly, the addition of the social escape condition did not reveal a social function for self-injury in any of the sample. This finding does not support previous literature (Hagopian, *et al.,* 2001; Tiger *et al.,* 2009) and may suggest that the function of social escape is not as prevalent as hypothesised in children with ASD and self-injury. It is possible that despite their deficits in social interaction, children with ASD may still be motivated to engage in social interaction, or at least do not find it aversive enough to occasion self-injury (Wing & Gould, 1979). Further delineation of social motivation versus social competency and the interactions between these characteristics and enforced social situations may shed further light on the role of social escape as a maintaining factor in self-injury for individuals with ASD. However, a number of methodological issues may also account for this finding. First, in order to allow comparisons to previous research, the commonly employed functional analyses in this study employed a 'high attention' condition as a control. It is plausible that through the high attention conditions, the repeated presentation of the antecedent (social interaction) without the presentation of the consequence contingent upon self-injury (removal of social interaction), led to extinction prior to the social escape condition being conducted. Secondly, it is possible that the children did not distinguish between the high attention and social escape conditions. However, as the two conditions were run by two different researchers, the likelihood of this was minimised. Finally, it is possible that the aversive property of social interaction was associated with the quantity of time that social interaction was presented for. Specifically, that the first researcher, who conducted the high attention and demand conditions, was engaged in social interaction with the participant for 30 minutes across the commonly employed functional analyses, whereas the second researcher who conducted the social escape condition, was only engaged in social interaction for two and a half minutes. If motivation to escape social interaction with a specific person increases proportionally to the amount of time spent engaging in social interaction, then the utilisation of the second researcher to run the social escape condition actually provided the participant with a break from an increasingly aversive social interaction. These issues require further research, using experimental designs constructed to overcome the problems highlighted here.

The most striking finding from this study is the identification of social functions to self-injurious behaviour for six of the participants, when ASD related antecedents and consequences were programmed into the functional analyses. Similarly to previous findings, two of the participants (Three and Nine) demonstrated sensory escape functions to their self-injurious behaviour (Buckley & Newchock, 2006; Devlin *et al.,* 2008). These findings support the hypothesis that sensory escape functions may be common in individuals with ASD. In line with previous research, two further participants demonstrated self-injurious behaviour that functioned to reinstate a repetitive behaviour (Participant One; Hausman *et al.,* 2009; Murphy *et al.,* 2000) or to gain access to a material involved in a repetitive behaviour (Participant Two; Reese *et al.,* 2003; 2005). Additionally, one participant (Participant Six) displayed self-injurious behaviour that functioned to gain a preferred response to repetitive questioning. The prevalence of self-injury associated with sensory escape and repetitive behaviours in this sample does lend tentative support to a hypothesis of ASD related functions and more broadly phenotype x environment interactions. In these interactions, reinforcement for self-injury interacts with core features of ASD phenomenology, in this case sensory sensitivity and a preference for routines. Finally, social function was obtained for one topography of Participant Ten's self-injury. The self-injury was demonstrated to be maintained by attention, but *only* in a specific location. This finding suggests an interaction between location and social antecedents which requires further investigation (Lang, Sigafoos, Lancioni, Didden & Rispoli, 2010).

Taken together, these findings confirm the presence of ASD related social functions for self-injury in children with ASD. Building upon the hypothesis of gene or phenotype x environment interactions (Langthorne & McGill, 2008; Oliver, 1993; Tunnicliffe & Oliver, 2011), these results may indicate that the elevated prevalence of self-injury in individuals with ASD results from an interaction between ASD characteristics such a preference for routine, repetitive behaviour and sensory sensitivity and the environment. These behavioural characteristics in the form of a behavioural phenotype, would elevate the probability of frequently occurring events and stimuli being reinforcing to individuals with ASD, but not to other individuals with intellectual disabilities of heterogeneous aetiology. This may account for the heightened prevalence of self-injury in individuals with ASD. However, whilst these findings provide preliminary support for this hypothesis, further experimental evidence, contrasting individuals with ASD with other participant groups is still required.

For Participants Four, Five, Seven and Eight, no social function was identified, and therefore automatic reinforcement may be assumed. However, a series of issues must be considered for two of the participants. Participant Four exhibited very high levels of distress when engaging in self-injury, and neither parents nor teachers could identify reliable triggers to his self-injury. Consequently, no ASD related functional analyses could be undertaken. Anecdotally carers reported that his self-injury seemed to "come out of nowhere", and was reported to be unrelated to his immediate environment. The results of the standard and extended functional analyses confirm this. Additionally a number of pain related behaviours were displayed by Participant Four, in close temporal proximity to self-injury. Following identification of this, Participant Four was treated for Gastroesophageal Reflux, and carers reported significant reduction in his self-injurious behaviour (Luzzani, Macchini, Valade, Milani & Selicorni, 2003). Unfortunately, within the constraints of this study it was not possible to confirm this change (see Section 1.3.3.3 for a further discussion of pain related self-injury). Participant Eight displayed very high levels of self-restraint behaviour, and therefore it was hypothesised that his self-injury was influenced by compromised behavioural inhibition (see Chapter 5 for a discussion of self-restraint and behavioural inhibition). Although in this case automatic reinforcement would be assumed, treatment options may be effective if they were focused upon increasing capacity for inhibitory control, rather than increasing enrichment in the environment. Consequently, even when commonly employed and ASD related functional analyses fail to demonstrate a social function, an assumption of automatic reinforcement may not be warranted. Further experimental investigation should follow from these anecdotal reports, to investigate the effect of pain upon self-injury in ASD and the resulting effects of self-restraint upon social function of self-injury.

There are a number of limitations to the findings presented in this study. Firstly, the order of conditions in the standard functional analyses was not randomised. Whilst this may have resulted in order and carry-over effects, this structure was chosen in order to ensure that all test conditions started from the same baseline. By preceding each test condition with a control condition, it was possible to ensure that establishing operations between test conditions were consistent. Secondly, as with all experimental functional analyses, it is not possible to identify from these results alone the proportion of naturally occurring self-injury that is accounted for by the ASD related functions identified in experimental settings. However, this preliminary study does provide proof of principle that it is possible to progress from undifferentiated functional analyses to identify ASD related functions for self-injury. Future work may usefully continue by assessing how frequently self-injury occurs in the natural environment, occasioned and reinforced by identified ASD related antecedents and consequences. Finally, it was beyond the remit of this study to conduct interventions derived from the functional analysis assessments. Future studies should seek to build upon these findings by including an intervention component in order to further validate the results of ASD related functions to self-injury.

Clinically, the findings of this study have significant implications. In accordance with the results of previous single case studies in individuals with ASD, several of the children displayed very low levels of self-injury across conditions (Participants One, Two, Three and Six; Hagopian *et al.,* 2007; Hausman *et al.,* 2009). Without the inclusion of the ASD modified functional analyses, it would be assumed wrongly that these children did not show self-injury in response to environmental antecedents. Likewise, several of the children displayed high, undifferentiated levels of self-injury across the conditions (Participants Nine and Ten; Healey *et al.,* 2001; McKerchar *et al.,* 2001). In clinical practice and in previous studies investigating challenging behaviour in ASD, these high levels of undifferentiated behaviour have been interpreted as evidence for automatic reinforcement (O'Reilly *et al.,* 2010), and the possibility of a social function to the behaviour has been disregarded. Although it is likely that automatic reinforcement accounts for some of the self-injury in these participants, this study has demonstrated that there is also a social function to their self-injury, for which behavioural interventions could be employed. These results highlight the importance of progressing beyond the commonly employed functional analysis for self-injurious behaviour in children with ASD.

The results of this study demonstrate proof of principle; that it is possible to identify a social function to self-injury for children with ASD through the employment of ASD related functional analyses. It has been hypothesised that these ASD related functions may be more likely in individuals with ASD than in individuals with intellectual disability, due to an interaction between the behavioural phenotype of ASD (with a preference for routine, repetitive behaviour and sensory sensitivity) and the environment. If this were the case, a key implication for clinical practise would be to build a tailored set of assessments for self-injury for individuals with ASD. However, this hypothesis requires further research in order to be tested. To extend the findings of this study and test this hypothesis, a replication study could be undertaken, with a larger sample size and wider age range of participants. In order to attribute these hypothesised 'ASD related functions' to an ASD population, the study would also require a matched comparison group of individuals with comparable levels of intellectual disability and no ASD diagnosis. All participants would receive the same commonly employed experimental functional analysis assessments, ideally including both a 'standard' component, testing demand escape and attention maintained functions, and an 'ASD' component, testing functions of sensory escape and avoidance of interruption to routine/ repetitive behaviour. The inclusion of a post-assessment intervention and treatment evaluation would add further validity to the findings. From the results of this study, and previous research (Reese *et al.*, 2003; 2005) it is hypothesised that the ASD related functions would occur significantly more frequently in the ASD population, and the 'standard' functions would occur significantly more frequently in the intellectual disability population.

In summary, this study demonstrated that, through the utilisation of ASD related experimental functional analyses, it was possible to identify social function for the majority of the children with ASD. This finding was discussed in relation to the possibility of ASD related functions.

CHAPTER 6

Temporal Associations between Self-Injurious Behaviour, Repetitive Behaviour, Challenging Behaviour, Proto-Imperative Communication and Self-restraint in Children with Autism Spectrum Disorder

---

6.1 Preface

The study in Chapter 5 evaluated one corollary of operant theory; that behaviour will systematically fluctuate across environments dependent upon reinforcement contingences. The study used experimental functional analysis methodologies to evaluate the function of self-injury in children with ASD. The results indicated that it was possible to ascertain a social function for self-injury for six of the ten participants, through the utilisation of functional analyses modified to identify ASD related antecedents. A second corollary of operant theory indicates that self-injury will occur within response classes within an individual's behavioural repertoire (see Sections 1.3.3.1 & 1.5.2). Therefore, in order to further explore the utility of operant theory in delineating self-injury in ASD, the present study presents a fine grained analysis of the behaviours displayed by the same sample of children during the experimental functional analyses. Specifically, temporal associations between self-injury and repetitive behaviours, proto-imperative communication, self-restraint and challenging behaviours will be explored, in order to evaluate the organisation of other behaviours relative to self-injury in ASD.

6.2 Introduction

The prevalence of self-injury in individuals with ASD has been robustly estimated at between 40 and 50%, which is significantly higher than the prevalence of self-injury in individuals with intellectual disability of heterogeneous aetiology (See Sections 1.4.1, 2.4.1, 3.4.2 & 4.4.2). Operant theory has been usefully employed to account for the development and maintenance of self-injury (see Section 1.3.3.1). One corollary of operant theory, demonstrated in Chapter 5, is that self-injury will systematically fluctuate across environmental conditions (see Section 5.4.1). A second corollary indicates that due to environmental learning, behaviour may become clustered within an individual's repertoire and that these clusters of behaviour can be temporally organised.

Temporal associations between self-injury and other behaviours have been investigated previously for both theoretical and pragmatic purposes. Investigation of how behaviour is organised within an individual's repertoire has led to the delineation of response classes, where temporally proximal behaviours group into functionally equivalent classes (Baer, 1982). By definition, each behaviour in the class is evoked by the same establishing operation and maintained by the same reinforcement contingencies. Behaviours within a response class may be organised in a hierarchy in which the initial response requires the least effort but is also the least likely to receive the required consequence. Pragmatically, this has led to the evaluation of precursors to severe self-injury, such as repetitive behaviour, communicative behaviour and lower intensity challenging behaviour (Borrero & Borrero 2008). Researchers have demonstrated that assessment and intervention for these precursor behaviours can reduce self-injury, without the need to evoke self-injury (Hagopian, Paclawskyj & Kuhn, 2005; Langdon, Carr & Owen-DeSchryver, 2008; Smith & Churchill, 2002). These findings have significant implications for the ethical dilemmas often identified in assessment and intervention for self-injury. However, little research has investigated the temporal associations and behavioural organisation of self-injury and co-occurring behaviours in ASD. More specifically, an evaluation of the temporal associations between self-injury and challenging behaviour, repetitive behaviours, proto-imperatives and self-restraint may have significant implications for both theoretical and pragmatic models of self-injury in ASD.

Self-injurious behaviour has been conceptualised in some theories as a stereotypic motor disorder rather than a learnt, operant behaviour maintained by environmental contingencies (Bodfish *et al.,* 1995; King, 1993). Within this model, self-injury is broadly perceived as purposeless and unrelated to environmental influences. This conceptualisation of self-injury can lead to therapeutic nihilism and an avoidance of behavioural interventions commonly employed when behaviour is determined to be maintained by environmental contingencies. This may be particularly important for individuals with ASD, where repetitive motor behaviours are significantly more common than in the intellectual disability population (Turner, 1999). Consequently, within populations with ASD, self-injury may be more likely to be perceived as an additional repetitive behaviour with no operant function. One source of evidence indicating that self-injurious behaviour is not simply a motor disorder is the close temporal association between self-injury and other challenging behaviours such as aggression and property destruction. Petty, Allen and Oliver (2009) demonstrated that for a subset of four children who displayed both self-injury and other challenging behaviours, other challenging behaviours were significant precursors to self-injury. This temporal clustering of behaviours suggests that self-injury is not simply a motor disorder, but rather a member of a response class, which an individual displays contingent upon environmental antecedents.

Similarly, Smith and Churchill (2002) demonstrated that an aggressive behaviour (grabbing others) displayed by an individual was maintained by the same environmental contingencies which maintained the individual's self-injurious behaviour, supporting the concept of aggressive and self-injurious behaviours forming a functionally equivalent response class. There is limited evidence for temporal associations between self-injurious behaviour and other challenging behaviour in individuals with ASD. One of the cohort of four children reported in the study by Petty *et al.* (2009) was previously diagnosed with ASD, however, this diagnosis was not confirmed within the study. Consequently, further evidence from a sample diagnosed with ASD is required to examine the possibility of self-injury as temporally associated to other challenging behaviours and thus potentially a member of a functionally equivalent response class.

Temporal associations between self-injury and repetitive behaviour may help to provide a more comprehensive understanding of the developmental progression of self-injury in ASD. The findings reported in Chapters 2, 3 and 4 revealed that repetitive behaviours were associated with the presence of self-injury (see Sections 2.4.4, 3.4.3 & 4.4.4) and that repetitive behaviours were independently predictive of self-injury in children with ASD and severe self-injury in adults with ASD (see Section 4.4.6). Importantly, Guess and Carr (1991) proposed that self-injury initially evolves from internally regulated repetitive behaviours that are present in typical development, but persist longer when developmental delay occurs. Guess and Carr (1991) hypothesise that repetitive behaviours then begin to function to maintain an optimal level of stimulation for the individual through a process of social and non-social reinforcement, and then transform into self-injury which is shaped and maintained by social operant contingencies. This model takes on particular significance in individuals with ASD who exhibit elevated levels of repetitive behaviour (Estes *et al.,* 2011; Richler, Bishop, Kleinke & Lord, 2007; Turner, 1999). If the model proposed by Guess and Carr (1991) is valid, then the raised prevalence of self-injury in ASD may be causally related to the increased prevalence of repetitive behaviours in the population. The elevated presence of repetitive behaviours may provide more opportunities for the behaviours to be shaped by social reinforcement into self-injury. However, there is little direct longitudinal evidence for transitions between the stages of the model proposed by Guess and Carr (1991).

Hall, Thorns and Oliver (2003) demonstrated that repetitive behaviours were sensitive to non-social and social reinforcement, allowing for the possibility of reinforcement shaping repetitive behaviours to self-injury. Additionally, Richman and Lindauer (2005) demonstrated the progression from a stereotyped behaviour to a functionally equivalent self-injurious behaviour over time. However, this pattern was only evident in one child, for one topography of behaviour, from a total cohort of 12 children. The paucity of longitudinal evidence for the transitions between repetitive behaviour and self-injury may in part be due to the inherent methodological problems in conducting longitudinal studies, of recruiting and maintaining a sample over time. These problems are particularly salient when the focus of the research is to identify a transition to a behaviour that has not yet emerged in the population, necessitating a large sample in order to ensure that the behaviour will occur in some cases.

Petty *et al.,* (2009) proposed an alternative method through which the model proposed by Guess and Carr (1991) could be evaluated prior to conducting more extensive longitudinal studies. Superstitious reinforcement has been hypothesised as one method by which transition between the stages of Guess and Carr's model may occur. Superstitious reinforcement occurs when behaviours occurring in close temporal proximity to the original reinforced behaviour, are inadvertently reinforced and then gain the function of the original behaviour (Kennedy, 2002; Oliver, 1993; 1995). Based upon this model, Petty *et al.* (2009) hypothesise that if the Guess and Carr transitional model is correct, then temporal associations between repetitive behaviour, proto-imperatives and self-injury would exist within an individual's behavioural repertoire, as evidence of how the behaviours became socially maintained. Utilising lag sequential analyses of behaviours exhibited by a cohort of children during experimental functional analysis and natural observations, Petty *et al.* (2009) demonstrated that for four of the six children, repetitive behaviours were significant immediate precursors to self-injury. Importantly, two of the children in this study had a diagnosis of ASD and for both of these children repetitive behaviours were significant temporal precursors to self-injury.

In addition to this cohort study, several single case studies have reported reliable repetitive behaviour as immediate precursors to self-injury, such as stereotyped waving of hands in front of the face, hand clasping, hand posturing and jerky body movements. (Hagopian *et al.,* 2005; Herscovitch, Roscoe, Libby, Bourret and Ahearn, 2009; Langdon, *et al.,* 2008). Interestingly, the studies conducted by Herscovitch *et al.* (2009) and Langdon *et al.* (2008) included individuals with ASD, supporting a hypothesis that repetitive behaviours may be precursors to self-injury in ASD populations. The reliable temporal association between repetitive behaviours and self-injury in ASD provides preliminary support for the hypothesis that self-injury evolves from repetitive behaviours through a process of reinforcement. However, the evidence thus far has been drawn from single case studies, or studies in which ASD diagnosis has not been independently confirmed. Additionally, the studies by Herscovitch *et al.* (2009) and Langdon *et al.* (2008) used informant report to establish precursor behaviours prior to experimental observations and only conducted analysis for the pre-selected precursor behaviours. Therefore, it is not possible to more broadly establish the temporal associations between repetitive behaviours and self-injurious behaviour in ASD. Finally, only the study conducted by Petty *et al* (2009) conducted statistical analyses in order to confirm that the probability of repetitive behaviour was significantly higher prior to self-injury, than it was at other times. Consequently, there is a need to investigate statistically the naturally occurring temporal association between repetitive behaviours and self-injury in a cohort of individuals with ASD, where an assessment of ASD diagnosis is also conducted.

In addition to investigating theoretical models of self-injurious behaviour, an evaluation of behaviours temporally associated to self-injury can also have pragmatic value. Interventions for self-injury have successfully used functional communication training (FCT; Carr & Durand, 1985), in which an individual is taught a communicative behaviour which is functionally equivalent to their self-injurious behaviour. This behaviour is then reinforced with more immediacy, magnitude and consistency than the self-injurious behaviour (Emerson, 1998; Horner & Day, 1991). As the new functionally equivalent communicative act increases in efficiency, it replaces the self-injurious behaviour in the individual's behavioural repertoire. Proto-imperative behaviours are pragmatic, functional communicative behaviours, such as negative vocalisations and approach or dissent behaviours, which influence a child's environment (Petty *et al.,* 2009). The presence of proto-imperatives in a child's behavioural repertoire, in close temporal proximity to self-injurious behaviour has two important implications for interventions such as FCT. Firstly, if the proto-imperative consistently precedes self-injurious behaviour, then the proto-imperative behaviour can highlight to others the point in the response chain at which a functionally equivalent response would be most effective. Secondly, given the communicative nature of a proto-imperative act, the proto-imperative behaviour itself can be shaped and utilised to be the functionally equivalent response which is reinforced and selected over self-injurious behaviour.

In support of this, Smith and Churchill (2002) demonstrated that proto-imperatives such as reaching for another person, screaming, crying and vocalisations, were maintained by the same contingencies as the problem behaviour which they preceded. They also demonstrated that reinforcement of the proto-imperative behaviour resulted in decreases in the problem behaviour. In a single case study of a child with ASD, Herscovitch *et al.* (2009) demonstrated that loud vocalisations were present in a response class of low intensity self-injurious behaviours which preceded higher severity self-injury. Additionally, proto-imperatives were identified as precursors to self-injury for the two children with ASD included in the cohort described by Petty *et al.* (2009). However, these studies did not conduct an assessment of ASD phenomenology and consequently there is limited evidence regarding the prevalence of naturally occurring proto-imperatives in immediate temporal proximity to self-injury in individuals with ASD. Additionally, all of the studies were conducted with individuals where a social function for self-injury was indentified and therefore there are currently no data regarding the presence and organisation of proto-imperatives around ‘non-functional’ self-injurious behaviour. Given the direct implications for intervention, further research is required to delineate the association between proto-imperative communication and self-injurious behaviour in a cohort of individuals with confirmed ASD diagnosis.

