CHALLENGING BEHAVIOUR IN CHILDREN WITH SEVERE INTELLECTUAL DISABILITIES: IDENTIFICATION AND CHARACTERISTICS OF THOSE AT HIGH RISK

By

LOUISE ELLEN DAVIES

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School of Psychology
University of Birmingham
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Abstract

Background: The prevalence, persistence and often early onset of challenging behaviour in individuals with severe intellectual disabilities allude to the potential of early intervention. Identification of children at high risk of challenging behaviour would enable effective implementation of this strategy.

Method: Questionnaire studies examined the association between child characteristics and the presence of challenging behaviour at one point in time and 18 months later using the Challenging Behaviour Screening Questionnaire (CBSQ). Natural observations, questionnaires and objective measures were utilised to assess the validity of the CBSQ. The functional and communicative nature of the challenging behaviour demonstrated by participants at high risk was examined using experimental functional and descriptive analyses.

Results: The relative risk of challenging behaviour at one point in time and its persistence 18 months later was significantly increased by repetitive, restricted, overactive and impulsive behaviour. The concurrent and convergent validity of the CBSQ was demonstrated. Much of the challenging behaviour demonstrated by high risk participants appeared functional and closely associated with communicative behaviours.

Discussion: Theoretical underpinnings of challenging behaviour in this population are examined with emphasis on the interaction between child characteristics and environmental variables and the potential success of early intervention programmes for these children proposed.

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Chapter 1

Introduction

1.1 Challenging behaviour in people with intellectual disabilities

People who have intellectual disabilities face adversity across a range of social and personal dimensions. A significantly greater number of households supporting an individual with an intellectual disability live below the poverty line compared to the average household, a finding largely replicated across countries and individuals with varying degrees of intellectual disability (Drews, Yeargin-Allsopp, Decoufle & Murphy, 1995; Emerson, 2003; Fujiura, 1998; Leonard & Wen, 2002). The employment rate of people with intellectual disabilities is also considerably lower than that for the general population (Taanila, Rantakallio, Koiranen, vonWendt & Jarvelin, 2005). With regard to the physical and mental health of people with intellectual disabilities, higher rates of physical illness have been reported in this population, related either to their disability or inadequate health promotion (Kerr, Fraser & Felce, 1996). The prevalence of mental health problems (Borthwick-Duffy, 1994; Campbell & Malone, 1991) and challenging behaviours also appear to be higher in people with intellectual disabilities than the typically developing population. Indeed, despite decades of research, challenging behaviour remains a significant issue for this population.

Challenging behaviour has been defined as

> "culturally abnormal behaviour(s) of such intensity, frequency or duration that the physical safety of the person or others is likely to be placed in serious

> jeopardy, or behaviour which is likely to seriously limit use of, or result in the person being denied access to, ordinary community facilities" (Emerson, 1995, p. 4-5).

Encompassing a wide range of behaviours, the term challenging behaviour includes all behaviours which are perceived as presenting a challenge to services (Blunden & Allen, 1987). This definition, broadly derived from a social perspective on difference, became widely used in the hope that it would focus attention on the need to develop services which could cater effectively for the needs of individuals with intellectual disabilities. However, this administrative definition has led to the clustering of behaviours with very different qualities and diverse consequences for the individuals, so that the term "challenging behaviour" is perhaps more useful for those organising services for this population than the individuals themselves. This all encompassing definition can lead to difficulties in interpreting data regarding the examination of general challenging behaviour, as the prevalence and cause of challenging behaviour might depend on the exact form. The term challenging behaviour is used in this thesis as an umbrella term for aggression, destruction and self-injury alone, although traditionally the definition of challenging behaviour is often more wide reaching.

The prevalence of challenging behaviour reported in the literature indicates a range of 10 to 15% of all people with intellectual disability (Emerson *et al.,* 2001a; Emerson & Bromley, 1995; Kiernan & Qureshi, 1993; Lowe *et al.,* 2007). Similar prevalence rates for self-injury have also been reported (e.g. Ando & Yoshimura, 1978; Ballinger, 1971; Borthwick-Duffy, 1984; Eyman & Call, 1977; Jacobson, 1982; Kebbon & Windahl, 1986; Oliver, Murphy & Corbett, 1987; Saloviita, 2000), whilst

rates appear to be slightly higher for aggression (range 2 to 20%; e.g. Cooper, 1998; Crocker *et al.,* 2006; Harris 1993; Sigafoos, Elkins, Kerr & Attwood, 1994) and destruction (range 4 to 24%; e.g. Ando & Yoshimura; Crocker *et al*.; Deb, Thomas & Bright, 2001). These wide ranging prevalence rates are likely to be caused by the varying definition of challenging behaviour used (i.e. different approaches to the measurement of challenging behaviour, one defining behaviour in terms of impact and another topographically) and the length of time the behaviour was measured, as well as the sample employed and other methodological variables.