A final behaviour of interest in relation to the presence of self-injury is self-restraint. Self restraint behaviours 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 behaviours are associated with the presence of self-injurious behaviour (Fovel, Lash, Barron & Roberts, 1989; Hyman, Oliver & Hall, 2002) and occur at a high prevalence in individuals who self-injure (46% to 76 %; Oliver, Murphy, Hall, Arron & Leggett, 2003; Powell, Bodfish, Parker, Crawford & Lewis, 1996).

The findings in Chapter 4 revealed a prevalence of self-restraint in children with ASD of 40.9% and 52.6% in adults with ASD (see Section 4.4.3). Similarly to previous research, the prevalence of self-restraint was significantly higher in children and adults who engaged in self-injury (56.8% and 57.5% respectively) revealing a robust association between self-injury and self-restraint. Importantly, the study also demonstrated a significant potential causal contribution of repetitive and overactive and impulsive behaviours to the presence of self-injury and self-restraint (see Section 4.4.4). These associations between self-injury, self-restraint and behavioural indicators of impaired inhibition lend support to the hypothesis of self-injury as a 'compulsive' behaviour, occurring beyond an individual's control (King, 1993; see Sections 4.2 & 4.5 for an extended discussion of this hypothesis). Within this hypothesis, self-restraint behaviours are displayed by an individual in an attempt to inhibit 'compulsive'[1] self-injury. This hypothesis is supported by physiological evidence which demonstrated elevated heart rate in an individual when restraint was unavailable, and resting heart rate when restraint was allowed (Jennett, Hagopian & Beaulieu, 2011).

However, despite the strong association between self-injurious behaviour and self-restraint and the hypothesised association with impaired behavioural inhibition, the temporal

[1] This term is used to represent behaviours which are difficult to inhibit and impulsive in nature, rather than compulsive behaviours which compulsions function to relieve anxiety or prevent a feared consequence.

association between self-injury and self-restraint has been largely ignored in research. One study that did investigate this association was conducted by Forman, Hall and Oliver (2002). Through lag sequential analysis, they demonstrated that self-injury increased significantly prior to self-restraint, occurred at near zero levels during self-restraint and increased again following self-restraint. This finding may support the theory of self-restraint as mechanism to inhibit 'compulsive' self-injury, with self-restraint functioning to stop the self-injurious behaviour once it reached a critical level. However, this evidence was demonstrated in a single case study and no replication of these findings has been conducted. Furthermore, no investigation of the temporal association between self-restraint and self-injury has been conducted in an ASD population. Consequently, preliminary investigation of the temporal association between self-injury and self-restraint, in a cohort with ASD is necessary in order to understand the function of self-restraint in relation to self-injury and in order to hypothesise further about the association with impaired behavioural inhibition.

The studies highlighted above present preliminary evidence for a temporal association between self-injury and 1) other challenging behaviours (Petty *et al.,* 2009; Smith & Churchill, 2002), 2) repetitive behaviour (Hagopian *et al.,* 2005; Herscovitch *et al.,* 2009; Langdon *et al.,* 2008; Petty *et al.,* 2009), 3) proto-imperatives (Herscovitch *et al.,* 2009; Petty *et al.,* 2009) and 4) self-restraint (Forman, Hall & Oliver, 2002). A limited selection of this evidence has been evaluated in ASD populations (Herscovitch *et al.,* 2009; Langdon *et al.,* 2008; Petty *et al.,* 2009). However, the majority of the evidence is reported in single case studies, where ASD diagnosis is not assessed independently. Additionally, in many of the studies investigating precursor behaviours, carer report was utilised in order to identify participants showing precursors and to identify specific behaviours which preceded self-injury

(Herscovitch *et al.,* 2009; Langdon *et al.,* 2008; Smith and Churchill, 2002). This approach prevents the research from being 'data-led', resulting in a limited number of behaviours being investigated, in a pre-selected sample known to evidence the behaviours. Within these studies, visual inspection of the probabilistic associations between behaviours has been utilised (Hagopian *et al.,* 2005; Langdon *et al.,* 2008; Smith & Churchill, 2002) and all participants included in the studies demonstrated self-injurious behaviour maintained by social reinforcement. Therefore, further research is necessary to investigate behaviours temporally associated with self-injury in a cohort of individuals with ASD. Whilst functional equivalence can not be assumed from simply evaluating temporal associations, an evaluation of these the temporal relationships will still provide data useful for theoretical and pragmatic inferences. Temporal associations between behaviours should be investigated without a-priori confirmation of their proximity to self-injury. Additionally, robust statistical analysis is required in order to evaluate the strength of the temporal associations between self-injury and other behaviours.

Given the paucity of research regarding behaviours temporally associated with self-injury in ASD populations, no specific hypotheses can be stated for this study. Rather, the study has the aim of examining the temporal associations between self-injury and: repetitive behaviours, proto-imperative behaviours, challenging behaviours and self-restraint in a cohort of children with ASD.

6.3 Methods

6.3.1 Recruitment, Participants and Procedure

The present study used the same participants and procedure as those reported in Chapter 5. Refer to Sections 5.3.1 for recruitment, Section 5.3.2 and Table 5.1 for participant information, Section 5.3.3 for the measures administered and Section 5.3.4.1 for the data collection procedure.

6.3.2 Data Coding and Analysis

6.3.2.1 Data Coding

Behaviour in all standard, extended and ASD modified functional analyses were coded for each child. Child behaviours were coded using real time Obswin software (Martin, Oliver & Hall, 2000). Some forms of behaviour were common across children and some topographies were specific to individual children. Behaviours were either coded as duration variables (e.g., approach), where the behaviour onset and offset was recorded, or as event variables, where only the behaviour occurrence was recorded (e.g., head hit). In order to evaluate the temporal associations between classes of behaviour, the individual behaviours were grouped into self-injurious behaviour, proto-imperatives, repetitive behaviour, challenging behaviour and self-restraint. Operational definitions for each behaviour and a list of participants who were recorded to show this behaviour can be seen in Table 6.1. Inter-observer reliability using 3 second time intervals was calculated for 25% of all video recorded sessions. Mean Cohen's Kappa values across all behaviours was .84 (range 0.47 – 1.00) illustrating good agreement (Fleiss, 1981).

Table 6.1. Coded behaviours, operational definitions, participants who displayed this behaviour and Kappa values

	Behaviour	Definition	Kappa	Participants
Self-Injurious Behaviour Mean Kappa = .87	Bite hard object	Child clenches jaw and teeth around a solid object (e.g., wood, chair, table). Must use top and bottom teeth and definite biting movement.	.82	4,5,
	Bite self	Child clenches jaw and teeth around own body part	.87	4,5,9,10
	Cheek press	Child forcefully pushes thumbs into cheek	.92	10
	Eye Press	Child pushes fingers on eye, pulls or pokes eyelashes	.97	8,
	Finger bite	Child clenches jaw and teeth on side of finger	1.00	3,
	Hand bite	Child clenches jaw and teeth on hands or arms	.67	6,
	Hand to hand hit	Child forcefully brings hands together, palm to palm	.81	8,
	Hand to object hit	Child forcefully makes contact with hard surface with palm of hand	.47	1,4,5,6,
	Head bang	Child forcefully makes contact with head against hard surface	.80	4,5,6,9,
	Head hit	Child forcefully makes contact with fist and/or open palm on their head	1.00	2,7,
	Hit self	Child forcefully makes contact with fist and/or open palm on own body part	.80	5,6,7,8,9
	Leg slap	Child forcefully makes contact on leg with palm of hand	.79	2,
	Mouth hit	Child forcefully makes contact on mouth/around mouth with palm of hand	1.00	3,
	Scratch self	Child drags nails across or pinches their own body part	1.00	2,9
	Skin pick	Child scrapes skin using forefinger at right angles to the surface of skin	.89	7,
	Wrist hit	Child forcefully makes contact with top teeth against object	1.00	8,
	Teeth hit	Child forcefully makes wrist to wrist contact	1.00	4,

Table 6.1 cont. Coded behaviours, operational definitions, participants who displayed this behaviour and Kappa values.

	Behaviour	Definition	Kappa	Participants
Proto-Imperative Behaviour Mean Kappa = .73	Approach	Child physically comes towards examiner, tugs at clothing, attempts to gain attention	.74	1,2,4,5,6,7,8,9,10
	Look towards examiner	Child orients head and eyes towards adult - this does not need to necessitate adult eye contact, simply that the child has looked towards the adult	.70	1,2,3,4,5,6,7,8,9,10
	Neutral vocalisation	Child makes any noise that has no clear affect - this does not include recognisable speech	.67	1,2,3,4,5,6,7,8,9,10
	Negative vocalisation	Child makes any noise that has clear negative affect – this includes crying, whining and protesting	.80	1,2,3,4,5,6,8,9,10
	Dissent	Child physically moves away from examiner or task – this includes attempts to leave room	.76	1,2,3,4,5,6,7,8,9,10
Challenging Behaviour Mean Kappa = .91	Grab examiner	Child reaches for and briefly holds onto the examiner's body or clothes	.89	1,7,9
	Kick examiner	Child strikes foot out, making contact with examiner	1.00	1,
	Pull examiner	Child holds onto examiner's body or clothes, moving them towards themselves	.80	2,5,
	Push examiner	Child reaches for examiner's body or clothes, makes contact and moves them away from themselves	.90	1,3,10
	Hit examiner	Child quickly strikes examiner with hand	1.00	7,
	Kick object	Child quickly strikes object with foot	.75	4,5,
	Push object	Child reaches for object and moves it quickly away from themselves	.91	4,5,7,
	Throw object	Child picks up object and pitches it away from themselves	1.00	4,5,

Table 6.1 cont. Coded behaviours, operational definitions, participants who displayed this behaviour and Kappa values.

	Behaviour	Definition	Kappa	Participants
Repetitive Behaviour Mean Kappa = .81	Body stereotypy	A non contact movement in which the child rocks, twists or holds body unusually	.87	1,8,9,10
	Hand stereotypy	A non contact movement in which the child twists or holds hand unusually	.74	1,2,3,5,6,7,9
	Hand to object stereotypy	A contact movement in which the child twists or holds hand unusually, making contact with an object	.72	4,
	Head stereotypy	A non contact movement in which the child rocks, twists or holds head unusually	.84	10
	Mouth stereotypy	A non contact movement in which the child opens, closes or poses mouth unusually	.73	1,7,9,
	Hand to eye stereotypy	A contact movement in which the child twists or places their hand by their eye unusually	.93	4,
	Saliva stereotypy	A non contact movement in which the child spits onto objects or hands, plays with and/or peers as saliva	.94	5,
	Hand to face stereotypy	A contact movement in which the child rubs their face with their hand in a repetitive motion	.71	2,8,
	Echolalia	The child immediately repeats a word or phrase spoken by the examiner	.80	6,9,
	Pick mouth	A contact movement in which the child repeatedly rubs at the inside of the mouth with their finger	.59	6,
	Skin rub	A contact movement in which the child repeatedly touches their skin with their finger	.96	7,
	Wipe mouth	A contact movement in which the child repeatedly rubs at the outside of the mouth with their finger, arm or tongue	.89	3,

Table 6.1 cont. Coded behaviours, operational definitions, participants who displayed this behaviour and Kappa values.

	Behaviour	**Definition**	**Kappa**	**Participants**
Self-Restraint Behaviour Mean Kappa = .79	Wrapping hands in clothes	Child puts hands inside of clothes, preventing movement	.86	6,7,
	Sitting on hands	Child places hands under own body, preventing movement	.88	6,7,
	Holding hands in hands	Child clasps own hands together, preventing movement	.53	6,7,8,
	Seeking restraint from others	Child offers hands to examiner, seeking hand holding, or placing hands under examiner’s legs or body, preventing movement	.88	6,7,

6.3.2.2 Data Analysis

In order to evaluate the temporal association between self-injury and proto-imperatives, challenging behaviour, repetitive behaviour and self-restraint, lag sequential analyses were conducted. These analyses consider the unconditional probability of the participant engaging in a target behaviour (proto-imperative, challenging behaviour, repetitive behaviour or self-restraint) against the conditional probability of the participant engaging in the target behaviour *given* that they are engaging in the criterion behaviour (e.g. displaying a proto-imperative, given that they then show self-injurious behaviour[2]). For all analyses, lags were examined for ten, three second intervals prior to and after the presence of the target behaviour. Lag zero indicates the epoch during which the self-injurious behaviour was displayed. The lag sequential analyses were *unrestricted* (i.e., the lag was conducted for 30 seconds prior to and after the occurrence of the target behaviour, even if another occurrence of the target behaviour was identified in that time period). Tests were conducted to identify differences between the unconditional and conditional probabilities. Given the large number of tests being conducted, a more stringent alpha levels of $p < .001$ was utilised, consequently a significant degree of difference between the unconditional and conditional probabilities is shown by a z score greater than 3.10.

For each participant the total frequency of each behaviour and the frequency per minute was calculated prior to conducting the lag sequential analyses. Table 6.2 displays the results of this. In order to ensure that there were sufficient data to conduct the lag sequential analyses,

[2] For participant ten, the two forms of self-injurious behaviour displayed (bite self and cheek press) were evaluated separately utilising separate lag analyses. This was due to different operant functions for these two behaviours being revealed in previous analysis (see Section 5.4.3). As no differences in function for topographies of self-injury were identified for any other participants, their topographies of self-injury were combined.

any criterion behaviours which occurred with a frequency of less than 10 were excluded from the analyses.

Table 6.2 Frequency of behaviour (frequency per minute in parentheses) for self-injury, proto-imperatives, challenging behaviour, repetitive behaviour and self-restraint displayed by each participant

	Criterion Variable	**Target Variables**			
Participant	**Self-Injurious Behaviour**	**Proto-Imperatives**	**Challenging Behaviour**	**Repetitive Behaviour**	**Self-Restraint**
One	32 (0.19)	669 (9.19)	115 (0.67)	265 (1.54)	0* (0)
Two	70 (0.77)	428 (18.49)	6* (0.20)	438 (3.97)	0* (0)
Three	19 (0.29)	183 (13.10)	9* (0.30)	136 (15.02)	0* (0)
Four	81 (1.12)	430 (22.48)	123 (1.05)	139 (13.25)	0* (0)
Five	71 (3.35)	425 (19.50)	38 (0.37)	70 (7.46)	0* (0)
Six	27 (0.61)	215 (12.27)	0* (0)	119 (4.03)	130 (28.89)
Seven	81 (2.08)	243 (7.03)	82 (1.97)	279 (3.63)	108 (13.15)
Eight	519 (27.24)	360 (13.79)	0* (0)	153 (4.03)	54 (1.86)
Nine	40 (1.26)	466 (15.36)	5* (0.14)	248 (2.10)	0* (0)
Ten	Bite self: 205 (10.34) Cheek press: 37 (1.49)	461 (15.44)	11 (1.15)	138 (4.84)	0* (0)

* indicates that the frequency of the criterion behaviour < 10, therefore lag sequential analyses were not conducted.

6.4 Results

In order to explore the association between self-injury and proto-imperatives, repetitive behaviour and challenging behaviour a series of lag sequential analyses were conducted. Figure 6.1 presents the results of these and Table 6.3 summarises these results for all participants.

The results revealed that seven of the ten participants evidenced a significant temporal association between self-injury and at least one of: repetitive behaviour, proto-imperative behaviour and/or challenging behaviour. Of the three participants for whom no temporal associations were identified, two had previously been identified as exhibiting self-injurious behaviour maintained by socially mediated consequences. Of the seven for whom temporal associations between self-injury and other behaviours were identified, four had been previously identified as exhibiting self-injurious behaviour maintained by socially medicated consequences.

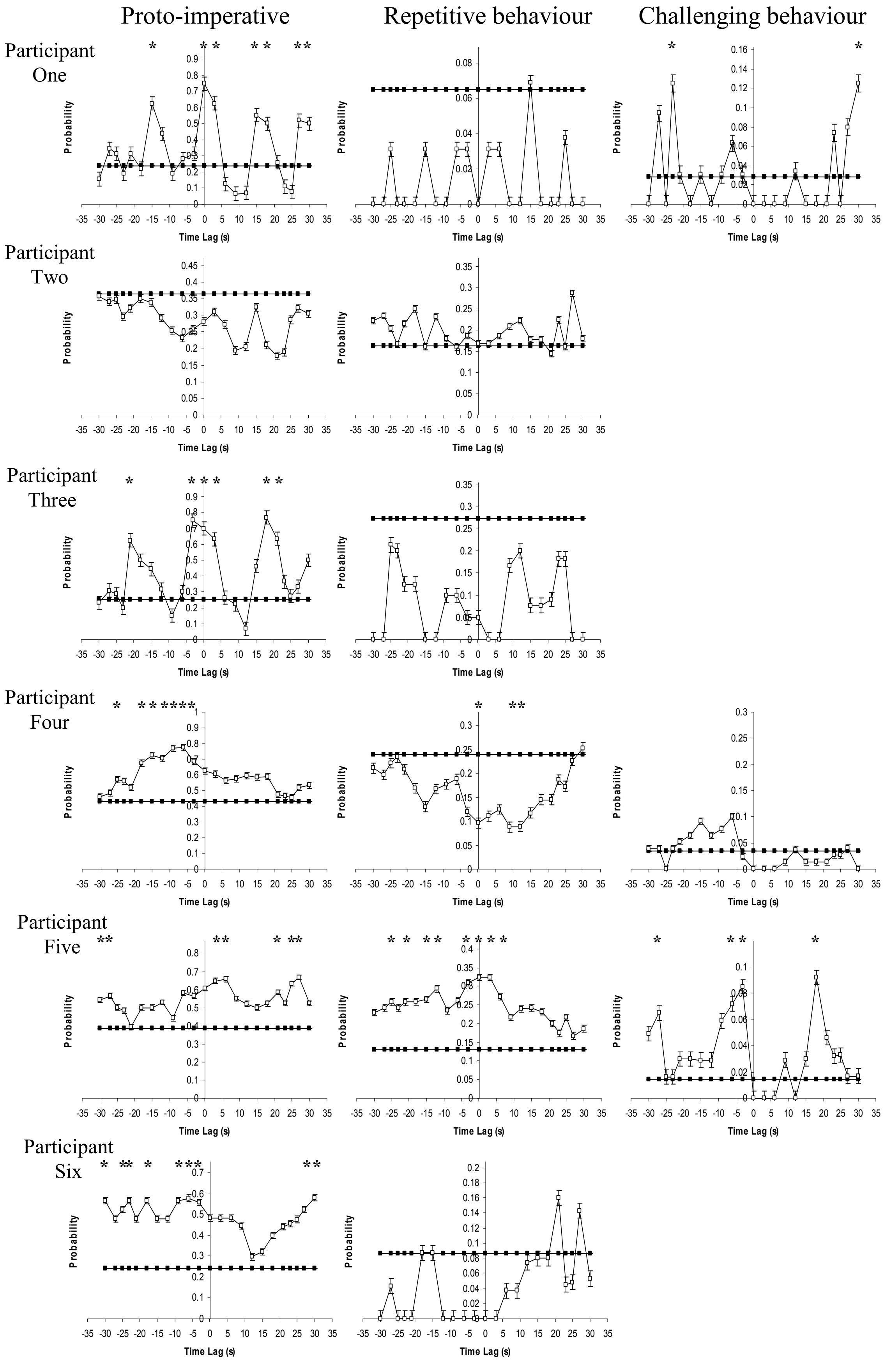


Figure 6.1 Mean unconditional probability (filled squares) of the child engaging in either proto-imperative, challenging behaviour or repetitive behaviour and conditional probability (unfilled squares) of the child engaging in proto-imperative, challenging behaviour or repetitive behaviour given that they are engaging in self-injury, for 21s before and after self-injury occurs (* = z >3.10, p<.001).