Age related changes in the prevalence of challenging behaviour have been reported in some studies. Examining the prevalence of self-injury specifically, Oliver *et al.* (1987) and Rojahn (1986) reported an increase in prevalence until the mid 30's at which point the prevalence declines. Broadly supporting these results, Borthwick-Duffy (1994) reported the highest prevalence of self-injurious behaviour to be around the early 20's. The results of this same study also appeared to indicate a slight increase in the prevalence of aggression in individuals after the age of 20 years. However, not all studies investigating challenging behaviour have found age related changes (e.g. Fraser, Leudar, Gray & Campbell, 1986; Hill & Bruininks, 1984; Hillery & Mulcahy, 1996) indicating the need for a more systematic evaluation of changes in challenging behaviour with age.

The relatively high prevalence of challenging behaviour in people with intellectual disabilities in combination with probable increases in this behaviour with age until early adulthood indicates the significance of challenging behaviour for this population.

1.2 Models of challenging behaviour

Explanations of the causes of challenging behaviour are based on two broad approaches, biological and behavioural. Whilst not necessarily in opposition, little attempt has been made to integrate the two to account for robust findings supporting each perspective (Oliver, 1993).

1.2.1 Biological models of challenging behaviour

1.2.1.1 Neurotransmitter theories

Neurobiological theories of challenging behaviour have focused on the role of neurotransmitters, focusing on the serotonergic, opiatergic and dopaminergic systems. Whilst the serotonergic system is involved in the expression of both self-injury and aggression, neurotransmitter theories generally are more relevant to self-injury with far less emphasis on the demonstration of aggression.

1.2.1.1.1 The serotonergic system

With regard to both aggression and self-injury, research has indicated a negative association between levels of serotonin and these forms of challenging behaviour. Illustrating this relationship, Baumeister and Sevin (1990) demonstrated that increased serotonin synthesis led to a reduction in levels of aggression whilst inhibited serotonin synthesis caused an increase in this behaviour in non humans. Similarly, interventions which increase levels of serotonin, including reuptake inhibitors, have been related to reduced levels of self-injury (Aman, Arnold & Armstrong, 1999; Ellis, Singh & Ruane, 1999). A more recent review examining the effect of various selective serotonin reuptake inhibitors found mixed evidence with regard to their

effectiveness however and also noted a lack of empirically sound studies in this area (McDougle, Kresch & Posey, 2000).

1.2.1.1.2 The opiatergic system

Associated with self-injury, β-endorphin, an opioid peptide from the opiatergic system, has been reported to influence this behaviour via two pathways, analgesia and addiction. According to the work of Sandman, Spence and Smith (1999), excess opioid activity increases pain thresholds whilst the release of β-endorphin contingent upon self-injury automatically reinforces the behaviour by inducing a state of euphoria (although reduced levels of withdrawal are perhaps more likely). This model of β-endorphin function is supported by the findings of a review indicating the effective action of Naltrexone, an opiate antagonist, which was shown to reduce self-injury in around 80% of individuals, although less than half showed a clinically significant decrease (Symons, Thompson & Rodriguez, 2004). However, these results might also be accounted for by the increase in pain associated with the use of Naltrexone, with obvious ethical implications. Concerns regarding the methodology employed by the studies included in the review were also highlighted.

1.2.1.1.3 The dopaminergic system

Evidence for the involvement of dopamine in the expression of self-injury is derived primarily from the study of Lesch-Nyhan syndrome and animal models of this syndrome. Lesch-Nyhan syndrome is a genetically determined syndrome in which self-biting is almost universal after the age of two with animal models revealing decreased levels of dopamine and related metabolites as a result of a deficiency in specific dopamine pathways (Breese *et al.,* 1995). Investigation of dopamine agonists

based on animal models has also led to the prescription of atypical antipsychotics for self-injury with some success (Turner & Lewis, 2002; Breese *et al.,* 2005).