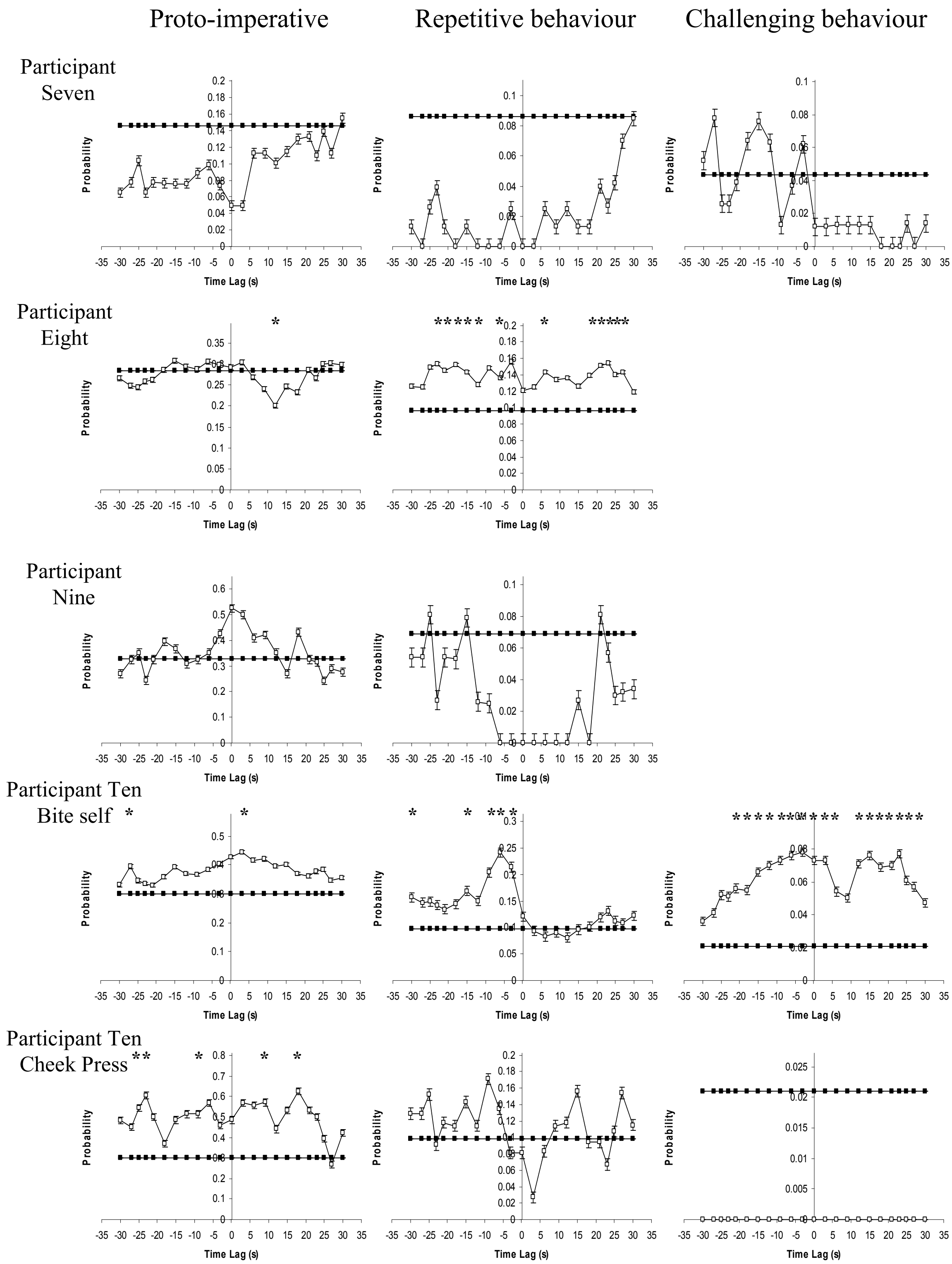


Figure 6.1 cont. Mean unconditional probability (filled squares) of the child engaging in either proto-imperative, challenging behaviour or repetitive behaviour and conditional probability (unfilled squares) of the child engaging in proto-imperative, challenging behaviour or repetitive behaviour given that they are engaging in self-injury, for 21s before and after self-injury occurs (* = z >3.10, p<.001).

Table 6.3 Social function of self-injurious behaviour and strength of social function (Cliffs *d* statistic) and temporal association between self-injurious behaviour and challenging behaviour, repetitive behaviour and proto-imperatives for each participant. + indicates a significant difference between one or more unconditional and conditional probabilities in the time period. o indicates that there were no significant differences between the unconditional and conditional probabilities in this time period.

Social function			**Challenging behaviour**			**Repetitive behaviours**			**Proto-imperatives**		
Participant	**Description**	***d* value**	**Before SIB**[3]	**During SIB**	**After SIB**	**Before SIB**	**During SIB**	**After SIB**	**Before SIB**	**During SIB**	**After SIB**
One	Access to repetitive behaviour[4]	1.00	+	o	+	o	o	o	+	+	+
Two	Access to materials involved in repetitive behaviour	1.00	N/A	N/A	N/A	o	o	o	o	o	o
Three	Sensory escape	1.00	N/A	N/A	N/A	o	o	o	+	+	+
Four	No social function identified	-	o	o	o	o	+[*]	+[*]	+	+	+
Five	No social function identified	-	+	o	+	+	+	+	+	o	+
Six	Gaining answers to repetitive questions	.78	N/A	NA	N/A	o	o	o	+	o	+
Seven	No social function identified	-	o	o	o	o	o	o	o	o	o
Eight	No social function identified	-	N/A	N/A	N/A	+	o	+	o	o	+[*]
Nine	Sensory escape	.83	N/A	N/A	N/A	o	o	o	o	o	o
Ten: bite self	Access to attention in specific location	.75	+	+	+	+	o	o	+	o	+
Ten: cheek press	No social function identified	-	o	o	o	o	o	o	+	o	+

[3] The time period 'Before Self-Injurious Behaviour' is defined as lags between -30 seconds and -3 seconds; the time period 'During Self-Injurious Behaviour' is defined as a lag of 0 i.e. when the self-injurious behaviour occurs; the time period 'After Self-Injurious Behaviour' is defined as lags between 3 seconds and 30 seconds
[4] The repetitive behaviour associated with the identified social function was *not* included in the category of repetitive behaviour for which lag analyses were conducted
[*] In these cases, the conditional probability is significantly *lower* than the unconditional probability

6.4.1 Temporal association between challenging behaviour and self-injurious behaviour

Of the five participants who displayed challenging behaviour, two evidenced a significant association between challenging behaviour and self-injury (Participants One and Five), with challenging behaviour being significantly more likely in the time periods before and after self-injurious behaviour. Both of these participants were identified as exhibiting self-injurious behaviour maintained by socially mediated consequences. Participant 10 exhibited a differential association between topographies of self-injurious behaviour and challenging behaviour. The topography of self-injurious behaviour which was socially maintained (biting self) was temporally associated with challenging behaviour, with challenging behaviour being significantly more likely to occur prior to, during and after self-biting. However, the topography of self-injurious behaviour for which no social function had been identified (cheek press) was not temporally associated with challenging behaviour.

Finally, participants four and seven, for whom no social function for self-injury had been identified, exhibited no temporal association between challenging behaviour and self-injurious behaviour. In summary, a temporal association was identified between challenging behaviour and self-injurious behaviour for three of the five participants who displayed challenging behaviour.

6.4.2 Temporal association between repetitive behaviour and self-injurious behaviour

Four of the ten participants exhibited a temporal association between self-injurious behaviour and repetitive behaviour (Participants Four, Five and Eight; one topography of Participant Ten). For three of these participants, repetitive behaviour was identified as significant precursor to self-injury, being significantly more likely to occur in the 30 second time period before self-injury. Participant Four exhibited an inverse of this association, with repetitive behaviour being significantly less likely during and after self-injurious behaviour occurred. Of the four participants for whom a temporal association between repetitive behaviour and self-injurious behaviour was identified, three exhibited self-injurious behaviour that was *not* maintained by social consequences (Participants Four, Five and Eight). In summary, a temporal association between repetitive and self-injurious behaviours was identified for four of the ten participants.

6.4.3 Temporal association between proto-imperatives and self-injurious behaviour

Seven of the ten participants exhibited a temporal association between proto-imperatives and self-injurious behaviour. Importantly, six of these participants were significantly more likely to evidence proto-imperatives prior to displaying self-injury (Participants One, Three, Four, Five, Six and Ten). An inverse association between proto-imperatives and self-injury was identified for Participant Eight, with proto-imperatives being significantly *less* likely to occur after self-injurious behaviour.

A socially mediated function for self-injury was identified for three of the seven participants for whom a temporal association between proto-imperatives and self-injury was identified (Participants One, Three and Six). However, no social function was identified for a further three of the seven participants (Participants Four, Five and Eight). Both topographies of Participant Ten's self-injury were temporally associated with proto-imperatives.

Interestingly, the three participants for whom no temporal association was evidenced between proto-imperatives and self-injurious behaviour, were the same participants for whom no temporal associations between self-injury and *any* behaviour were evidenced (Participants Two, Seven and Nine). In summary, a temporal association between self-injurious behaviour and proto-imperatives was identified for seven of the ten participants.

6.4.4 Temporal association between self-restraint and self-injurious behaviour

In order to evaluate the association between self-restraint and self-injurious behaviour, lag sequential analyses were conducted for the three individuals who engaged in self-restraint behaviours. Figure 6.2 presents the results of these, revealing that all three participants exhibited a significant temporal association between self-injurious behaviour and self-restraint.

Self-Restraint Behaviour

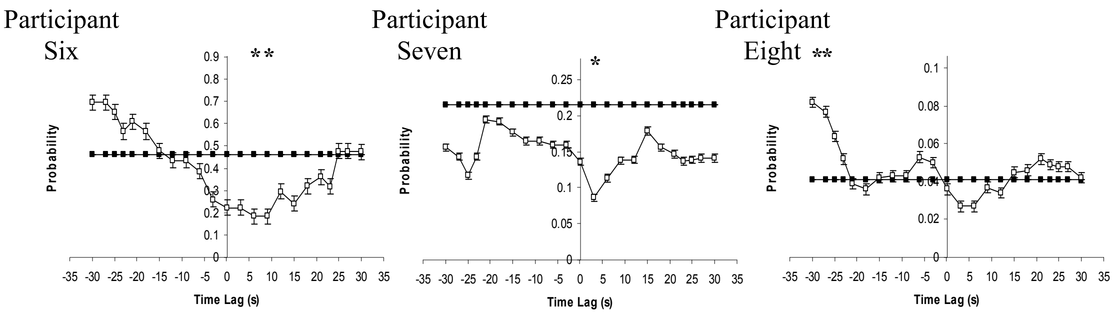


Figure 6.2 Mean unconditional probability (filled squares) of the child engaging in self-restraint and conditional probability (unfilled squares) of the child engaging in self-restraint, given that they are engaging in self-injurious behaviour, for 21 seconds before and after self-injury occurs (* = z >3.10, p<.001).

A common pattern of conditional probabilities is demonstrated across all three participants. The probability of self-restraint is highest prior to self-injury, with Participant Eight being significantly more likely to exhibit self-restraint prior to self-injurious behaviour. The conditional probability of self-restraint then gradually decreases for all participants, through to the time at which the child engages in self-injury. The lowest probability of self-restraint occurring is exhibited in the time period directly after self-injurious behaviour. For Participants Six and Seven, self-restraint is significantly less likely to occur following self-injury. For all participants, the conditional probability of self-restraint then rises. In summary, a significant temporal association between self-restraint and self-injury was evidenced for all three participants.

6.5 Discussion

Significant temporal associations between self-injurious behaviour and: challenging behaviour, repetitive behaviour, proto-imperatives and self-restraint were identified in this study. This was the first study to investigate these associations in a cohort with ASD and was also the largest cohort study investigating behaviours temporally associated with self-injury in any population. The inclusion of individuals with an identified social function to their self-injury *and* individuals without an indentified social function extends findings reported in previous research. All behaviours were operationally defined and a high level of inter-rater reliability was achieved. The study improved upon previous research by assessing ASD diagnosis in the cohort with instruments with good reliability and validity. Additionally, the study employed robust statistical methods to evaluate the temporal associations between behaviours for individuals with ASD. These behaviours were investigated without a-priori confirmation of their temporal proximity to self-injury, allowing for a more complete examination of their associations with self-injury. The results of the study revealed that seven of the ten participants evidenced a significant temporal association between self-injury and at least one of the identified behaviours. Despite the variation in these associations between participants, this finding indicates that self-injurious behaviour in ASD is unlikely to be a randomly occurring motor disorder, but rather is part of a larger class of behaviours within an individual's repertoire. This finding has significant implications for both theoretical and pragmatic issues regarding models of self-injury and intervention for self-injury in ASD populations.

The results indicated that the majority of the participants evidenced significant temporal associations between self-injurious behaviour and at least one other behaviour. More specifically, half of the participants who exhibited other forms of challenging behaviour, demonstrated a significant temporal association between challenging behaviour and self-injurious behaviour. In these cases, challenging behaviours were a significant precursor to self-injury, being more likely to occur in the 30 seconds prior to self-injurious behaviour than in time periods in which self-injury did not occur. This result supports previous findings reported by Petty *et al.* (2009), who also demonstrated other challenging behaviours to be significant precursors to self-injury. The finding provides evidence to discount the conceptualisation of self-injury in ASD as a motor disorder (Bodfish *et al.,* 1995; King, 1993). The temporal proximity of challenging behaviour and self-injury in these participants supports the hypothesis that these behaviours are members of the same response class, although functional equivalence can not be inferred from temporal associations alone. It is likely, given the short time period in which these behaviours co-occur, that they may be subject to the same reinforcement contingencies and evoked by the same antecedents. However, further research is required in order to support the hypothesis of functional equivalence, utilising experimental functional analyses to confirm independently the function of each behaviour.

Although half of the individuals who exhibited challenging behaviour evidenced a temporal association between self-injury and challenging behaviour, a number of individuals' behaviours were not associated in this way. Participants Four, Seven and Ten exhibited challenging behaviour that was not temporally associated with their self injury (in the case of Participant Ten, one topography of self-injury). Interestingly, no social function was identified for the self-injury displayed in these three cases. Taken together, these findings perhaps indicate that the organisation of behaviours may be different in those for whom self-injury is not socially maintained. It could be hypothesised that if self-injury is not socially maintained and therefore is not evoked and maintained by environmental consequences, then functionally equivalent response classes which include self-injury and other challenging behaviours would not form within an individual's behavioural repertoire. In these cases, self-injury would occur with no associations to environmental stimuli, maintained by automatic sensory reinforcement or perhaps associated with pain. Self-injury would have no temporal associations to other forms of challenging behaviour which are unlikely to be reinforced by automatic or sensory contingencies. As no other studies have investigated the temporal associations between behaviours in individuals for whom no social function has been identified, this finding warrants further investigation.

In addition to associations with challenging behaviour, the results also demonstrated that significant temporal associations existed between self-injury and repetitive behaviours for four of the participants. For three children, repetitive behaviour was identified as a significant precursor to self-injury (Participants Five, Eight and Ten). This finding partially supports those reported by Petty *et al.* (2009) who demonstrated that repetitive behaviours were a significant precursor to self-injury, in four individuals from a cohort of six. The finding is also in line with studies that preselected participants who exhibited precursor behaviours and subsequently demonstrated that repetitive behaviours can be significant precursors to self-injury (Hagopian *et al.,* 2005; Herscovitch *et al.,* 2009; Langdon *et al.,* 2008). Taken together, these results indicate that for some individuals with ASD, repetitive behaviour and self-injury occur in close temporal proximity and therefore may be members of the same response class. The temporal proximity of repetitive behaviour and self-injury suggests that the process of superstitious reinforcement is a possible explanation in this population and therefore provides tangential support to the transitional model proposed by Guess and Carr (1991).

However, six participants with ASD did not demonstrate any temporal association between self-injury and repetitive behaviour. This finding is particularly surprising given the high levels of repetitive behaviour reported in individuals with ASD (Estes *et al.,* 2011; Richler *et al.,* 2007; Turner, 1999). In some cases, this may be due to the relatively conservative statistical analysis conducted in this study. For example, visual inspection of Participant Ten's Cheek Press data, reveals that the conditional probability of repetitive behaviour is highest prior to self-injury and then a large decrease in the conditional probability occurs following self-injury. However, the analysis conducted compares the conditional probability to the unconditional probability over time and therefore, this large decrease in conditional probability does not reach significance, although the changes in behaviour observed may have pragmatic and clinical significance. However, utilisation of a similar statistical method revealed repetitive behaviour as a significant precursor to self-injury in the majority of cases reported by Petty *et al.* (2009) and consequently, conservative statistical analysis cannot fully explain this pattern of results.

An alternative explanation may be that the temporal associations between repetitive behaviour and self-injury in ASD are mediated by social function. The results from this study revealed that the majority of the individuals for whom a temporal association was identified between self-injury and repetitive behaviour, displayed self-injury that was *not* maintained by socially mediated reinforcement. It may be that for individuals with ASD, who exhibit 'non-functional' self-injury, repetitive behaviour clusters around their self-injury and is therefore a member of a response class reinforced by automatic or sensory reinforcement. Conversely, it could be hypothesised that in individuals with ASD and self-injury that is maintained by social reinforcement, repetitive behaviour rarely forms a response class with self-injury. Repetitive behaviour may be predominantly automatically reinforced in individuals with ASD and once self-injurious behaviour has gained a socially mediated function, repetitive behaviour and self-injury would subsequently not be evoked by the same establishing operations. This would lead to repetitive behaviours being infrequently shown in close temporal proximity to socially maintained self-injurious behaviour. These hypotheses require significant future research attention, in samples large enough to test for significant difference between groups with socially mediated self-injury and groups with automatically reinforced self-injury.

In contrast to the varied results for temporal associations between challenging behaviour, repetitive behaviour and self-injury, a more homogeneous pattern of results was evidenced for the temporal associations between proto-imperatives and self-injury. Seven of the participants exhibited proto-imperatives that were temporally associated to self-injurious behaviour. Importantly, for six of these participants, proto-imperatives were significant precursors to self-injury. This supports findings reported by Petty *et al.* (2009) who demonstrated that proto-imperatives were significant precursors to self-injury for all of their cohort of six participants. This finding is novel in an ASD sample and has significant implications for both theoretical and pragmatic models of self-injury in ASD. First, the close temporal proximity of proto-imperatives and self-injury supports the possibility of superstitious learning occurring and provides tangential support to the model proposed by Guess and Carr (1991). This model is further strengthened by two cases in which *both* proto-imperatives and repetitive behaviour were significant precursors to self-injury, indicating that the three behaviours may form a response class (Participants Five and Ten). The close temporal proximity of these three behaviours means that both the repetitive and self-injurious behaviours are more likely to receive inadvertent social reinforcement when the proto-imperatives are reinforced. In this way, the repetitive behaviours may have been exposed to social reinforcement and shaped over time to self-injurious behaviour. This preliminary finding reinforces the necessity of longitudinal studies to evaluate the emergence of self-injurious behaviour in ASD.

Second, the close temporal proximity of proto-imperatives prior to self-injury in the majority of participants with ASD has important implications for intervention. The results extend findings reporting communicative acts as precursors to self-injury in those individuals who are known to display precursors (Herscovitch *et al.,* 2009). This study demonstrates that in the majority of individuals with ASD, with both ‘functional’ and ‘non-functional’ self-injury, proto-imperatives are naturally occurring within an individual’s behavioural repertoire proximate to and before self-injury (Borrero & Borrero, 2008). This allows for the utilisation of naturally occurring proto-imperatives to signal to carers the point in the response chain at which a functionally equivalent response would be most effective. Importantly, it also suggests that these proto-imperatives could be shaped for use as functionally equivalent responses in FCT, to replace self-injury. Although this study has not demonstrated functional equivalence of proto-imperatives and self-injurious behaviour, the temporal proximity of these behaviours makes it likely that they are inadvertently receiving the same reinforcement contingencies. Consequently, the naturally occurring proto-imperatives would provide a logical and identifiable starting place for FCT, reducing the intensive intervention required to teach a new communicative response and mitigating the ethical issues associated with intervention for self-injury. Future research should investigate the plausibility of shaping naturally occurring proto-imperatives in ASD, to be used as functionally equivalent responses to replace self-injurious behaviour.

A final significant finding in this study was that all individuals who engaged in self-restraint behaviours, exhibited a significant temporal association between self-restraint and self-injury. This finding supports previous research associating the presence of self-injury and self-restraint (Fovel *et al.,* 1989; Hyman *et al.,* 2002; Sections 4.4.3 & 4.4.5) and extends findings reporting a temporal association between self-injury and self-restraint to an ASD population (Forman *et al.,* 2002). Interestingly, the temporal association between self-injury and self-restraint was broadly similar across all three participants who evidenced both behaviours. In all three cases, the conditional probability of self-restraint decreased proximal to self-injury, with the lowest conditional probability occurring just after self-injurious behaviour had been displayed. Following this, the conditional probability of self-restraint rose for all participants. This pattern of probabilities, arguably, provides support for the model of self-injurious behaviour as a failure in inhibitory control, presented in Chapter 4. If self-restraint functions to inhibit impulsive self-injury, it can be seen that the steadily decreasing probability of self-restraint are reflective of an inverse, increasing compulsion to engage in self-injury. It can be hypothesised that, at the point of self-injurious behaviour occurring, a ‘threshold’ of inhibitory control has been reached and the individual is no longer able to inhibit their compulsive self-injury. Consequently self-injury occurs and self-restraint ceases. Immediately following the occurrence of self-injury, the drive to engage in self-injury would be weaker, hence the very low conditional probabilities of self-restraint. Subsequently, the compulsion to re-engage in self-injury rises, mirrored by a rise in the conditional probability of self-restraint, to inhibit the compulsive self-injury. This tentative hypothesis requires experimental confirmation in an ASD population. If these preliminary findings are further substantiated, one clinical implication may be that interventions to protect individuals from self-injurious behaviour should be available immediately when the individual ceases to self-restrain.

The results of this study provide provisional support for self-injury being part of a response class in children with ASD. However, a key limitation of the study was that function of each class of behaviour was not evaluated experimentally. Additionally, the lag analyses conducted to investigate the temporal associations between behaviours were unrestricted (i.e., they may have inadvertently included the preceding or following episodes of self-injurious behaviour). This may have resulted in the data in the graphs being broadly flattened across time. However, for the majority of participants, visual inspection of the data reveals that this was not the case. Future research should build upon these initial findings and incorporate experimental functional analyses for all classes of behaviours, collecting a greater volume of data for each participant in order to conduct restricted analyses. These modifications would build upon the preliminary findings reported in this study.

The findings in this study also provide tentative support for the developmental model of self-injury proposed by Guess and Carr (1991). The results allude to a potential difference in the temporal associations of behaviours in those with socially maintained and non-socially maintained self-injury. However, these tentative results highlight an increasing need for large sample, robust longitudinal studies to evaluate the emergence and development of self-injurious behaviour. These studies are necessary not only in ASD populations, but also in intellectual disability populations. An effective early intervention strategy for self-injury necessitates a greater understanding of the early developmental trajectory of self-injury in very young children, which is currently absent from the research literature. There is a pressing need for empirical data, in studies specifically designed to test the hypotheses generated from developmental models of self-injury, such as the one proposed by Guess and Carr (1991).

In summary, the findings of the study demonstrated significant temporal associations between self-injury and challenging behaviour, repetitive behaviour and proto-imperatives for the majority of individuals with ASD. Additionally, all individuals who exhibited self-restraint, evidenced a significant temporal association between self-injury and self-restraint. These findings have implications for theories of response classes, developmental models of self-injury, theories of inhibitory control in self-injury and the utilisation of communication interventions for self-injury in ASD.

CHAPTER 7

GENERAL DISCUSSION

7.1 Preface

The study in Chapter 6 described the temporal associations between self-injury and other behaviours in a group of children with ASD. Fine grained lag sequential analyses were employed to address both theoretical and pragmatic questions regarding the application of operant theory to self-injury in ASD. The results identified significant temporal associations between self-injury and other forms of challenging and repetitive behaviours and proto-imperative communicative behaviours for the majority of the sample. These findings indicate that self-injury appears to have the properties of a functionally communicative behaviour which is associated with repetitive behaviours that might have featured at an earlier developmental stage. Additionally, these results suggest that interventions for self-injury such as functional communication training may be usefully employed in individuals with ASD. The results also delineated a temporal association between self-injurious behaviour and self-restraint. This contributed to a developing model of impaired inhibitory control in some cases of self-injury in ASD.

In this chapter, the results from all of the studies will be discussed and synthesised with existing literature, with a view to developing a dynamic model of self-injury in ASD.

7.2 <u>Introduction</u>

Self-injurious behaviour is displayed by between 4 and 12% of individuals with intellectual disability of heterogeneous aetiology (Cohen *et al.,* 2010; Cooper *et al.,* 2009; Emerson *et al.,* 1997; Holden & Gitlesen, 2006; Lowe *et al.,* 2007; Oliver, Murphy & Corbett, 1987). Differing causal explanations have highlighted a role for neurobiological (Sandman, 1992; Sandman, Barron, Chicz-DeMet & DeMet, 1990; Symons, Clark, Hatton, Skinner, & Bailey, 2003; Symons & Thompson, 1997), pain related (Carr & Owen-Deschryver, 2007; Carr, Smith, Giacin, Whelan & Pancari, 2003; Christensen *et al.*. 2009; O' Reilly, 1997) and operant influences upon self-injury (Emerson, 1998; Carr & Durand, 1985; Iwata, Dorsey, Slifer, Bauman & Richman, 1994; Oliver, 1993; 1995). The consequences of self-injury are known to be pervasive and damaging, influencing quality of life (Beadle-Brown, Murphy & DiTerlizzi, 2009), care provision and practices (Allen, Lowe, Brophy & Moore, 2009; McGill *et al.,* 2009) and the lives of carers and families (Hastings, 2003; Totsika, Hastings, Emerson, Lancaster & Berridge, 2011; Seltzer, *et al.,* 2010).

Preliminary data suggest that the prevalence of self-injury is heightened in individuals with ASD compared to those with intellectual disability but not ASD, with prevalence data ranging from 10.3% to 70.6% (Ando & Yoshimura, 1979a; 1979b; Baghdadli, Pascal, Grisi, & Aussilloux, 2003; Bartak & Rutter, 1976; Bradley, Summers, Wood & Bryson, 2004; Billstedt, Gillberg & Gillberg, 2005; LeCavalier, 2006; McTiernan, Leader, Healy & Mannion, 2011; Murphy, Healy & Leader, 2009; Shattuck *et al.,* 2007). Despite this elevated prevalence, research investigating self-injury in individuals with ASD is somewhat limited. As highlighted in Chapter 1 the validity of the conclusions drawn in prevalence studies has been threatened by absent or limited descriptions of participant characteristics. Additionally, few data have been generated to delineate person characteristics associated with self-injury in ASD or to develop a model of self-injury in which the elevated prevalence is accounted for. Therefore, the broad aim of this thesis was to utilise a multi-method approach to detail the epidemiology, associated person characteristics and operant function of self-injury in multiple samples of individuals with ASD. The results of these studies would then contribute toward a more comprehensive model of self-injury in ASD.

To achieve this aim, five empirical studies were conducted. In three studies survey methodologies were used to assess comparatively large populations of individuals with ASD. These studies generated novel data regarding the prevalence and persistence of self-injury, and allowed for the identification and evaluation of person characteristics as putative risk markers for self-injury in ASD. A fine grained cohort study was undertaken, in order to evaluate the impact of operant influences upon self-injury in ASD. Further statistical analysis allowed for the delineation of temporal associations within each individual's behavioural repertoire. The findings, strengths, limitations and clinical implications of this research will be discussed below, with reference to the existing literature. A preliminary model of self-injury in ASD will be proposed, allowing for hypothesis generation for future research.

7.3 Main Findings

Given the broad aim of this thesis and the diverse range of methods employed, the key results and implications will be considered most usefully within three domains: epidemiology, person characteristics and function of self-injury in individuals with ASD.

7.3.1 Epidemiology of self-injurious behaviour in autism spectrum disorder

7.3.1.1 Prevalence and risk of self-injurious behaviour

A key aim of this thesis was to generate a robust and reliable estimate of the prevalence of self-injurious behaviour in individuals with ASD. Previous prevalence estimates had been highly divergent and limited by exclusive reliance on prior clinical diagnosis of ASD (Ando & Yoshimura,1979a; Bartak & Rutter, 1976; Janicki & Jacobson, 1983; Lecavalier, 2006; Murphy *et al.,* 2009), limited examination of intellectual or adaptive functioning (Cooper *et al.,* 2009; Janicki & Jacobson, 1983), small sample sizes (Bradley *et al,* 2004; Bartak & Rutter, 1976) and differences in the specified time frames for self-injury. The results of the studies presented in Chapters 2, 3 and 4 estimated prevalence rates of self-injury in the preceding month of 50%, 41.7% and 47.4%[1] respectively. Taken together, these figures indicate the prevalence of self-injury at between 40 and 50%, which is in line with the most robust published prevalence data (Ando & Yoshimura, 1979a; 1979b; Baghdadli *et al.,* 2003; Bradley *et al,* 2004; Billstedt, Gillberg & Gillberg, 2005; Murphy *et al.,* 2009; Shattuck *et al.,* 2007). The inclusion of a screen for ASD phenomenology and assessment of adaptive functioning in Chapters 2 and 3 strengthens the external validity of the findings. Additionally, the large samples surveyed and the employment of standardised, reliable and valid measures to collect the data further support the validity of the findings. The consistency of the reported

[1] Summed prevalence for both child and adult samples reported here. For the child sample alone, prevalence = 45.7%. For the adult sample alone, prevalence = 49.1%.

prevalence figures in this thesis, across three survey studies, suggests that a prevalence figure of 40% to 50% is robust and replicable.

This finding has significant implications for clinical practice and service provision, as it indicates that at any time, as many as one in two individuals with ASD will have engaged in self-injury in the previous month. Given the significantly lower prevalence rates reported in populations with intellectual disability of heterogeneous aetiology, this finding also alludes to the increased risk of self-injury reported in ASD populations (McClintock, Hall & Oliver, 2003). The concept of increased risk of self-injury was supported by data presented in Chapter 2, which revealed that individuals with ASD are 2.67 times more likely to engage in self-injury than individuals with Down syndrome. This demonstrates that individuals with ASD are at heightened risk of developing self-injurious behaviour compared to individuals with intellectual disability of heterogeneous aetiology or specific syndromes, such as Down syndrome, in which self-injury is typically observed at levels comparable to those seen in individuals with intellectual disability of heterogeneous aetiology.

Importantly, the association between ASD and self-injury was further examined in this thesis by investigating the associations between ASD phenomenology and self-injury in other populations. The results indicated that self-injury was significantly associated with the total score on a measure of ASD phenomenology in samples with Fragile X and Down syndromes. This finding supports data collected in groups of individuals with rare genetic syndromes and heterogeneous aetiology of intellectual disability, where ASD phenomenology was also associated with self-injury (Arron, Oliver, Berg, Moss & Burbidge, 2011; Bhaumik, Branford, McGrother & Thorp, 1997; Collacott, Cooper, Branford & McGrother, 1998; Lowe *et al.*.

2007; Murphy *et al.,* 2005). Importantly, these findings extend the concept of ASD diagnosis as a risk marker for self-injury, and suggest that it is the cumulative presence of ASD behaviours, rather than idiopathic autism per se, that is associated with self-injury. These findings can be interpreted in light of operant theories regarding the communicative function of self-injury and the development of self-injury from repetitive behaviours (Guess & Carr, 1991). An increased risk of self-injury associated with an increase in ASD type repetitive behaviours has face validity if self-injury is seen to develop from repetitive and stereotyped behaviour (Guess & Carr, 1991; Petty, Oliver & Allen, 2009).

7.3.1.2 Topography and severity of self-injurious behaviour

The results in Chapter 2 also delineated the topography and severity of self-injury displayed by individuals with ASD. The analysis of forms of self-injury in this population was novel, and the results indicated that whilst a wide range of topographies were displayed, the most prevalent form of self-injury was self hitting. The prevalence data were consistent across studies with 29.5% and 26.2% of the ASD samples engaging in this behaviour (results from Chapters 2 and 4[2] respectively). Additionally, the results in Chapter 2 revealed that individuals with ASD were approximately 4.8 times more likely to engage in self-hitting than individuals with Down syndrome, perhaps suggesting a specificity of behaviour in individuals with ASD, or potentially a low prevalence of this topography in individuals with Down syndrome. Finally, the study in Chapter 3 revealed that all topographies of self-injury were stable over a three year period, with self-hitting being the most persistent form of self-injury over time. Given the putative associations between 'targeted' self-injury and theories of pain related self-injury, the specificity of topography of self-injury in individuals with ASD

[2] Summed prevalence for both child and adult samples reported here. For the child sample alone, prevalence = 24.5%. For the adult sample alone, prevalence = 28.2%.

warrants further investigation (Breau, Camfield, McGrath & Finley, 2003; Symons, Clark, Hatton, Skinner & Bailey, 2003; Symons & Thompson, 1997).

An analysis of the severity of self-injury in individuals with ASD revealed that self-injury displayed in this population was no more severe than self-injury displayed by individuals with Down syndrome or Fragile X syndrome. However, the results from the longitudinal study revealed that self-injury in individuals with ASD remained at a stable level of severity over time.

7.3.1.3 <u>Persistence of self-injurious behaviour</u>

This thesis included the first study of the persistence of self-injury in an ASD sample that were not recruited from services providing clinical interventions. The results demonstrated that self-injury was persistent in 77.8% of individuals with ASD over three years. This finding replicates robust results from populations of individuals with intellectual disability, where self-injury is reported to be persistent in 71% and 84% of individuals, over 7 and 20 years respectively (Emerson *et al.,* 2001b; Taylor, Oliver & Murphy, 2011). Clinically these findings suggest that individuals with ASD are at high risk of developing self-injury, and that once the behaviour is established, it is unlikely to reduce without intervention. If a pragmatic argument is accepted, that intervention may be more effective when children are smaller and learning histories are shorter, these data indicate that an early intervention strategy to reduce self-injury in individuals with ASD may be warranted.

7.3.1.4 Summary of epidemiology of self-injurious behaviour in autism spectrum disorder

In summary, the results of this thesis have reliably demonstrated that self-injury is displayed by approximately 50% of individuals with ASD. The concept of an increased risk of self-injury in ASD has been supported and extended to demonstrate that the presence of ASD phenomenology in other populations is also associated with self-injury. Importantly, the results have also demonstrated that self-injurious behaviour is persistent across 3 years in close to 80% of individuals with ASD.

7.3.2 Person characteristics associated with self-injurious behaviour in autism spectrum disorder

A second key aim of this thesis was to delineate person characteristics associated with the presence and persistence of self-injury in ASD. These findings allow further delineation of putative risk markers for self-injury and support a comprehensive model of contributory factors in the development and maintenance of self-injury. Through this thesis, four key person characteristics have been identified as associating with self-injury in ASD: adaptive functioning, painful health conditions, repetitive behaviours and overactive and impulsive behaviours.

7.3.2.1 Adaptive functioning

Greater degree of intellectual disability, as evidenced through compromised expressive language (Collacott *et al.,* 1998; Emerson *et al.,* 2001a; Murphy *et al.,* 2005; Schneider, Bijam-Schulte, Janssen & Stolk, 1996), impaired mobility (Murphy, Hall, Oliver & Kissi-Debra, 1999) and reduced cognitive functioning (Collacott *et al.,* 1998; Schroeder, Schroeder, Smith & Dalldorf, 1978) have all been associated with self-injury in individuals with intellectual disability of heterogeneous aetiology. Importantly, adaptive functioning, expressive language and ASD symptoms are often confounded when the impact of intellectual disability upon self-injury is examined (McClintock *et al.,* 2003). The results in Chapters 2 and 3 demonstrated that, in a population where ASD symptomatology is clearly defined, both poorer expressive language and lower levels of adaptive functioning are significantly associated with self-injury. The association between lower adaptive functioning and self-injury was replicated in the large child sample assessed in Chapter 4. These findings support previous data associating deficits in expressive language (Dominick, Davis, Lainhart, Tager-Flusberg & Folstein, 2007) and low levels of adaptive functioning (Baghdadli *et al.,* 2003) with self-injury in individuals with ASD. The findings in this thesis extend these preliminary results by delineating the association between lower ability and self-injury in a population where ASD phenomenology has been carefully screened. Thus, the results indicate that even in populations with ASD, lower levels of adaptive functioning are still associated with the presence of self-injury.

However, the results of this thesis also suggest that the relationship between adaptive functioning and self-injury in individuals with ASD is not as simple as perhaps first thought. Whilst some associations were identified between variables indicative of lower ability and self-injury, these associations were not consistent across all data sets. For example, whilst deficits in expressive language were associated with self-injury at T_1 in Chapter 2 and T_2 in Chapter 3, these expressive language deficits did not predict the persistence of self-injury over time. Similarly, lower levels of adaptive functioning were associated with self-injury at T_2, but not at T_1. Likewise, logistic regressions revealed that poorer adaptive functioning independently contributed to the presence of frequent self-injury in the children, but did not significantly influence the presence of self-injury or frequent self-injury in adults (results from Chapter 4). These findings suggest a more complex relationship between ability level and self-injury in individuals with ASD that may vary across time and between age groups. Therefore, whilst ability level may be associated with self-injury at certain times and in certain age groups, further longitudinal research is required in order to evaluate the validity of lower levels of ability as a consistent risk marker for self-injury in individuals with ASD.

7.3.2.2 Painful health conditions

In addition to assessing the association between ability levels and self-injury in ASD, this thesis included the first large scale study of the prevalence of health problems and their associations with self-injury in children and adults with ASD. In accordance with previous research in individuals with intellectual disability of heterogeneous aetiology, health problems were found to be very common in both children (38.5%) and adults (61.1%) with ASD (Berg, Arron, Burbidge, Moss & Oliver, 2007, Haveman et al., 2000; Jansen, Krol, Groothoff, & Post, 2004; van Schrojenstein Lantman-De Valk *et al.,* 2000; van Schrojenstein Lantman-de Valk, Linehan, Kerr & Noonan-Walsh, 2007). Interestingly, despite the elevated prevalence of health problems in the adult sample, health problems were only associated with self-injury in the child sample, replicating findings associating painful health conditions and self-injury in children with intellectual disability (Davies, 2010; Petty, Bacarese-Hamilton & Oliver, In Prep). Importantly, this thesis demonstrated that the presence of one or more painful health conditions, increased the likelihood of children with ASD engaging in self-injury by 2.3 times and increased the likelihood of them engaging in very frequent self-injury by 3.5 times.

This novel finding, associating painful health problems and self-injury in children with ASD has significant clinical implications. Given that interventions to reduce painful health conditions have resulted in reductions in self-injury, these findings suggest that assessment and treatment of painful health conditions should be a component of any evidence based assessment of self-injury in ASD (Christensen *et al.*, 2009; Bosch, Van Dyke, Smith & Poulton, 1997). Additionally, with a view toward an early intervention strategy, carer vigilance to observable painful health conditions and changes in individuals pain related behaviours may provide a low cost, high benefit intervention.

Whilst longitudinal conclusions cannot be drawn from the cross sectional findings in this thesis, the differences in the child and adult data presented in Chapter 4 are interesting with regard to the early development of self-injury. From these data, it could be hypothesised that painful health conditions are implicated in the early development and maintenance of self-injury during childhood, but have less influence in adulthood once the behaviour is established in an individual's repertoire. This type of model is similar to that proposed by Guess and Carr (1991), with painful health conditions fulfilling a similar role to repetitive behaviours in Stages One and Two of the model. The presence of painful health conditions may subsequently lead to self-injury in an attempt to remove or 'gate' painful experiences (Melzack & Wall, 1965). Once environmental reinforcement maintains self-injury, the causal role of painful health conditions may be diminished, although pain may still moderate the influence of environmental contingencies in the form of setting events (Carr *et al.,* 2003; Carr & Blakeley-Smith, 2006; Carr & Smith, 1995). This hypothesis requires further investigation.

7.3.2.3 Repetitive behaviours

A third important association identified in this thesis was between repetitive behaviours and self-injury in individuals with ASD. The longitudinal study reported in Chapters 2 and 3 and the large scale survey study in Chapter 4 demonstrated that the presence of repetitive behaviours was associated with the presence of self-injury. The logistic regression model presented in Chapter 4 revealed that children with high levels of repetitive behaviour were 3.9 times more likely to engage in self-injury, and adults with high levels of repetitive behaviour were 2.5 times more likely to engage in frequent self-injury. This supports previous findings associating repetitive behaviours and self-injury in individuals with intellectual disability (Collacott *et al.,* 1998; Davies, 2010; Emerson *et al.,* 2001a) and ASD (Dominick *et al.,* 2007). The results in this study extend these preliminary findings by delineating the specific forms of repetitive behaviour which were associated with self-injury, revealing that levels of compulsive behaviours and insistence on sameness differed between individuals with and without self-injury. Importantly, this finding was identified in a matched case-control analysis, suggesting that differences in ASD phenomenology and ability level do not account for the difference in repetitive behaviour. Finally, compulsive repetitive behaviours remained associated with self-injury at T_2 of the longitudinal study and, in a composite with overactive and impulsive behaviours, predicted persistent self-injury over absent self-injury across 3 years. A caveat to these findings is that repetitive behaviours were not associated with both the presence and frequency of self-injury in both the adult and child samples in Chapter 4. Nonetheless, these findings do suggest that in individuals with ASD, repetitive behaviours are a relatively reliable risk marker for the presence of self-injury.

One explanation to account for the association between self-injurious and repetitive behaviours can be drawn from Guess and Carr's (1991) model of the emergence of self-injury. Guess and Carr (1991) proposed that self-injury developed from repetitive behaviours through a process of social and non-social reinforcement. This model indicates a direct link between repetitive behaviours as a longitudinal precursor to self-injury, and therefore the evidence identifying repetitive behaviours as risk markers for self-injury has good face validity. The study in Chapter 6 also provides tangential support for this explanation by revealing temporal associations between individual episodes of self-injury and repetitive behaviour for four of the ten participants. Similar temporal associations have also been identified in individuals with intellectual disability of heterogeneous aetiology (Petty *et al.,* 2009). However, this explanation is limited by the fact that only four of the ten participants in this thesis evidenced this temporal association between repetitive behaviours and self-injury. It is plausible that for the additional six participants, repetitive behaviours are replaced in the behavioural repertoire with self-injurious behaviour, however, it may be expected from Guess and Carr's model (1991) that this would occur in all or none of the cases, rather than in a proportion of them. Alternatively, self-injury may only develop from repetitive behaviours for a proportion of individuals with ASD, with painful health conditions contributing to another developmental pathway in some cases.