1.2.1.1.4 The basal ganglia hypothesis

Unifying neurotransmitter theories of challenging behaviour regarding the influence of serotonin and dopamine is the basal ganglia hypothesis. Dysregulation of the basal ganglia, a group of brain nuclei, has been identified in individuals with disorders associated with dopamine depletion and self-injury (Obeso *et al.,* 2000; Visser, Bar & Jinnah, 2000). Serotonergic pathways are also closely involved in the modulation of the basal ganglia (Wolf & Schutz, 1999) which are also implicated in self-injury, as previously discussed in section 1.2.1.1.1.

1.2.1.1.5 Monoamine oxidase A

A more recent biological theory of challenging behaviour proposed is based upon the functional polymorphism in the promoter of the gene encoding monoamine oxidase A (MAOA). MAOA, an enzyme involved in the biodegradation of serotonin and other monoaminergic neurotransmitters, has been associated with an aggressive phenotype in both animal models (e.g. Cases *et al.,* 1995) and studies involving typically developing humans (Brunner, Nelen, Breakefield, Ropers & van Oost, 1993; Caspi *et al.,* 2002). More recently, May *et al.* (2009) reported a significantly higher prevalence of the short allele MAOA polymorphism in individuals with intellectual disabilities and challenging behaviour than individuals with or without intellectual disabilities and no challenging behaviour. Some studies however, have failed to identify this association (Kim-Cohen *et al.,* 2006, Taylor & Kim-Cohen, 2007).

1.2.1.2 The compulsive behaviour hypothesis

The compulsive behaviour hypothesis is a neurobiological model of self-injury proposed by King (1993) whereby self-injury is posited to result from unspecified cerebral damage and to have a compulsive quality. This supposition is based on animal models demonstrating direct relationships between cerebral damage and self-injurious behaviour. This model is also supported by the results of empirical studies with clinical samples which have demonstrated close associations between self-injury and compulsive behaviour in individuals with Cornelia de Lange syndrome (Hyman, Oliver & Hall, 2002) and general intellectual disability (Bodfish *et al.,* 1995).

1.2.1.3 The neuropsychological hypothesis

According to Barkley's (1997a, 1997b) theory of Attention Deficit Hyperactivity Disorder (ADHD), compromised behavioural inhibition is specified as the central deficiency unique to this disorder. Subsequent impairments in other executive functions are also proposed to be associated with the impulsivity and overactivity demonstrated by individuals with the disorder. Compromised behavioural inhibition is also implicated in Turner's (1997, 1999) theory of repetitive behaviour and restricted interests in autism, whereby individuals with the disorder cannot inhibit an ongoing or inappropriate response and as a result demonstrate repetitive behaviour. It can therefore be argued from these models that impulsive, overactive and repetitive behaviours are associated with deficits in executive functioning and inhibition in particular. Thus, if children with severe intellectual disabilities were to demonstrate impulsive, overactive and repetitive behaviour (section 1.3.3) it might also be proposed that these individuals might well have compromised behavioural inhibition. This deficit could also aid our understanding of challenging behaviour in this

population, as a difficulty terminating ongoing responses could mean that children with compromised inhibition find it harder to inhibit this behaviour. This neuropsychological hypothesis could also complement operant models, so that children with compromised behavioural inhibition find it harder to inhibit challenging behaviour which has been reinforced previously (section 1.2.2.1).

Therefore, as discussed, there are currently several theories of challenging behaviour based on biological models. Neurotransmitter theories have proved particularly influential with regard to challenging behaviour interventions, although the effectiveness of medications according to large reviews (e.g. Baumeister, Todd & Sevin, 1993) is disappointing. In order to provide a robust biological account of challenging behaviour, it might be beneficial to perceive biological theories not as separate entities, but as associated or interacting causes, so that, for example, neurotransmitter dysfunction might underlie compromised behavioural inhibition leading to challenging behaviour.

1.2.2. Behavioural theories of challenging behaviour

The behavioural model of challenging behaviour, in all its various forms, proposes that challenging behaviour, like all other operant behaviour, is learned and maintained by its consequences.

1.2.2.1. Operant theory

Operant theory proposes that challenging behaviours are operant behaviours with the likelihood of future challenging behaviour increased by the contingent presentation of a reinforcer or reward (positive reinforcement) or the contingent removal of aversive

stimuli (negative reinforcement). Antecedents to challenging behaviour function as establishing operations or discriminative stimuli, acting as motivational states for the behaviour or indicating that reinforcement is available respectively (Michael, 1982). To illustrate, a common establishing operation for challenging behaviour is a low level of adult attention, or attention deprivation. Thus, adult attention contingent upon challenging behaviour acts as a positive reinforcer, a supposition supported by the empirical results of Hall, Oliver and Murphy (2001). The discriminative stimulus for this behaviour could be presence of an adult.