An alternative and complementary explanation may draw upon Turner's (1999) theory of repetitive behaviour. Turner (1999) proposed that impairments in executive functioning underpin the development of all repetitive behaviour. Thus, repetitive behaviours may be a risk marker for self-injury as both repetitive and self-injurious behaviours and control of both behaviours is by the same underlying cognitive processes. This explanation differs slightly from the explanation drawn from Guess and Carr's model (1991). Using Guess and Carr's model, repetitive behaviours would be a *risk factor* for self-injury as they provide a necessary and causal contribution to the development of self-injury. However, using Turner's model, impairments in behavioural control provide the causal contribution and thus repetitive behaviours may co-occur with self-injury as a *risk marker*, without necessarily having a causal contribution to the development of self-injury. Therefore, the explanation developed from Turner's model allows for the slightly changeable associations between self-injury and repetitive behaviours identified in this study. As repetitive behaviours are hypothesised to have no direct association with self-injury apart from being underpinned by the same causal mechanism, the observable association between repetitive behaviour and self-injury at any given time point is free to vary.

Alternatively, it is possible that both explanations have validity in modelling the association between repetitive behaviours and self-injury. Drawing upon Turner's model (1999), both behaviours may be underpinned by deficits in behavioural control. However, drawing upon Guess and Carr's model (1991), there may be a developmental progression from repetitive behaviours initially to self-injurious behaviours. The trajectory of this progression may be influenced by the degree of impairment in behavioural control, with poorer behavioural control leading to both an increased emergence of repetitive behaviours (Turner, 1999) and a quicker progression from repetitive behaviour to self-injury. Importantly, this model may also predict that the effects of impaired behavioural control would be seen in other classes of behaviour, such as overactivity and impulsivity, in addition to the effects seen in repetitive behaviours and self-injury.

7.3.2.4 Overactive and impulsive behaviours

A novel pair of person characteristics identified in this thesis as being associated with self-injury was overactivity and impulsivity. Findings from individuals with intellectual disability of heterogeneous aetiology indicated that overactivity and impulsivity were risk markers for self-injury (Arron *et al.,* 2011; Cooper *et al.,* 2009; Collacott *et al.,* 1998; Oliver, Sloneem, Hall & Arron, 2009; Schneider *et al.,* 1996). This thesis supported those findings, demonstrating that for individuals with ASD, both overactivity and impulsivity were reliably associated with self-injury at T_1 and T_2 of the longitudinal study. Additionally, a composite of overactivity and impulsivity was demonstrated to increase the risk of self-injury and frequent self-injury in both children and adults between two and six fold. Finally, the presence of overactive and impulsive behaviours was demonstrated to predict the persistence of self-injury over a three year period. Importantly, the combination of overactivity and impulsivity was the only person characteristic which was consistently associated with self-injury across all three of the survey studies in this thesis. The consistent association of overactivity and impulsivity, both independently and combined, suggests that in individuals with ASD these two characteristics may be usefully considered as one risk marker for self-injury.

These findings have supported a model of self-injury developed through theories of Attention Deficit Hyperactivity Disorder (ADHD; Barkley, 1997) and repetitive behaviour (Turner, 1999), in which impairments in behavioural inhibition are seen to drive the development of self-injury. The identified associations between self-injury, repetitive behaviour and impulsivity\overactivity support a hypothesis in which a fundamental deficit in behavioural control leads to an increased persistence and severity of self-injury. Impaired behaviour control may lead to self-injury being initiated at a low threshold in the presence of discriminative stimuli and/or self-injury being difficult for the individual to terminate.

Importantly, tangential evidence of difficulties terminating self-injury was identified in this thesis through an examination of self-restraint behaviours. Data in this thesis demonstrated that self-restraint was associated with self-injury and impulsive/overactive behaviours, supporting a theory that these behaviours are underpinned by a common deficit in behavioural inhibition. Additionally, the concept of self-restraint functioning to inhibit self-injury was supported by the study of temporal associations in Chapter 6, where self-restraint was evidenced to significantly increase prior to self-injury and significantly decrease following self-injury. These novel findings have significantly extended the theoretical understanding of self-injury in ASD, and have posited a putative cognitive mechanism by which these behaviours are moderated.

7.3.2.5 Summary of person characteristics associated with self-injurious behaviour in autism spectrum disorder

In summary, the results of this thesis have identified four person characteristics to be reliably associated with self-injury in ASD: low ability levels, painful health conditions, repetitive behaviours and impulsive/overactive behaviours. These findings have important implications for both theoretical and clinical understanding of self-injury in ASD. Firstly, the findings have contributed to a hypothesised causal model where impairments in behavioural inhibition and/or the presence of health conditions are identified as influencing the presence of self-injury in individuals with ASD. Secondly, these findings have highlighted those person characteristics which may have utility as risk markers for self-injury in individuals with ASD. Thus, the plausibility of an early intervention strategy for self-injury in ASD is enhanced, with the possibility of being able to identify those individuals with ASD most at risk of developing self-injury.

7.3.3 Function of self-injurious behaviour in autism spectrum disorder

A final key aim of this thesis was to evaluate systematically the utility of an operant model in accounting for the maintenance of self-injury in individuals with ASD.

7.3.3.1 Operant function

Previous research had suggested that the most commonly employed functional analyses failed to identify social functions for self-injury in individuals with ASD (Hagopian, Bruzek, Bowman, & Jennett, 2007; Hausman *et al.,* 2009; Healey *et al.,* 2001; McKerchar *et al.,* 2001). Consequently, researchers had concluded that self-injury may be more frequently automatically maintained in individuals with ASD than in individuals with intellectual disability of heterogeneous aetiology (O'Reilly *et al.,* 2010). The results of this study supported the data presented in previous research, by demonstrating that the most commonly employed conditions in experimental functional analyses failed to identify social functions for self-injurious behaviour in ten children with ASD. However, in a novel methodological progression, the systematic assessment of ASD weighted functions for self-injury revealed socially mediated functions in six of the ten children. Supporting previous case studies, the majority of the functions identified in this cohort were associated with ASD person characteristics such as high levels of sensory sensitivity (Buckley & Newchock, 2006; Devlin, Healy, Leader & Reed, 2008) or preference for repetitive behaviours and routines (Hausman *et al.,* 2009; Murphy, McDonald, Hall & Oliver, 2000; Reese, Richman, Belmont & Morse, 2005; Reese, Richman, Zarcone & Zarcone, 2003). The results of this study were strengthened by the utilisation of robust assessments for ASD phenomenology and intellectual functioning, and through the delineation of reliable behavioural codes and statistical analysis of function.

These results have validated the potential application of operant theories for the understanding of the maintenance of self-injury in individuals with ASD. Clinically, these findings suggest that commonly employed behavioural interventions designed to reduce challenging behaviour would be applicable in this population, provided they were targeted towards correctly identified ASD weighted antecedents and consequences. These findings also have theoretical implications to an understanding of the elevated prevalence of self-injury in ASD, building upon the hypothesis of gene or phenotype x environment interactions (Langthorne & McGill, 2008; Oliver, 1993; Tunnicliffe & Oliver, 2011). The elevated prevalence of self-injury in individuals with ASD can be explained through an interaction between ASD characteristics such as preference for routine, repetitive behaviours and sensory sensitivity, and the environment. These ASD characteristics would elevate the probability of frequently occurring events and stimuli being aversive or reinforcing to individuals with ASD, but not to other individuals with intellectual disabilities of heterogeneous aetiology, therefore occasioning more self-injury in ASD populations.

7.3.3.2 Temporal associations

The results regarding operant functions for self-injury in ASD were supported and extended by findings from fine grained temporal analysis of behaviours presented in Chapter 6. These findings revealed that for the majority of individuals with ASD, self-injury was significantly temporally associated with other behaviours in individuals' repertoires, supporting the assertion from the functional results that self-injurious behaviour in ASD does not occur randomly. Importantly some individuals evidenced significant temporal relationships between repetitive behaviours and self-injury, providing tangential support for the early development model of self-injury proposed by Guess and Carr (1991).

Clinically, this study provided important findings that for the majority of individuals with ASD, self-injury is reliably preceded by proto-imperative communicative behaviours. This finding is novel in cohorts of individuals with ASD, and supports data collected in individuals with intellectual disability of heterogeneous aetiology (Petty *et al.,* 2009). The close temporal association between self-injury and proto-imperative acts again supports the data collected in the study of operant function, and suggests that self-injury in ASD may often serve a communicative purpose. Critically, this finding implies that commonly employed interventions to replace challenging behaviour with a functionally equivalent response (Functional Communication Training, FCT; Carr & Durand, 1985) could be usefully employed in populations of individuals with ASD.

7.3.3.3 <u>Summary of function of self-injurious behaviour in autism spectrum disorder</u>

In summary, the results of this thesis demonstrated proof of principle; that self-injury can be maintained by environmental contingencies in children with ASD. Whilst this may not entail the ‘typical’ social functions of acquiring attention or escaping demands, this study demonstrated the operation of socially mediated functions associated with person characteristics of individuals with ASD. Implications from the study of temporal associations in Chapter 6 support the assertion that self-injury often serves a communicative function in children with ASD and interventions such as FCT may be usefully employed to reduce self-injury.

7.3.4 Model of self-injurious behaviour in autism spectrum disorder

A final aim of this thesis was to translate findings regarding the epidemiology, risk markers and function of self-injury into a dynamic model of self-injury in individuals with ASD which can be utilised to generate hypotheses for further research and indicate points of intervention. Given the utility of Guess and Carr's (1991) developmental stage model of self-injury, a similar set of stages are hypothesised as the basis of this model of self-injury. However, as identified in Chapter 1, whilst Guess and Carr's (1991) model has utility in explaining the development of self-injury, it does not account for differing levels of probability or trajectories of development that are influenced by person characteristics. Therefore, adaptations to this model will be made in order to account for the elevated prevalence of self-injury in ASD and the associations between health problems, repetitive behaviours, impaired behavioural control and self-injury.

Guess and Carr's model postulates three stages in the development and maintenance of self-injury. Stage One encompasses the occurrence of rhythmic repetitive behaviours in an individuals' behavioural repertoire. In Stage Two, Guess and Carr (1991) proposed that these repetitive behaviours become sensitive to arousal levels and function to maintain optimal arousal for the individual by increasing or decreasing the level of stimulation experienced by the individual. During Stage Three these behaviours become sensitive to environmental reinforcement and are shaped into increasingly severe behaviour. A baseline trajectory for the development of self-injury in individuals with intellectual disability is plotted in accordance with Guess and Carr's (1991) model. Repetitive behaviours occur and then become sensitive to internal states. Over time, these behaviours increase in severity and probability as they are shaped by the environment. The level of external social control over these behaviours increases from low to high as environmental reinforcement becomes more consistent. Figure 7.1 displays this model of self-injury in ASD diagrammatically, with the addition of the following modifications based on the findings of this thesis.

7.3.4.1 Modifications to the stages of the model

Modifications are proposed to Guess and Carr's original model at Stage Two. Additionally, a putative Stage Four is hypothesised.

Stage Two: Guess and Carr's (1991) original Stage Two encompassed the sensitisation of repetitive behaviours to arousal levels. The results of this thesis have indicated a potential role for self-injury in response to painful health conditions and therefore Stage Two has been extended to encompass behaviours becoming sensitive to *all internal states*, including pain. This addition allows for a functional interaction between self-injury, pain and painful health conditions at Stage Two of the model.

Stage Four: The results of this thesis support the inclusion of a putative Stage Four, following the establishment of environmental control in Stage Three. In Stage Four, it is proposed that environmental social control is less influential and self-injury is no longer wholly within the individual's control. During this stage, self-restraint behaviours are hypothesised to occur as the individual inhibits their self-injurious behaviour.

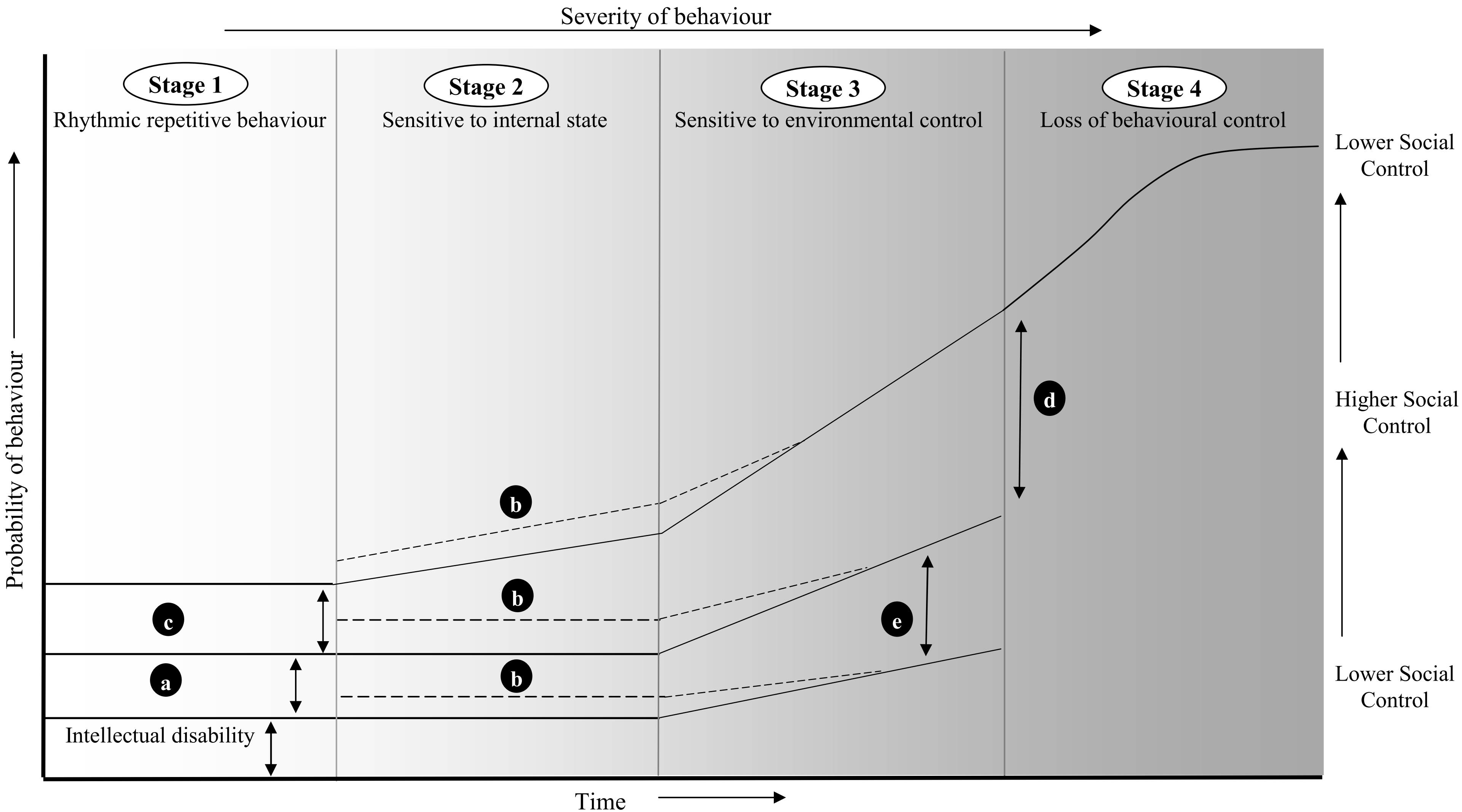


Figure 7.1 Hypothesised dynamic model of the development and maintenance of self-injury in ASD over time ('a': probability is raised due to 'ASD' impairments; 'b': probability of self-injury is raised and an alternative mechanism for the emergence of self-injury occurs due to the development of painful health conditions; 'c': probability is raised due to impairments in behavioural control; 'd' Impaired behavioural control drives self-injury, evidenced through self-restraint; 'e': both ASD weighted and typical reinforcement contingencies are active conditions).

7.3.4.2 Additions to the developmental trajectory of the model

From the delineation of risk markers and person characteristics associated with self-injury in ASD, three modifications to the slope of the trajectory of self-injury are proposed. These additions are associated with the presence of 'ASD' impairments, impairments in behavioural inhibition and the presence of painful health conditions.

'ASD' Impairments:

This thesis has demonstrated that the presence of 'ASD' impairments such as repetitive behaviours and deficits in social interaction, rather than a diagnosis of idiopathic autism, increase the risk of self-injury (results from Chapter 2). Therefore, a differing trajectory for the development of self-injury is hypothesised for individuals who have these characteristics. Individuals with ASD engage in high levels of repetitive behaviour (Estes *et al.,* 2011; Richler, Bishop, Kleinke & Lord, 2007; Turner, 1999), and therefore it is proposed that during Stage One, the probability of behaviour and the subsequent trajectory of development is already elevated above the intellectual disability trajectory (see point 'a' in Figure 7.1). This heightened trajectory remains stable during Stage Two as the behaviours become sensitive to internal states. In Stage Three (see point 'e' in Figure 7.1) as environmental reinforcement shapes the behaviour, it is hypothesised that the probability of self-injury in any typical environment in individuals with 'ASD' impairments is further heightened. Results from Chapter 5 demonstrated proof of principle that operant reinforcement paradigms delineating 'ASD weighted' contingencies account for self-injury in individuals with ASD. These results have been interpreted within a phenotype x environment framework in which establishing operations and specific antecedents are hypothesised to be more frequently occurring and more aversive for individuals with 'ASD' impairments. For example these individuals may have a lower tolerance for aversive noises *and* so find a higher proportion of typically tolerable noises aversive, resulting in more opportunities for noise to occasion self-injury.

Therefore, for individuals with 'ASD' impairments, there is both a higher probability of repetitive behaviours being shaped to self-injury given the heightened prevalence of repetitive behaviours *and* a higher probability of these self-injurious behaviours being shaped and maintained by frequently occurring environmental contingencies. Taken together the hypothesised trajectory accounts for the heightened probability of self-injurious behaviour in individuals with associated 'ASD' impairments, and charts the development of self-injury in these individuals using Guess and Carr's (1991) stage model.

Impaired Behavioural Control:

A second person characteristic and risk marker which the model must account for is the influence of impaired behavioural control on the developmental trajectory of self-injury. Impaired behavioural control, as evidenced by ADHD type behaviours (Barkley, 1997) is hypothesised to effect the development of self-injury at all stages of the model. In Stage One, it is hypothesised that impaired behavioural control would lead to a heightened prevalence of repetitive behaviours (see point 'c' in Figure 7.1), as repetitive behaviours are understood to be directly underpinned by impairments in executive functioning (Turner, 1999). During Stage Two of the model, behaviour becomes sensitive to internal state and functions to regulate this. Barkley (1997) proposed that impaired behavioural control leads to behaviours being initiated at a low threshold in the presence of discriminative stimuli. Thus, as behaviour becomes sensitive to internal state, this prepotent response becomes increasingly difficult for the individual to inhibit in response to the internal state. Therefore, the probability and consequently developmental trajectory of self-injury is elevated in individuals with impaired behavioural control during Stage Two. Similarly, during Stage Three, prepotent responses to environmental antecedents are difficult to inhibit and thus self-injury is initiated more and more frequently.

It is hypothesised that for individuals with impaired behavioural control, it eventually becomes impossible to fully inhibit these prepotent responses and the individual gradually ‘loses control’ over their self-injury. At this stage, the individual transitions into the final stage of the model (see point ‘d’ in Figure 7.1) and may develop self-restraint behaviours as a means of controlling their self-injury. For these individuals it is proposed that whilst environmental contingencies may still be active, self-injury is no longer wholly controlled by these contingencies. It is likely that at this point, alternative intervention strategies may need to be employed, rather than the typical behavioural interventions which are effective when environmental contingencies are active. The developmental trajectory for individuals with impaired behavioural control is therefore steepest and potentially most worrying.

Painful Health Conditions:

A final risk marker for self-injury identified in this thesis was that of painful health conditions. However, in contrast to ‘ASD’ impairments, it is hypothesised that painful health conditions do not influence the trajectory of self-injury in all stages of Guess and Carr’s (1991) original model. Instead it is hypothesised that painful health conditions provide a second pathway for self-injurious behaviour to enter into an individual’s behavioural repertoire. This occurs at Stage Two of the model, where behaviour is sensitive to internal states, and is indicated by points ‘b’ on Figure 7.1. It is hypothesised that the experience of painful health conditions leads to a heightened probability of individuals engaging in behaviour in an attempt to remove or ‘gate’ the painful experience (Melzack & Wall, 1965). Thus, the trajectory for the development of self-injury is heightened. However, the effect of painful health conditions is proposed to be intermittent throughout development, as unlike ‘ASD’ impairments and impaired behavioural inhibition, painful health conditions may occur acutely and then remit. Therefore, the trajectory of self-injury development given the presence of painful health condition is represented using an intermittent line over and above each of the previously described trajectories. Once established in an individual’s behavioural repertoire, these behaviours can then progress to be shaped by the environment in Stage Three of the model.

Similar to the argument presented about ‘ASD’ impairments, it is hypothesised that the presence of pain also increases the probability of self-injury in response to environmental antecedents. Pain is described as a setting event (Carr *et al.,* 2003; Carr & Blakeley-Smith, 2006; Carr & Smith, 1995), which, when present, leads to an increased proportion of antecedents being, for example, aversive for an individual. Therefore, painful health conditions in Stage Three of the model are hypothesised to increase the trajectory of self-injury. Thus, the hypothesised trajectory and influence of painful health conditions upon the development of self-injury can be seen to account for the data presented in this thesis, revealing painful health conditions to increase the risk of self-injury in individuals with ASD.

7.3.4.3 Summary of the model

In summary, this model demonstrates how person characteristics can alter the initial probability and trajectory of the development and maintenance of self-injury, utilising components of the original model proposed by Guess and Carr (1991). Individuals can be seen to accrue risk markers which may then alter the hypothesised developmental trajectory of self-injury. From this, it can be seen that individuals with 'ASD' impairments *and* painful health conditions *and* impaired behavioural control may evidence the highest probability/risk of self-injury and the steepest gradient of developmental trajectory. For these individuals, repetitive behaviours in Stage One are more likely, self-injury can develop in Stage Two via two pathways (repetitive behaviour and health problems), environmental antecedents are likely to be more frequent and more aversive in Stage Three and these individuals are most at risk of progressing into Stage Four where self-injury is no longer under environmental control. Thus, this model accounts for the prevalence, risk markers, and operant influence of self-injury in individuals with ASD identified in this thesis.

Whilst many of the hypotheses in this model are supported by the data collected in the thesis, it is acknowledged that some aspects of this model have yet to be supported by data. However, this model is intended to be both a synthesis of existing and new findings and generative; to lead to future research which can test and evaluate the hypotheses and implications posited by the model.

7.4 Limitations and Strengths of the Research

Whilst the findings in this thesis are novel and robust, a number of limitations to the conclusions and hypothesised model must be acknowledged. Firstly, whilst neurobiological theories of self-injury were delineated in Chapter 1, these theories were not evaluated in the empirical research in this thesis. Thus, the final proposed model of self-injury in ASD, presented in Figure 7.1, is unable to identify and include a role for neurobiology. This limitation is broadly representative of two divergent streams of research in challenging behaviour more generally. Whilst behavioural and biological research continue to be separate, comprehensive models of self-injury will lack a balanced evaluation of the potential contributions of both mechanisms.

Secondly, throughout the thesis, self-injury was defined by topography rather than by actual physical harm. Thus, all studies may have inadvertently included behaviours which have the capacity to result in injury, but do not currently result in actual physical harm. However, all studies employed detailed descriptions of the specific topographies of self-injury which helped to operationalise the definition. Given that all of these topographies necessarily indicated some form of contact that could potentially lead to physical harm (e.g., hits self with object; bites self) it was felt that this method of defining self-injury would be suitably robust. Additionally, necessitating observable physical harm in a definition of self-injury may lead to some potentially severe behaviours, such as head banging, being excluded from data as physical harm may be hidden from sight, e.g., below hair lines, or physical harm may be unobservable e.g., internal injury.

Finally, throughout the thesis there was a consistent trade off between depth of measurement and sample size. Whilst the survey studies had substantial samples sizes, assessment of ASD phenomenology and ability level was necessarily reliant upon informant report questionnaires. Conversely, the utilisation of standardised measures such as the Autism Diagnostic Observation Schedule (Lord *et al.,* 2000), Vineland Adaptive Behavior Scales (Sparrow, David & Cicchetti, 1985) and Mullen Scales of Early Learning (Mullen, 1995) in Chapters 5 and 6 may have improved the external validity of the studies, but did limit the possible sample sizes due to the time consuming nature of the assessments. Overall, it was felt that an appropriate compromise was reached across the whole of the thesis, through the combination of large scale survey studies and smaller, more detailed investigations of self-injury. The inclusion of both of these methodologies allowed for both robust assessments of person characteristics and the collection of datasets large enough to enable statistical modelling of putative risk markers.

Despite the limitations identified above, the reliability and validity of the findings reported in this thesis are robust. Throughout the thesis, attempts were made to delineate the characteristics of the samples, particularly regarding ASD phenomenology and intellectual functioning, as this was highlighted as a key limitation of previous research in Chapter 1. Additionally, the utilisation of a wide range of research techniques allowed for a comprehensive examination of self-injury in individuals with ASD. Conducting research at both macro and micro levels enabled an understanding of both the size and scale of the problem of self-injury, and also a more fine grained analysis of the presentation and function of self-injury in children with ASD.

A key strength of the research undertaken in this thesis is that it was motivated by clinical need and has identified novel data in response to this need. Despite the elevated prevalence of self-injury in ASD, little focused research had been conducted in this population. Thus, the results from this thesis regarding the prevalence and persistence of self-injury, associated person characteristics such as overactivity, impulsivity and painful health conditions, and the delineation of ASD weighted operant functions, contribute significantly to the research literature. Additionally, the generation of a novel model of self-injury from these data will enable future hypothesis driven research to continue in ASD populations.

7.5 Future Directions

As a result of the research in this thesis and the proposed model of self-injury in ASD, a number of key areas for future research can be identified. Firstly, a longitudinal study beginning at the earliest stage of the development of self-injury in ASD is warranted. This research should evaluate the role of pain and painful health conditions specifically, and the possible evolution of repetitive behaviours. This study would require careful methodological considerations, recruiting a substantial well documented sample of children with ASD at a very young age, in order to track the emergence and development of self-injury. Recent advances in early diagnosis of ASD may make sample identification and recruitment more plausible. The methods used in Chapter 6, for fine grained temporal analysis of associations between behaviours could be utilised in order to evaluate the evolution of repetitive behaviours over time. Regular simple health checks and monitoring of new health symptoms could also form part of the protocol. Given the potential interaction between health conditions and the emergence of self-injury, cross discipline research led by psychological and medical professionals may produce the most robust and reliable results.

In addition to highlighting the role of health problems and pain in self-injury, this thesis has identified an important potential role for impaired behavioural control in the development and maintenance of self-injury. However, inhibitory control is assessed indirectly as evidenced through ADHD type behaviours and self-restraint. Whilst this assumption has good face validity, it is critical that behavioural control is assessed directly in individuals with ASD who evidence self-injury. As reliable and robust measures of inhibitory control have been developed within executive functioning batteries in cognitive psychology, these measures could be readily applied to test and control groups of individuals with and without self-injury (e.g., The Test of Everyday Attention for Children, TEA-Ch, Manly, Robertson, Anderson, &

Nimmo-Smith, 1999; Delis-Kaplin Executive Function Scale, D-KEFS, Lopez, Lincoln, Ozonoff; Lai, 2005). Specifically, matched samples such as those identified in Chapter 2, with higher levels of functioning and either present or absent self-injury could be readily assessed in order to evaluate the relationship between behavioural inhibition and self-injury. However, as these measures are predominantly developed for individuals of typical development, alternative approaches may be required to assess inhibitory control in individuals with ASD and associated intellectual disability. Some developments have been made in creating assessments such as the 'Gift Delay' inhibition task and 'Object Retrieval' conflict inhibition task (Diamond, 1990; Kochanska, Murray & Harlan, 2000). Therefore, future research could evaluate the association between inhibitory control and self-injury in both intellectually disabled and higher functioning populations of individuals with ASD.

Finally, given the significant clinical implications of highly prevalent and persistent self-injury, research must now turn towards evaluating a comprehensive intervention strategy for self-injury in ASD. As discussed in Chapter 3, an early intervention approach may produce the most effective results. The findings in this thesis have highlighted individual characteristics which could help identify those individual's with ASD who are most at risk of engaging in persistent self-injury. The findings also indicate that any intervention approach should assess and target three key areas; pain and discomfort, operant influence and interactions with ASD characteristics, and behavioural inhibition. This 'triage' approach may see different types of intervention being utilised with different children, depending upon their profile of self-injury. Importantly, the strategies needed for the majority of these interventions are already widely used (e.g., simple health interventions, applied behaviour analysis techniques). However, what remains to be trialled is a consistent and structured application of these techniques, targeted to 'at risk' children at an early age.

7.6 Closing Summary

> "The outcome of any serious research can only be to make two questions grow where only one grew before" (Thorstein Veblen).

As this quote suggests, research has a tendency to create novel and interesting questions exponentially. However, whilst this thesis has generated many new research ideas, it has also given robust answers to some critical questions about the prevalence, persistence and function of self-injury in ASD. These findings show that self-injury is prevalent, persistent and often sensitive to identifiable environmental reinforcement. Thus, these findings indicate that intervention for self-injury in ASD is both necessary and, potentially, straight forward, inasmuch as interventions are readily available; drawing upon existing evidence based behavioural techniques and including thorough investigation of potential health conditions. Whilst further theoretical questions can always be posed, attention must be given to ensuring that existing data, and the pragmatic implications of these data, are translated effectively into clinical practice. Given that this thesis was broadly motivated by current clinical need, it is imperative that the direction of any future work remains focused upon reducing this clinical need, whilst acknowledging that further theoretical questions can, and should, be generated. In ensuring that clinical need and theoretical understanding remain closely entwined, successful research advances can be made which will improve the lives of carers, families and individuals with ASD.

REFERENCES

Allen, D., Lowe, K., Brophy, S., & Moore, K. (2009). Predictors of restrictive reactive strategy use in people with challenging behaviour. *Journal of Applied Research in Intellectual Disabilities, 22,* 159-168.

Amador-Campos, J. A., Forns-Santacana, M., Guardia-Olmos, J., & Pero-Cebollero, M. (2006). DSM-IV attention deficit hyperactivity disorder symptoms: Agreement between informants in prevalence and factor structure at different ages. *Journal of Psychopathology and Behavioral Assessment, 28,* 23-32.

American Psychiatric Association. Diagnostic and statistical manual of mental disorders. 4th edition. Washington, DC: American Psychiatric Association; 1994

Anderson, C. M. & Long, E. S. (2002). Use of a structured descriptive assessment methodology to identify variables affecting problem behavior. *Journal of Applied Behavior Analysis, 35,* 137-154.

Ando, H. & Yoshimura, I. (1979a). Speech skill levels and prevalence of maladaptive behaviors in autistic and mentally retarded children. *Child Psychiatry and Human Development, 10,* 85-90.

Ando, H. & Yoshimura, I. (1979b). Comprehension skill levels and prevalence of maladaptive behaviors in autistic and mentally retarded children. *Child Psychiatry and Human Development, 10,* 85-90.

Arron, K., Oliver, C., Moss, J., Berg, K., & Burbidge, C. (2011). The prevalence and phenomenology of self-injurious and aggressive behaviour in genetic syndromes. *Journal of Intellectual Disability Research, 55,* 109-120.

Baer, D. M. (1982). The imposition of structure on behavior and the demolition of behavior structures. In H. E. Howe (Ed.), *Nebraska Symposium on Motivation*. Lincoln: University of Nebraska Press.

Baghdadli, A., Pascal, C., Grisi, S., & Aussilloux, C. (2003). Risk factors for self-injurious behaviours among 222 young children with autistic disorders. *Journal of Intellectual Disability Research, 47,* 622-627.

Baghdadli, A., Picot, M. C., Pry, R., Michelon, C., Burzstejn, C., Lazartigues, A. *et al.* (2008). What factors are related to a negative outcome of self-injurious behaviour during childhood in pervasive developmental disorders? *Journal of Applied Research in Intellectual Disabilities, 21,* 142-149.

Baird, G., Simonoff, E., Pickles, A., Chandler, S., Loucas, T., Meldrum, D. *et al.* (2006). Prevalence of disorders of the autism spectrum in a population cohort of children in South Thames: the Special Needs and Autism Project (SNAP). *Lancet, 368,* 210-215.

Barkley, R. A. (1997). *ADHD and the nature of self-control.* New York, London: The Guilford Press.

Barrett, R. P., Feinstein, C., & Hole, W. T. (1989). Effects of Naloxone and Naltrexone on Self-Injury - A Double-Blind, Placebo-Controlled Analysis. *American Journal on Mental Retardation, 93,* 644-651.

Bartak, L. & Rutter, M. (1976). Difference between mentally retarded and normally intelligent autistic children. *Journal of Autism and Childhood Schizophrenia, 6,* 109-120.

Baumeister, A. A., Frye, G. & Schroeder, S. R. (1985). Neurochemical correlates of self-injurious behaviour. In Mulick, J. A. and Mallory, B. L (Eds.). *Transitions in Mental Retardation: Advocacy, Technology and Science.* Nowood, NJ: Ablex Publishing Corporation.

Beadle-Brown, J., Murphy, G., & DiTerlizzi, M. (2009). Quality of life for the Camberwell Cohort. *Journal of Applied Research in Intellectual Disabilities, 22,* 380-390.

Berg, K., Arron, K., Burbidge, C., Moss, J., & Oliver, C. (2007). Carer-reported contemporary health problems in people with severe and profound intellectual disability and genetic syndromes. *Journal of Policy and Practice in Intellectual Disabilities, 4,* 120-128.

Berument, S. K., Rutter, M., Lord, C., Pickles, A., & Bailey, A. (1999). Autism screening questionnaire: diagnostic validity. *British Journal of Psychiatry, 175,* 444-451.

Bhaumik, S., Branford, D., McGrother, C., & Thorp, C. (1997). Autistic traits in adults with learning disabilities. *The British Journal of Psychiatry, 170,* 502-506.

Bijou, S. W. & Baer, D. M. (1978). *Behavior analysis of child development.* Englewood Cliffs, NJ: Prentice-Hall.

Billstedt, E., Gillberg, C., & Gillberg, C. (2005). Autism after adolescence: Population-based 13- to 22-year follow up study of 120 individuals with autism diagnosed in childhood. *Journal of Autism and Developmental Disorders, 35,* 351-360.

Bishop, D. V. M. & Norbury, C. F. (2002). Exploring the borderlands of autistic disorder and specific language impairment: a study using standardised diagnostic instruments. *Journal of Child Psychology and Psychiatry and Allied Disciplines, 43,* 917-929.

Blacher, J. & McIntyre, L. L. (2006). Syndrome specificity and behavioural disorders in young adults with intellectual disability: Cultural differences in family impact. *Journal of Intellectual Disability Research, 50,* 184-198.

Blakeley-Smith, A., Carr, E. G., Cale, S. I., & Owen-DeSchryver, J. S. (2009). Environmental fit: A model for assessing and treating problem behavior associated with curricular difficulties in children with autism spectrum disorders. *Focus on Autism and Other Developmental Disabilities, 24,* 131-145.

Blanks, R. G., Moss, S. M., McGahan, C. E., Quinn, M. J., & Babb, P. J. (2000). Effect of NHS breast screening programme on mortality from breast cancer in England and Wales, 1990-8: comparison of observed with predicted mortality. *British Medical Journal, 321,* 665-669.

Bodfish, J. W., Crawford, T. W., Powell, S. B., Parker, D. E., Golden, R. N., & Lewis, M. H. (1995). Compulsions in adults with mental retardation: Prevalence, phenomenology, and comorbidity with stereotypy and self-injury. *American Journal on Mental Retardation, 100,* 183-192.

Bodfish, J. W., Symons, F. J., Parker, D. E., & Lewis, M. H. (2000). Varieties of repetitive behavior in autism: Comparisons to mental retardation. *Journal of Autism and Developmental Disorders, 30,* 237-243.

Borrero, C. S. W. & Borrero, J. C. (2008). Descriptive and experimental analyses of potential precursers to problem behavior. *Journal of Applied Behavior Analysis, 41,* 83-96.

Bosch, J., Van Dyke, D. C., Smith, S. M., & Poulton, S. (1997). Role of medical conditions in exacerbation of self-injurious behavior: An exploratory study. *Mental Retardation, 35,* 124-130.

Bradley, E. A., Summers, J. A., Wood, H. L., & Bryson, S. E. (2004). Comparing rates of psychiatric and behavior disorder in adolescents and young adults with severe intellectual disability with and without autism. *Journal of Autism and Developmental Disorders, 34,* 151-161.

Breau, L., Camfield, C. S., McGrath, P. J., & Finley, A. (2003). The incidence of pain in children with severe cognitive impairments. *Archives of Pediatrics and Adolescent Medicine, 157,* 1219-1226.

Breau, L., Finley, A., McGrath, P. J., & Camfield, C. S. (2002). Validation of the non-communicating children's pain checklist - Postoperative version. *Anesthesiology, 96,* 528-535.

Brereton, A. V., Tonge, B. J., & Einfeld, S. L. (2006). Psychopathology in children and adolescents with autism compared to young people with intellectual disability. *Journal of Autism and Developmental Disorders, 36,* 863-870.

Buckley, S. D. & Newchok, D. K. (2006). Analysis and treatment of problem behavior evoked by music. *Journal of Applied Behavior Analysis, 39,* 141-144.

Burbidge, C., Oliver, C., Moss, J., Arron, K., Berg, K., Furniss, F. *et al.* (2010). The association between repetitive behaviours, impulsivity and hyperactivity in people with intellectual disability. *Journal of Intellectual Disability Research, 54,* 1078-1092.

Burns, G. L., Keortge, S. G., Formea, G. M., & Sternberger, L. G. (1996). Revision of the Padua Inventory of obsessive compulsive disorder symptoms: Distinctions between worry, obsessions, and compulsions. *Behaviour Research and Therapy, 34,* 163-173.

Carr, E. G. & Owen-DeSchryver, J. S. (2007). Physical illness, pain and problem behavior in minimally verbal people with developmental disabilities. *Journal of Autism and Developmental Disorders, 37,* 413-424.

Carr, E. G. & Smith, C. E. (1995). Biological setting events for self-injury. Mental Retardation and Developmental Disabilities 1, 94-98.

Carr, E. G., Smith, C. E., Giacin, T. A., Whelan, B. M., & Pancari, J. (2003). Menstrual discomfort as a biological setting event for severe problem behavior: Assessment and intervention. *American Journal on Mental Retardation, 108,* 117-133.

Carr, E. G. & Blakeley-Smith, A. (2006). Classroom intervention for illness-related problem behavior in children with developmental disabilities. *Behavior Modification, 30,* 901-924.

Carr, E. G. & Durand, V. M. (1985). Reducing Behavior Problems Through Functional Communication Training. *Journal of Applied Behavior Analysis, 18,* 111-126.

Chadwick, O., Piroth, N., Walker, J., Bernard, S., & Taylor, E. (2000). Factors affecting the risk of behaviour problems in children with severe intellectual disability. *Journal of Intellectual Disability Research, 44,* 108-123.

Charach, A., Chen, S., Hogg-Johnson, S., & Schachar, R. J. (2009). Using the Conners' Teacher Rating Scale-Revised in School Children Referred for Assessment. *Canadian Journal of Psychiatry, 54,* 232-241.

Christensen, T. J., Ringdahl, J. E., Bosch, J., Falcomata, T. S., Luke, J. R., & Andelman, M. S. (2009). Constipation associated with self-injurious and aggressive behavior exhibited by a child diagnosed with autism. *Education and Treatment of Children, 32,* 89-103.

Christie, R., Bay, C., Kaufman, I. A., Bakay, B., Borden, M. & Nyhan, W. L. (1982). Lesch-Nyan disease: clinical experience with nineteen patients. *Developmental Medicine and Child Neurology, 24,* 293-306.

Cliff, N. (1993). Dominance Statistics - Ordinal Analyses to Answer Ordinal Questions. *Psychological Bulletin, 114,* 494-509.

Cohen, D., Pichard, N., Tordjman, S., Baumann, C., Burglen, L., Excoffier, E. *et al.* (2005). Specific Genetic Disorders and Autism: Clinical Contribution Towards their Identification. *Journal of Autism and Developmental Disorders, 35,* 103-116.

Cohen, H., merine-Dickens, M., & Smith, T. (2006). Early intensive behavioral treatment: Replication of the UCLA model in a community setting. *Journal of Developmental and Behavioral Pediatrics, 27,* S145-S155.

Cohen, I., Tsiouris, J., Flory, M., Kim, S. Y., Freedland, R., Heaney, G. *et al.* (2010). A Large Scale Study of the Psychometric Characteristics of the IBR Modified Overt Aggression Scale: Findings and Evidence for Increased Self-Destructive Behaviors in Adult Females with Autism Spectrum Disorder. *Journal of Autism and Developmental Disorders, 40,* 599-609.

Collacott, R. A., Cooper, S. A., Branford, D., & McGrother, C. (1998). Epidemiology of self-injurious behaviour in adults with learning disabilities. *The British Journal of Psychiatry, 173,* 428-432.

Colley, A., Leversha, M., Voullaire, L. & Rogers, J. (1990). Five cases demonstrating the distinctive behavioural features of chromosomal deletion 17 (p11.2p11.2) (Smith-Magenis syndrome). *Journal of Paediatrics and Child Health, 26* 17-21.

Conners, J., Iwata, B. A., Kahng, S., Hanley, G. P., Wordsell, A. S., & Thomson, R. H. (2000). Differential responding in the presence and absence of discriminative stimuli during multi-element functional analyses. *Journal of Applied Behavior Analysis, 33,* 299-308.

Cooper, S. A., Smiley, E., Allan, L. M., Jackson, A., Finlayson, J., Mantry, D. *et al.* (2009). Adults with intellectual disabilities: Prevalence, incidence and remission of self-injurious behaviour, and related factors. *Journal of Intellectual Disability Research*.

Cooper, S. A. & van der Speck, R. (2009). Epidemiology of mental ill health in adults with intellectual disabilities. *Current Opinion in Psychiatry, 22,* 431-436.