Operant conditioning plays a vital role in the development and maintenance of self-injury in Guess and Carr's (1991) stage model of the behaviour. In the first stage of the model, repetitive behaviours are proposed to be developmentally appropriate, state dependent internally regulated behaviours. During the second transitional phase of the model, these repetitive behaviours begin to influence, and be influenced by, the environment and thus become adaptive responses enabling homeostasis. The third and final stage of the model involves the evolution of these repetitive behaviours into self-injurious behaviour via operant processes. Oliver (1993) and Kennedy (2002) also proposed the development of self-injury from repetitive behaviour via several possible behavioural mechanisms. General support for this model is provided by Richman and Lindauer (2005) who found that some topographies of self-injury evolved from early stereotypic behaviour exhibited by children with intellectual disabilities.

Further enhancing Guess and Carr's (1991) model of self-injury, Oliver (Oliver & Head, 1990; Oliver, 1993; Oliver, 1995) developed a mutual reinforcement paradigm of self-injury based on the operant reinforcement of the behaviour consistent with

Guess and Carr's model, but also including biological variables, providing an integrated model of self-injury. Longitudinal empirical evidence for the role of social reinforcement in the development of self-injury has also been provided (Oliver, Hall & Murphy, 2005). The most recent evidence in support of both Guess & Carr's (1991) and Oliver's (Oliver & Head, 1990; Oliver, 1993; Oliver, 1995) model is provided by Petty, Allen and Oliver (2009) who identified a strong temporal association between repetitive and self-injurious behaviour, indicating the potential emergence of self-injury from repetitive behaviours.

1.2.2.2 Automatic reinforcement

Operant behaviours might also be reinforced by variables internal to the individual as opposed to within the environment, as indicated by automatic reinforcement (Kennedy, 1994; Vollmer, 1994). For example, health problems associated with pain, such as an irritating skin condition, might lead to challenging behaviour in the form of scratching as this relieves the irritation and removes the aversive stimulation and thus self-injury is negatively reinforced.

1.2.2.3 The communication hypothesis

Acknowledging the importance of social reinforcement, as in operant theory, but focusing more specifically on pragmatic communication, Carr and Durand (1985) proposed the communication hypothesis of challenging behaviour. The main premise of this hypothesis is that challenging behaviours function as nonverbal communicative acts and are thus similar to other nonverbal behaviours, such as pointing demonstrated by infants (Bates, Camaioni & Volteraa, 1975). Support for this hypothesis comes from the inverse relationship between communicative skill and behaviour problems

(e.g. Foxx & Livesay, 1984), indicating that for individuals with severe intellectual disabilities, challenging behaviours function like communicative behaviours, in order to gain attention or escape demands or obtain a desired object (proto-imperatives) for example (Bates, 1976). Research has also shown that typically developing two, but not three, year old children demonstrate communicatively functional aggression (Brownlee & Bakeman, 1981). The extinction of aggression in this older age group was, arguably, due to the functionally equivalent verbal communicative behaviours acquired, replacing challenging behaviour. Children with severe intellectual disabilities are less likely to acquire these functionally equivalent behaviours and thus retain challenging behaviour within their behavioural repertoire.

Based on the communication hypothesis, Carr and Durand (1985) established functional communication training, proposing that since challenging behaviour functions as a form of communication, teaching individuals appropriate functionally equivalent communicative behaviour would reduce the frequency of challenging behaviour. Having identified the function of challenging behaviour in four children with intellectual disabilities, Carr and Durand taught each participant a relevant and irrelevant response and found that only the functionally equivalent relevant response led to a reduction in challenging behaviour. Numerous studies have provided further evidence for the effectiveness of functional communication training in reducing the frequency of challenging behaviour, as well as demonstrating this reduction across new tasks, environments, teachers and over time (Durand & Carr, 1991; Durand & Carr, 1992).

Thus, there is a great deal of theoretical and empirical support for the importance of operant theories of challenging behaviour. Analogous with operant theory, the communication hypothesis also recognises the importance of social reinforcement, alongside pragmatic communication, providing the basis for functional communication training. Therefore, behavioural theories of challenging behaviour are fundamental to the understanding of the behaviour, as well as providing effective interventions.