Cowley, A., Newton, J., Sturmey, P., Bouras, N., & Holt, G. (2005). Psychiatric inpatient admissions of adults with intellectual disabilities: Predictive factors. *American Journal on Mental Retardation, 110,* 216-225.

Danquah, A., Limb, K., Chapman, M., Burke, C., Flood, A., Gore, S. *et al.* (2009). An investigation of the factors predictive of continued self-injurious behaviour in an intellectual disability service. *Journal of Applied Research in Intellectual Disabilities*.

Davies, L. (2010). Challenging behaviour in children with severe intellectual disabilities: identification and characteristics of those at high risk. Unpublished doctoral dissertation, University of Birmingham, Birmingham, UK.

de Winter, C. F., Jansen, A. A. C., & Evenhuis, H. M. (2011). Physical conditions and challenging behaviour in people with intellectual disability: a systematic review. *Journal of Intellectual Disability Research, 55,* 675-698.

Devlin, S., Healy, O., Leader, G., & Reed, P. (2008). The analysis and treatment of problem behavior evoked by auditory stimulation. *Research in Autism Spectrum Disorders, 2,* 671-680.

Diamond, A. (1990). Developmental time course in infants and infant monkeys, and the neural bases of inhibitory control in reaching. In A. Diamond (Ed.), *The development and neural bases of higher cognitive functions* (Annals of the New York Academy of Sciences Vol. 608, pp. 637–676). New York: New York Academy of Sciences.

Dominick, K. C., Davis, N. O., Lainhart, J., Tager-Flusberg, H., & Folstein, S. (2007). Atypical behaviors in children with autism and children with a history of language impairment. *Research in Developmental Disabilities, 28,* 145-162.

Dykens, E. M. & Hodapp, R. M. (2001). Research in mental retardation: toward an etiologic approach. *Journal of child psychology and psychiatry and allied disciplines, 42*, 49-71.

Dykens, E.M. & Volkmar, F. (1997). Medical Conditions Associated with Autism. In Cohen, D. & Volkmar, F. (Eds.) *Autism and Pervasive Developmental Disorders* (2nd Edition). NY: John Wiley & Sons, Inc.

Eikeseth, S., Smith, T., Jahr, E., & Eldevik, S. (2007). Outcome for children with autism who began intensive behavioral treatment between ages 4 and 7 - A comparison controlled study. *Behavior Modification, 31,* 264-278.

Emerson, E., Alborz, A., Reeves, D., Mason, H., Swarbrick, R., Kiernan, C. *et al.* (1997). The HARC challenging behaviour project. Report 2. The prevalence of challenging behaviour, Hester Adrian Research Centre, University of Manchester, Manchester.

Emerson, E. (1998). Working with people with challenging behaviour. In: Emerson, E., Hatton, C., Bromley, J. & Caine, A (Eds). *Clinical Psychology and People with Intellectual Disability* , 128-153. Wiley: Chichester.

Emerson, E., Kiernan, C., Alborz, A., Reeves, D., Mason, H., Swarbrick, R. *et al.* (2001a). The prevalence of challenging behaviors: a total population study. *Research in Developmental Disabilities, 22,* 77-93.

Emerson, E., Kiernan, C., Alborz, A., Reeves, D., Mason, H., Swarbrick, R. *et al.* (2001b). Predicting the persistence of severe self-injurious behavior. *Research in Developmental Disabilities, 22,* 67-75.

Esbensen, A. J., Seltzer, M. M., Lam, K. S. L., & Bodfish, J. W. (2009). Age related differences in restricted repetitive behaviors in autism spectrum disorders. *Journal of Autism and Developmental Disorders, 39,* 57-66.

Estes, A., Shaw, D. W. W., Sparks, B. F., Friedman, S., Giedd, J. N., Dawson, G. *et al.* (2011). Basal ganglia morphometry and repetitive behavior in young children with autism spectrum disorder. *Autism Research, 4,* 212-220.

Fleiss, J. L. (1981). *Statistical Methods for Rates and Proportions*. John Wiley & Sons, Inc., New York.

Forman, D., Hall, S., & Oliver, C. (2002). Descriptive analysis of self-injurious behaviour and self-restraint. *Journal of Applied Research in Intellectual Disabilities, 15,* 1-7.

Fovel, J. T., Lash, P. S., Barron, D. A., & Roberts, M. S. (1989). A survey of self-restraint, self-injury, and other maladaptive behaviors in an institutionalized retarded population. *Research in Developmental Disabilities, 10,* 377-382.

Gillberg, C. & Billstedt, E. (2000). Autism and Asperger syndrome: coexistence with other clinical disorders. *Acta Psychiatrica Scandinavica, 102,* 321-330.

Grant, J. E. & Potenza, M. N. (2006). Compulsive aspects of impulse-control disorders. *Psychiatric Clinics of North America, 29,* 539-+.

Greenberg, J. S., Seltzer, M. M., Hong, J., & Orsmond, G. I. (2006). Bidirectional effects of expressed emotion and behavior problems and symptoms in adolescents and adults with autism. *American Journal on Mental Retardation, 111,* 229-249.

Griffin, J. C., Williams, D. E., Stark, M. T., Altmeyer, B. K., & Mason, M. (1986). Self-injurious behavior: A state-wide prevalence survey of the extent and circumstances. *Applied Research in Mental Retardation, 7,* 105-116.

Griffith, G., Hastings, R., Nash, S., & Hill, C. (2010). Using matched groups to explore child behavior problems and maternal well-being in children with Down syndrome and autism. *Journal of Autism and Developmental Disorders, 40,* 610-619.

Guess, D. & Carr, E. G. (1991). Emergence and maintenance of stereotypy and self-injury. *American Journal on Mental Retardation, 96,* 299-319.

Hagopian, L. P., Bruzek, J. L., Bowman, L. G., & Jennett, H. K. (2007). Assessment and treatment of problem behavior occasioned by interruption of free-operant behavior. *Journal of Applied Behavior Analysis, 40,* 89-103.

Hagopian, L. P., Wilson, D. M., & Wilder, D. A. (2001). Assessment and treatment of problem behavior maintained by escape from attention and access to tangible items. *Journal of Applied Behavior Analysis, 34,* 229-232.

Hagopian, L. P., Paclawskyj, T. R., & Kuhn, S. C. (2005). The use of conditional probability analysis to identify a response chain leading to the occurrence of eye poking. *Research in Developmental Disabilities, 26,* 393-397.

Hall, S. (2005). Comparing descriptive, experimental and informant-based assessments of problem behaviors. *Research in Developmental Disabilities, 26,* 514-526.

Hall, S., Lightbody, A. A., & Reiss, A. L. (2008a). Compulsive, Self-injurious and autistic behavior in children and adolescents with Fragile X syndrome. *American Journal on Mental Retardation, 113,* 44-53.

Hall, S., Oliver, C., & Murphy, G. (2001). Early development of self-injurious behaviour: An empirical study. *American Journal on Mental Retardation, 106,* 189-199.

Hall, S. & Oliver, C. (1992). Differential-Effects of Severe Self-Injurious-Behavior on the Behavior of Others. *Behavioural Psychotherapy, 20,* 355-365.

Hall, S., Thorns, T., & Oliver, C. (2003). Structural and environmental characteristics of stereotyped behaviors. *American Journal on Mental Retardation, 108,* 391-402.

Hall, S., Arron, K., Sloneem, J., & Oliver, C. (2008b). Health and sleep problems in Cornelia de Lange Syndrome: a case control study. *Journal of Intellectual Disability Research, 52,* 458-468.

Hanley, G. P., Iwata, B. A., & McCord, B. E. (2003). Functional analysis of problem behaviour: A review. *Journal of Applied Behavior Analysis, 36,* 147-185.

Happe, F. (1994). Autism: An introduction to psychological theory. UCL Press: London.

Harvey, S. T., Boer, D., Meyer, L. H., & Evans, I. M. (2009). Updating a meta-analysis of intervention research with challenging behaviour: Treatment validity and standards practice. *Journal of Intellectual and Developmental Disability, 34,* 67-80.

Hastings, R. P. (2003). Child behaviour problems and partner mental health as correlate of stress in mothers and fathers of children with autism. *Journal of Intellectual Disability Research, 47,* 231-237.

Hastings, R. P. & Brown, T. (2002). Coping strategies and the impact of challenging behaviors on special educators burnout. *Mental Retardation, 40,* 148-156.

Hausman, N., Kahng, S., Farrell, E., & Mongeon, C. (2009). Idiosyncratic functions: Severe problem behavior maintained by access to ritualistic behaviors. *Education and Treatment of Children, 32,* 77-87.

Haveman, M., Van Berkum, G., Holscher, P., & Van Walleghem, M. (2000). Needs for services expressed by parents with young intellectually disabled children: A comparison of results in Germany, the Netherlands and Belgium. *Journal of Intellectual Disability Research, 44,* 471.

Hayes, S., McGuire, B., O'Neill, M., Oliver, C., & Morrison, T. (2011). Low mood and challenging behaviour in people with severe and profound intellectual disabilities. *Journal of Intellectual Disability Research, 55,* 182-189.

Healey, J. J., Ahearn, W. H., Graff, R. B., & Libby, M. E. (2001). Extended analysis and treatment of self-injurious behavior. *Behavioral Interventions, 16,* 181-195.

Herscovitch, B., Roscoe, E. M., Libby, M. E., Bourret, J. C., & Ahearn, W. H. (2009). A Procedure for Identifying Precursors to Problem Behavior. *Journal of Applied Behavior Analysis, 42,* 697-702.

Hertz-Picciotto, I. & Delwiche, L. (2009). The Rise in Autism and the Role of Age at Diagnosis. *Epidemiology, 20,* 84-90.

Hill, J. & Furniss, F. (2006). Patterns of emotional and behavioural disturbance associated with autistic traits in young people with severe intellectual disabilities and challenging behaviours. *Research in Developmental Disabilities, 27,* 517-528.

Holden, B. & Gitlesen, J. P. (2006). A total population study of challenging behaviour in the county of Hedmark, Norway: Prevalence, and risk markers. *Research in Developmental Disabilities, 27,* 456-465.

Horner, R. H. & Day, H. M. (1991). The Effects of Response Efficiency on Functionally Equivalent Competing Behaviors. *Journal of Applied Behavior Analysis, 24,* 719-732.

Howells, G. (1986). Are the Medical Needs of Mentally-Handicapped Adults Being Met. *Journal of the Royal College of General Practitioners, 36,* 449-453.

Howlin, P. & Karpf, J. (2004). Using the Social Communication Questionnaire to identify 'autistic spectrum' disorders associated with other genetic conditions - Findings from a study of individuals with Cohen syndrome. *Autism, 8,* 175-182.

Hutt, C. & Hutt, S. J. (1965). Effects of Environmental Complexity on Stereotyped Behaviours of Children. *Animal Behaviour, 13,* 1-&.

Hyman, P., Oliver, C., & Hall, S. (2002). Self-injurious behavior, self-restraint and compulsive behaviours in Cornelia de Lange syndrome. *American Journal on Mental Retardation, 107,* 146-154.

World Health Organisation. (1992). ICD-10 Classifications of Mental and Behavioural Disorder: Clinical Descriptions and Diagnostic Guidelines. Geneva. World Health Organisation.

Iwata, B. A., Dorsey, M. F., Slifer, K. J., Bauman, K. E., & Richman, G. S. (1994). Toward A Functional-Analysis of Self-Injury. *Analysis and Intervention in Developmental Disabilities, 2,* 3-20.

Jackson, L., Kline, A. D., Barr, M. A., & Koch, S. (1993). Delange-Syndrome - A Clinical Review of 310 Individuals. *American Journal of Medical Genetics, 47,* 940-946.

Janicki, M. P. & Jacobson, J. W. (1983). Selected Clinical-Features and Service Characteristics of Autistic Adults. *Psychological Reports, 52,* 387-390.

Jansen, D. E. M. C., Krol, B., Groothoff, J. W., & Post, D. (2004). People with intellectual disability and their health problems: a review of comparative studies. *Journal of Intellectual Disability Research, 48,* 93-102.

Jennett, H., Hagopian, L. P., & Beaulieu, L. (2011). Analysis of heart rate and self-injury with and without restraint in an individual with autism. *Research in Autism Spectrum Disorders, 5,* 1110-1118.

Kawamura, Y., Takahashi, O., & Ishii, T. (2008). Reevaluating the incidence of pervasive developmental disorders: Impact of elevated rates of detection through implementation of an integrated system of screening in Toyota, Japan. *Psychiatry and Clinical Neurosciences, 62,* 152-159.

Kennedy, C. H. (2002). Evolution of stereotopy into self-injury. In S.R.Schroeder, M. L. Oster-Granite, & T. Thompson (Eds.), *Self-injurious behavior: Gene-brain-behavior relationships* (pp. 133-143).

Kientz, M. A. & Dunn, W. (1997). A comparison of the performance of children with and without autism on the sensory profile. *American Journal of Occupational Therapy, 51,* 530-537.

Knapp, M., Comas-Herrera, A., Astin, J., Beecham, J., & Pendaries, C. (2005). Intellectual disability, challenging behaviour and cost in care accommodation: what are the links? *Health & Social Care in the Community, 13,* 297-306.

Knapp, M., Romeo, R. & Beecham, J. (2009). Economic cost of autism in the UK. *Autism, 13,* 317-326.

Kochanska, G., Murray, K.T., & Harlan, E. (2000). Effortful control in early childhood: Continuity and change, antecedents, and implications for social development. *Developmental Psychology, 36,* 220-232.

Kolevzon, A., Newcorn, J. H., Kryzak, L., Chaplin, W., Watner, D., Hollander, E. *et al.* (2010). Relationship between whole blood seretonin and repetitive behaviors in autism. Psychiatric Research 175, 274-276.

King, B. (1993). Self-injury by people with mental retardation: A compulsive behavior hypothesis. *American Journal on Mental Retardation, 98,* 93–112.

Kushlick, A., Blunden, R., & Cox, G. (1973). Method of Rating Behavior Characteristics for Use in Large-Scale Surveys of Mental Handicap. *Psychological Medicine, 3,* 466-478.

LaChapelle, D. L., Hadjistavropoulos, T., & Craig, K. D. (1999). Pain Measurement in Persons with Intellectual Disabilities. *Clinical Journal of Pain, 15,* 13-23.

Lang, R., Sigafoos, J., Lancioni, G., Didden, R., & Rispoli, M. (2010). Influence of Assessment Setting on the Results of Functional Analyses of Problem Behavior. *Journal of Applied Behavior Analysis, 43,* 565-567.

Langdon, N. A., Carr, E. G., & Owen-DeSchryver, J. S. (2008). Functional Analysis of Precursors for Serious Problem Behavior and Related Intervention. *Behavior Modification, 32,* 804-827.

Langthorne, P. & McGill, P. (2008). Functional analysis of the early development of self-injurious behavior: Incorporating gene-environment interactions. *American Journal on Mental Retardation, 113,* 403-417.

Langthorne, P., McGill, P., & O'Reilly, M. (2007). Incorporating "motivation" into the functional analysis of challenging behavior - On the interactive and integrative potential of the motivating operation. *Behavior Modification, 31,* 466-487.

Lecavalier, L. (2006). Behavioral and emotional problems in young people with Pervasive Developmental Disorders: Relative prevalence, effects of subject charachteristics and empirical classification. *Journal of Autism and Developmental Disorders, 36,* 1101-1114.

Lewis, P., Abbeduto, L., Murphy, M., Richmond, E., Giles, N., Bruno, L. *et al.* (2006). Cognitive, language and social-cognitive skills of individuals with fragile X syndrome with and without autism. *Journal of Intellectual Disability Research, 50,* 532-545.

Lopez, B.R., Lincoln, A.J., Ozonoff, S. & Lai, Z. (2005). Examining the relationship between executive functions and restricted, repetitive symptoms of autistic disorder. *Journal of Autism & Developmental Disorders, 35,* 445-460.

Lord, C., Risi, S., Lambrecht, L., Cook, E. H., Leventhal, B. L., DiLavore, P. C. *et al.* (2000). The Autism Diagnostic Observation Schedule-Generic: A standard measure of social and communication deficits associated with the spectrum of autism. *Journal of Autism and Developmental Disorders, 30,* 205-223.

Lord, C., Rutter, M., & Lecouteur, A. (1994). Autism Diagnostic Interview-Revised - A Revised Version of A Diagnostic Interview for Caregivers of Individuals with Possible Pervasive Developmental Disorders. *Journal of Autism and Developmental Disorders, 24,* 659-685.

Lovaas, O. I. (1987). Behavioral Treatment and Normal Educational and Intellectual-Functioning in Young Autistic-Children. *Journal of Consulting and Clinical Psychology, 55,* 3-9.

Love, J. R., Carr, J. E., & LeBlanc, L. A. (2008). Functional assessment of problem behavior in children with autism spectrum disorders: A summary of 32 outpatient cases. *Journal of Autism and Developmental Disorders*.

Lowe, K., Allen, D., Jones, E., Brophy, S., Moore, K., & James, W. (2007). Challenging behaviours: prevalence and topographies. *Journal of Intellectual Disability Research, 51,* 625-636.

Luzzani, S., Macchini, F., Valade, A., Milani, D., & Selicorni, A. (2003). Gastroesophageal reflux and Cornelia de Lange syndrome: Typical and atypical symptoms. *American Journal of Medical Genetics Part A, 119A,* 283-287.

Mailis, A. (1996). Compulsive targeted self-injurious behaviour in humans with neuropathic pain: a counterpart of animal autonomy? Four case reports and literature review. *Pain, 64,* 569-578.

Mandell, D. S. (2008). Psychiatric hospitilization among children with autism spectrum disorder. *Journal of Autism and Developmental Disorders, 38,* 1059-1065.

Mandell, D. S. & Salzer, M. S. (2007). Who joins support groups among parents of children with autism. *Autism, 11,* 111-122.

Manly, T., Robertson, I. H., Anderson, V., & Nimmo-Smith, I. (1999). *The Test of Everyday Attention for Children (TEA-Ch).* Bury St Edmunds, UK: Thames Valley Test Company.

Marston, G. M., Perry, D. W., & Roy, A. (1997). Manifestations of depression in people with intellectual disability. *Journal of Intellectual Disability Research, 41,* 476-480.

Martin N., Oliver C. & Hall S. (2000) *Obswin*, Version 3.0. University of Birmingham, Birmingham.

Marzullo Kerth, D., Progar, P. R., & Morales, S. (2009). The effects of non-contingent self-restraint on self-injury. *Journal of Applied Research in Intellectual Disabilities, 22,* 187-193.

Matson, J. L. & LoVullo, S. V. (2008). A review of behavioral treatments for self-injurious behaviors of persons with autism spectrum disorders. *Behavior Modification 32,* 61-76.

Matson, J. L. & LoVullo, S. V. (2009). Trends and topics in autism spectrum disorders research. *Research in Autism Spectrum Disorders, 3,* 252-257.

Matson, J. L. & Shoemaker, M. (2009). Itellectual disability and its relationship to autism spectrum disorders. *Research in Developmental Disabilities, 30,* 1107-1114.

Matson, J. L., Baglio, C. S., Smiroldo, B. B., Hamilton, M., Packlowskyj, T., Williams, D. *et al.* (1996). Characteristics of autism as assessed by the diagnostic assessment for the severely handicapped-II (DASH-II). *Research in Developmental Disabilities, 17,* 135-143.

Matson, J. L., Mahan, S., Hess, J. A., Fodstad, J. C., & Neal, D. (2010). Progression of challenging behaviors in children and adolescents with Autism Spectrum Disorders as measured by the Autism Spectrum Disorders-Problem Behaviors for Children (ASD-PBC). *Research in Autism Spectrum Disorders, 4,* 400-404.

Matson, J. L. & Rivet, T. T. (2008). The effects of severity of autism and PDD-NOS symptoms on challenging behaviors in adults with intellectual disabilities. *Journal of Developmental and Physical Disabilities, 20,* 41-51.

McClintock, K., Hall, S., & Oliver, C. (2003). Risk markers associated with challenging behaviours in people with intellectual disabilities: a meta-analytic study. [Article]. *Journal of Intellectual Disability Research, 47,* 405-416.

McComas, J., Hoch, H., Paone, D., & El-Roy, D. (2000). Escape behavior during academic tasks: A preliminary analysis of idiosyncratic establishing operations. *Journal of Applied Behavior Analysis, 33,* 479-493.

McDermott, S., Zhou, L., & Mann, J. (2008). Injury treatment among children with autism or Pervasive Developmental Disorder. *Journal of Autism and Developmental Disorders, 38,* 626-633.

Mcelroy, S. L., Phillips, K. A., & Keck, P. E. (1994). Obsessive-Compulsive Spectrum Disorder. *Journal of Clinical Psychiatry, 55,* 33-53.

McKerchar, T. L., Kahng, S., Casioppo, E., & Wilson, D. (2001). Functional analysis of self-injury maintained by automatic reinforcement: Exposing masked social functions. *Behavioral Interventions, 16,* 59-63.

McTiernan, A., Leader, G., Healy, O., & Mannion, A. (2011). Analysis of risk factors and early predictors of challenging behavior for children with autism spectrum disorder. *Research in Autism Spectrum Disorders, 5,* 1215-1222.

Melzack, R. & Wall, P. D. (1965). Pain Mechanisms - A New Theory. *Science, 150,* 971-&.

Messmer, R. L., Nader, R., & Craig, K. D. (2008). Brief report: Judging pain intensity in children with autism undergoing venepuncture: The influence of facial activity. *Journal of Autism and Developmental Disorders, 38,* 1391-1394.

Meyer, L. H. & Evans, I. M. (1993). Science and Practice in Behavioral Intervention - Meaningful Outcomes, Research Validity, and Usable Knowledge. *Journal of the Association for Persons with Severe Handicaps, 18,* 224-234.

Michael, J. (1982). Distinguishing Between Discriminative and Motivational Functions of Stimuli. *Journal of the Experimental Analysis of Behavior, 37,* 149-155.

Miller, L.J., Reisman, J.E., McIntosh, D.N., & Simon, J. (2001). An ecological model of sensory modulation: Performance of children with fragile X syndrome, autistic disorder, attention-deficit/hyperactivity disorder, and sensory modulation dysfunction. In S. Smith-Roley, E.I. Blanche, & R.C. Schaaf (Eds.), *Understanding the nature of sensory integration with diverse populations* (pp. 57–88). San Antonio, TX: Therapy Skill Builders.

Moss, J. & Howlin, P. (2009). Autism spectrum disorders in genetic syndromes: implications for diagnosis, intervention and understanding the wider autism spectrum disorder population. *Journal of Intellectual Disability Research, 53,* 852-873.

Moss, J., Magiati, I., Charman, T., & Howlin, P. (2008a). Stability of the autism diagnostic interview-revised from pre-school to elementary school age in children with autism spectrum disorders. *Journal of Autism and Developmental Disorders, 38,* 1081-1091.

Moss, J., Oliver, C., Arron, K., Burbidge, C., & Berg, K. (2009). The Prevalence and Phenomenology of Repetitive Behavior in Genetic Syndromes. *Journal of Autism and Developmental Disorders, 39,* 572-588.

Moss, J. F., Oliver, C., Berg, K., Kaur, G., Jephcott, L., & Cornish, K. (2008b). Prevalence of Autism spectrum phenomenology in Cornelia de Lange and Cri du Chat syndromes. *American Journal on Mental Retardation, 113,* 278-291.

Mullen, E. M. (1995). *Mullen Scales of Early Learning* (AGS ed.). Circle Pines, MN: American Guidance Service Inc.

Murphy, G., Beadle-Brown, J., Wing, L., Gould, J., Shah, A., & Holmes, M. (2005). Chronicity of challenging behaviours in people with severe intellectual disabilities and/or Autism: A total population sample. *Journal of Autism and Developmental Disorders, 35,* 405-418.

Murphy, G., Hall, S., Oliver, C., & Kissi-Debra, R. (1999). Identification of early self-injurious behaviour in young children with intellectual disability. *Journal of Intellectual Disability Research, 43,* 149-163.

Murphy, G., Macdonald, S., Hall, S., & Oliver, C. (2000). Aggression and the termination of "rituals": a new variant of the escape function for challenging behavior? *Research in Developmental Disabilities, 21,* 43-59.

Murphy, G. & Wilson, B. (1985) *Self-injurious Behaviour*. Kidderminster: British Institute of Mental Handicap Publications.

Murphy, O., Healy, O., & Leader, G. (2009). Risk factors for challenging behaviors among 157 children with autism spectrum disorder in Ireland. *Research in Autism Spectrum Disorders, 3,* 474-482.

Nader, R., Oberlander, T. F., Chambers, C. T., & Craig, K. D. (2004). Expression of Pain in Children with Autism. *Clinical Journal of Pain, 20,* 88-97.

Nassar, N., Dixon, G., Bourke, J., Bower, C., Glasson, E., de Klerk, N. *et al.* (2009). Autism spectrum disorders in young children: effect of changes in diagnostic practices. *International Journal of Epidemiology, 38,* 1245-1254.

O'Reilly, M. F. (1997). Functional analysis of episodic self-injury correlated with recurrent otitis media. *Journal of Applied Behavior Analysis, 30,* 165-168.

O'Reilly, M. F., Lacey, C., & Lancioni, G. E. (2000). Assessment of the influence of background noise on escape-maintained problem behavior and pain behavior in a child with Willlams syndrome. *Journal of Applied Behavior Analysis, 33,* 511-514.

O'Reilly, M. F., Rispoli, M., Davis, T., Machalicek, W., Lang, R., Sigafoos, J. *et al.* (2010). Functional analysis of challenging behavior in children with autism spectrum disorders: A summary of 10 cases. *Research in Autism Spectrum Disorders 4*, 1-10.

O'Reilly, M. F., Sigafoos, J., Lancioni, G. E., Edrisinha, C., & Andrews, A. (2005). An examination of the effects of a classroom activity schedule on levels of self-injury engagement for a child with severe autism. *Journal of Autism and Developmental Disorders, 35,* 305-311.

Oliver, C. (1993). Self-injurious behaviour: From response to strategy. In C. Kiernan (Ed.) *Challenging behaviour and learning disabilities: Research to practice?: Implications of research on the challenging behaviour of people with learning disabilities* (p 135-188). Clevedon, Bristol: BILD Publications.

Oliver, C. (1995). Annotation: Self-injurious behaviour in children with learning disabilities: Recent advances in assessment and intervention. *Journal of Child Psychology and Psychiatry, 36,* 909-927.

Oliver, C., Hall, S., & Murphy, G. (2005). The early development of self-injurious behaviour: evaluating the role of social reinforcement. *Journal of Intellectual Disability Research, 49,* 591-599.

Oliver, C., Murphy, G., & Corbett, J. A. (1987). Self-injurious behaviour in people with mental handicap: A total population study. *Journal of Mental Deficiency Research, 31,* 147-162.

Oliver, C., Murphy, G., Hall, S., Arron, K., & Leggett, J. (2003). Phenomenology of self-restraint. *American Journal on Mental Retardation, 108,* 71-81.

Oliver, C., Arron, K., Sloneem, J., & Hall, S. (2008). Behavioural phenotype of Cornelia de Lange syndrome: case-control study. *British Journal of Psychiatry, 193,* 466-470.

Oliver, C., Murphy, G., Crayton, L., & Corbett, J. (1993). Self-Injurious-Behavior in Rett Syndrome - Interactions Between Features of Rett Syndrome and Operant-Conditioning. *Journal of Autism and Developmental Disorders, 23,* 91-109.

Oliver, C., Petty, J., Ruddick, L. and Bacarese-Hamilton, M. (In press). The association between repetitive, self-injurious and aggressive behavior in children with severe intellectual disability. *Journal of Autism and Developmental Disorders.*

Oliver, C., Sloneem, J., Hall, S. & Arron, K. (2009). Self-injurious behaviour in Cornelia de Lange syndrome: I. Prevalence and phenomenology. *Journal of Intellectual Disability Research, 53*, 575-589.

Olsson, M. B. & Hwang, C. P. (2001). Depression in mothers and fathers of children with intellectual disability. *Journal of Intellectual Disability Research, 45,* 535-543.

Ornitz, E. M., Guthrie, D., & Farley, A. H. (1977). Early Development of Autistic-Children. *Journal of Autism and Childhood Schizophrenia, 7,* 207-229.

Paclawskyj, T. R., Matson, J. L., Rush, K. S., Smalls, Y., & Vollmer, T. R. (2000). Questions about behavioral function (QABF): a behavioral checklist for functional assessment of aberrant behavior. *Research in Developmental Disabilities, 21,* 223-229.

Palmer, J. & Jenkins, J. (1982). The Wessex Behavior Rating System for Mentally-Handicapped People - Reliability Study. *British Journal of Mental Subnormality, 28,* 88-96.

Papageorgiou, V., Kalyva, E., Dafoulis, V., & Vostanis, P. (2008). Differences in Parents' and Teachers' Ratings of ADHD Symptoms and Other Mental Health Problems. *European Journal of Psychiatry, 22,* 200-210.

Pence, S. T., Roscoe, E. M., Bourret, J. C., & Ahearn, W. H. (2009). Relative contributions of three descriptive methods: implications for behavioral assessment. *Journal of Applied Behavior Analysis, 42,* 425-446.

Petty, J., Allen, D., & Oliver, C. (2009). Relationship Among Challenging, Repetitive, and Communicative Behaviors in Children With Severe Intellectual Disabilities. *American Journal on Intellectual and Developmental Disabilities, 114,* 356-368.

Petty, J., Bacarese-Hamilton, M. & Oliver, C. (In Prep.) Investigating risk-markers for self-injurious and other challenging behaviours in children under the age of five years.

Poustka, F. & Lisch, S. (1993). Autistic behaviour domains and their relation to self-injurious behaviour. *Acta Paedopsychiatr, 56,* 69-73.

Powell, S. B., Bodfish, J. W., Parker, D., Crawford, T. W., & Lewis, M. H. (1996). Self-restraint and self-injury: Occurrence and motivational significance. *American Journal on Mental Retardation, 101,* 41-48.

Reese, R. M., Richman, D. M., Belmont, J. M., & Morse, P. (2005). Functional characteristics of disruptive behavior in developmentally disabled children with and without autism. *Journal of Autism and Developmental Disorders, 35,* 419-428.

Reese, R. M., Richman, D. M., Zarcone, J., & Zarcone, T. (2003). Individualizing functional assessments for children with autism: the contribution of perseverative behavior and sensory disturbances to disruptive behavior. *Focus on Autism and Other Developmental Disabilities, 18,* 89-94.

Remington, B., Hastings, R. P., Kovshoff, H., Espinosa, F. D., Jahr, E., Brown, T. *et al.* (2007). Early intensive behavioral intervention: Outcomes for children with autism and their parents after two years. *American Journal on Mental Retardation, 112,* 418-438.

Richards, C., Moss, J., O'Farrell, L., Kaur, G., & Oliver, C. (2009). Social Anxiety in Cornelia de Lange Syndrome. *Journal of Autism and Developmental Disorders, 39,* 1155-1162.

Richler, J., Bishop, S. L., Kleinke, J. R., & Lord, C. (2007). Restricted and repetitive behaviors in young children with autism spectrum disorders. *Journal of Autism and Developmental Disorders, 37,* 73-85.

Richman, D. M. (2008). Early intervention and prevention of self-injurious behaviour exhibited by young children with developmental disabilities. *Journal of Intellectual Disability Research, 52,* 3-17.

Richman, D. M. & Lindauer, S. E. (2005). Longitudinal assessment of stereotypic, proto-injurious, and self-injurious behavior exhibited by young children with developmental delays. *American Journal on Mental Retardation, 110,* 439-450.

Robertson, J., Emerson, E., Pinkney, L., Caesar, E., Felce, D., Meek, A. *et al.* (2005). Treatment and management of challenging behaviours in congregate and noncongregate community-based supported accommodation. *Journal of Intellectual Disability Research, 49,* 63-72.

Rogers, S. J., Hepburn, S., & Wehner, E. (2003). Parent reports of sensory symptoms in toddlers with autism and those with other developmental disorders. *Journal of Autism and Developmental Disorders, 33,* 631-642.

Rojahn, J., Mulick, J. A., McCoy, D., & Schroeder, S. R. (1978). Setting events, adaptive clothing, and the modification of head banging and self-restraint in two profoundly retarded adults. *Behavioral Analysis and Modification 2,* 185-196.

Ross, E. & Oliver, C. (2002). The relationship between levels of mood, interest and pleasure and 'challenging behaviour' in adults with severe and profound intellectual disability. *Journal of Intellectual Disability Research, 46,* 191-197.

Ross, E. & Oliver, C. (2003). Preliminary analysis of the psychometric properties of the Mood, Interest and Pleasure Questionnaire (MIPQ) for adults with severe and profound learning disabilities. *British Journal of Clinical Psychology, 42,* 81-93.

Rutter, M., Bailey, A. & Lord, C. (2003) *Social Communication Questionnaire (SCQ)*. Los Angeles, LA: Western Psychological Services

Salkovskis, P. M. (1999). Understanding and treating obsessive-compulsive disorder. *Behaviour Research and Therapy, 37,* S29-S52.

Sandman, C. A., Hetrick, W., Taylor, D. V., Marlon, S. D., Touchette, P. E., Barron, J. L. *et al.* (2000). Long term effects of nalrexone on self-injurious behavior. *American Journal on Mental Retardation, 105,* 103-117.

Sandman, C. A. (1992). Various Endogenous Opioids and Autistic Behavior - A Response. *Journal of Autism and Developmental Disorders, 22,* 132-133.

Sandman, C. A., Barron, J. L., Chiczdemet, A., & Demet, E. M. (1990). Plasma B-Endorphin Levels in Patients with Self-Injurious-Behavior and Stereotypy. *American Journal on Mental Retardation, 95,* 84-92.

Sandman, C. A., Datta, P. C., Barron, J., Hoehler, F. K., Williams, C., & Swanson, J. M. (1983). Naloxone Attenuates Self-Abusive Behavior in Developmentally Disabled Clients. *Applied Research in Mental Retardation, 4,* 5-11.

Sandman, C. A. & Hetrick, W. P. (1995). Opiate mechanisms in self-injury. *Mental Retardation and Developmental Disabilities Research Reviews, 1,* 130-136.

Saracino, J., Noseworthy, J., Steiman, M., Reisinger, L., & Fombonne, E. (2010). Diagnostic and Assessment Issues in Autism Surveillance and Prevalence. *Journal of Developmental and Physical Disabilities, 22,* 317-330.

Sayers, N., Oliver, C., Ruddick, L., & Wallis, B. (2011). Stereotyped behaviour in children with autism and intellectual disability: an examination of the executive dysfunction hypothesis. *Journal of Intellectual Disability Research, 55,* 699-709.

Schneider, M. J., Bijam-Schulte, A. M., Janssen, C. G. C., & Stolk, J. (1996). The origin of self-injurious behaviour of children with mental retardation. *British Journal of Developmental Disabilities 42,* 136-148.

Schroeder, S. R., Oster-Granite, M. L., Berkson, G., Bodfish, J. W., Breese, G. R., Cataldo M. F., *et al.* (2001) Self-injurious behavior: gene, brain, behavior relationships. *Mental Retardation and Developmental Disabilities Research Reviews, 7,* 3–12.

Schroeder, S. R., Loupe, P. S., & Tessel, R. E. (2008). Animal Models of Self-Injurious Behavior: Induction, Prevention, and Recovery. *International Review of Research in Mental Retardation,, 36,* 195-231.

Schroeder, S. R., Schroeder, C. S., Smith, B., & Dalldorf, J. (1978). Prevalence of self-injurious behaviors in a large state facility for the retarded: A three year follow up study. *Journal of Autism and Childhood Schizophrenia, 8,* 261-269.

Schroeder, S, R. & Tessel, R(1994) *Dopaminergic and serotonergic mechanisms in self-injury and aggression. Destructive behavior in developmental disabilities: Diagnosis and treatment.* In Thompson, Travis (Ed); Gray, David B. (Ed), (1994). Destructive behavior in developmental disabilities: Diagnosis and treatment, Sage focus editions, Vol. 170 (pp. 198-210). Thousand Oaks, CA, US: Sage Publications.

Seltzer, M., Greenberg, J., Hong, J., Smith, L., Almeida, D., Coe, C. *et al.* (2010). Maternal Cortisol Levels and Behavior Problems in Adolescents and Adults with ASD. *Journal of Autism and Developmental Disorders, 40,* 457-469.

Seth, K. & Oliver, C. (In Prep.) Review of research trends in autism spectrum disorder.

Shattuck, P. T., Seltzer, M. M., Greenberg, J. S., Orsmond, G. I., Bolt, D., Kring, S. *et al.* (2007). Change in autism symptoms and maladaptive behaviors in adolescents and adults with an autism spectrum disorder. *Journal of Autism and Developmental Disorders, 37,* 1735-1747.

Simonoff, E., Pickles, A., Charman, T., Chandler, S., Loucas, T., & Baird, G. (2008). Psychiatric disorders in children with autism spectrum disorders: prevalence, comorbidity and associated factors. *Journal of the American Academy of Child and Adolescent Psychiatry, 8,* 921-929.

Smith, R. G. & Churchill, R. M. (2002). Identification of environmental determinants of behavior disorders through functional analysis of precursor behaviors. *Journal of Applied Behavior Analysis, 35,* 125-136.

Smith, R. G., Iwata, B. A., Vollmer, T. R., & Pace, G. M. (1992). On the Relationship Between Self-Injurious-Behavior and Self-Restraint. *Journal of Applied Behavior Analysis, 25,* 433-445.

Sparrow, S.S., Balla, D.A., & Cicchetti, D.V. (1985). *Vineland Adaptive Behavior Scales-Classroom Edition.* Circle Pines, MN: American Guidance Service.

Stallard, P., Williams, L., Lenton, S., & Velleman, R. (2007). Pain in cognitively impaired, non-communicating children. *Archives of Disease in Childhood, 85,* 460-462.

Symons, F. J., Clark, R. D., Hatton, D. D., Skinner, M., & Bailey, D. B. (2003). Self-injurious behavior in young boys with Fragile X syndrome. *American Journal of Medical Genetics, 118A,* 115-121.

Symons, F. J. & Danov, S. E. (2005). A prospective clinical analysis of pain behavior and self-injurious behavior. *Pain, 117,* 473-477.

Symons, F. J., Sperry, L. A., Dropik, P. L., & Bodfish, J. W. (2005). The early development of stereotypy and self-injury: a review of research methods. *Journal of Intellectual Disability Research, 49,* 144-158.

Symons, F. J. & Thompson, T. (1997). Self-injurious behaviour and body site preference. *Journal of Intellectual Disability Research, 41,* 456-468.

Tarbox, J., Wilke, A., Najdowski, A., Findel-Pyles, R., Balasanyan, S., Caveney, A. *et al.* (2009). Comparing Indirect, Descriptive, and Experimental Functional Assessments of Challenging Behavior in Children with Autism. *Journal of Developmental and Physical Disabilities, 21,* 493-514.

Taylor, L., Murphy, G. H., & Oliver, C. (2006). Self-injurious behaviour: A twenty year follow-up of the first total population study. *Journal of Applied Research in Intellectual Disabilities, 19,* 233.

Taylor, L., Oliver, C., & Murphy, G. (2011). The Chronicity of Self-Injurious Behaviour: A Long-Term Follow-Up of a Total Population Study. *Journal of Applied Research in Intellectual Disabilities, 24,* 105-117.

Thompson, T., Hackenberg, T., Cerutti, D., Baker, D., & Axtell, S. (1994). Opioid Antagonist Effects on Self-Injury in Adults with Mental-Retardation - Response Form and Location As Determinants of Medication Effects. *American Journal on Mental Retardation, 99,* 85-102.

Tiger, J. H., Fisher, W. W., Toussaint, K. A., & Kodak, T. (2009). Progressing from initially ambiguous functional analyses: three case examples. *Research in Developmental Disabilities, 30,* 910-926.

Totsika, V. & Hastings, R. P. (2009). Persistent challenging behaviour in people with an intellectual disability. *Current Opinion in Psychiatry, 22,* 437-441.

Totsika, V., Hastings, R. P., Emerson, E., Lancaster, G. A., & Berridge, D. M. (2011). A population-based investigation of behavioural and emotional problems and maternal mental health: associations with autism spectrum disorder and intellectual disability. *Journal of Child Psychology and Psychiatry, 52,* 91-99.

Tsakanikos, E., Costello, H., Holt, G., Sturmey, P., & Bouras, N. (2007). Behaviour management problems as predictors of psychotropic medication and use of psychiatric services in adults with autism. *Journal of Autism and Developmental Disorders, 37,* 1080-1085.

Turner, M. (1999). Annotation: Repetitive behaviour in autism: A review of psychological research. *Journal of Child Psychology and Psychiatry, 40,* 839-849.

Underwood, L., McCarthy, J., & Tsakanikos, E. (2010). Mental health of adults with autism spectrum disorders & ID. *Journal of Applied Research in Intellectual Disabilities, 23,* 421.

van Schrojenstein Lantman-De Valk, Linehan, C., Kerr, M., & Noonan-Walsh, P. (2007). Developing health indicators for people with intellectual disabilities. The method of the Pomona project. *Journal of Intellectual Disability Research, 51,* 427-434.

van Schrojenstein Lantman-De Valk, Metsemakers, J. F., Haveman, M. J., & Crebolder, H. F. (2000). Health problems in people with intellectual disability in general practice: a comparative study. *Fam Pract, 17,* 405-407.

White, P., O'Reilly, M., Fragale, C., Kang, S., Muhich, K., Falcomata, T. *et al.* (2011). An extended functional analysis protocol assesses the role of stereotypy in aggression in two young children with autism spectrum disorder. *Research in Autism Spectrum Disorders, 5,* 784-789.

Wing, L. & Gould, J. (1979). Severe Impairments of Social-Interaction and Associated Abnormalities in Children - Epidemiology and Classification. *Journal of Autism and Developmental Disorders, 9,* 11-29.