1.2.3 Child characteristics associated with challenging behaviour

Numerous studies using a variety of methodologies and samples have identified various child characteristics associated with challenging behaviour in people with intellectual disabilities. Arguably the most well researched of these is severity of intellectual disability, a more severe degree of which is associated with challenging behaviour (Baghdadli, Pascal, Grisis & Aussilloux, 2003; Chadwick, Piroth, Walker, Bernard & Taylor, 2000; Holden & Gitlesen, 2006; McClintock, Hall & Oliver, 2003). A meta-analysis based on the findings of 22 studies conducted by McClintock *et al.* has also identified an association between challenging behaviour and deficits in communication, although such deficits are likely to be an artefact of severity of intellectual disability and thus associated with challenging behaviour due to the overlap between the characteristics. More recent studies however have indicated that despite being a robust risk marker, greater severity of intellectual disability might not be associated with challenging behaviour in children with a severe intellectual disability and thus those already at high risk (Petty *et al.*, in preparation; Oliver *et al.,* in preparation). This might also be the case for communicative ability.

Two child characteristics also receiving much support from the literature as putative risk markers of challenging behaviour are age (Chadwick *et al.* 2000; Baghdadali *et al.* 1993; Holden & Gitlesen, 2006) and the presence of autism (Baghdadli, *et al.*; Baghdadli *et al*., 2008; Holden & Gitlesen; McClintock *et al.,* 2003). Also emerging as a putative risk marker are health problems and associated pain. Additionally, although only recently identified as being associated with self-injury (Petty *et al.,* in preparation), health has long been implicated with challenging behaviour in typically developing children (de Lissovoy, 1962; Hart, Bax & Jenkins, 1984).

More recent studies examining the putative risk markers of challenging behaviour have focused on behavioural characteristics. Illustrating this, Oliver *et al.* (in preparation) identified high frequency repetitive and ritualistic behaviour as predictors of challenging behaviour, a finding also reported in two recent empirical studies (Oliver, Sloneem, Hall & Arron, 2009; Arron, Oliver, Berg, Moss & Burbidge, in review). Replicating these findings, Petty *et al.* (in preparation) also showed a positive association between repetitive, overactive and impulsive behaviours and challenging behaviour, indicating their roles as putative risk markers for challenging behaviour. The identification of overactive and impulsive behaviour as putative risk markers has also been supported by the recently identified associations between self-injurious and aggressive behaviour and attention deficit hyperactivity disorder, also in individuals with intellectual disabilities (Cooper, Smiley, Allan *et al*., 2009; Cooper, Smiley, Jackson *et al*., 2009).

Particular forms of challenging behaviour have also been reported to be more prevalent in individuals with specific genetic syndromes. For example, self-injury is

more commonly reported in Lowe, Smith-Magenis, Lesch-Nyhan and Prader-Willi syndromes (Clarke & Boer, 1998; Kenworthy & Charnas, 1995; Nyhan, 1994).

Research has thus indicated that the presence of particular child characteristics is associated with challenging behaviour, the identification of which might enable clinicians to target interventions at those individuals with intellectual disabilities most at risk of challenging behaviour. To understand how these characteristics function to increase the prevalence of challenging behaviour, consideration should be given to operant theory.

1.2.4 Operant and child characteristic interactions

According to operant learning theory, there should be no difference in the prevalence of challenging behaviour across the population of people with intellectual disabilities because environmental influences would, presumably, be randomly distributed across groups. However, as described in section 1.2.3, child characteristics associated with challenging behaviour have been identified, indicating that particular children with these characteristics are more likely to demonstrate challenging behaviour. Thus, it is likely that for some individuals with intellectual disability, challenging behaviour might arise as a result of operant reinforcement or because of the presence of a particular characteristic. However, it is also possible that challenging behaviour might develop or be maintained as the result of an interaction between the presence of a particular child characteristic and operant reinforcement. For example, repetitive behaviour has been identified as a risk marker for challenging behaviour, but to understand why that might be, consideration must be given to social reinforcement, as in Guess and Carr's (1991) model of the development of challenging behaviour

(section 1.2.2.1). Similarly in the case of health, automatic reinforcement might lead to challenging behaviour; however, this automatically reinforced behaviour might also be perceived adversely and thus positively reinforced by adults, so that the behaviour is maintained by operant reinforcement.

1.3. The cost of challenging behaviour

It has long been recognised that challenging behaviour is both costly and resource intensive. Mansell (1992) found that despite being in the minority, individuals with challenging behaviour use a disproportionate amount of resources. Illustrating the exact costs of service provision for individuals with intellectual disabilities and challenging behaviour, Hallam and Trieman (2001) identified an 86% increase in the mean weekly cost of services for individuals deemed difficult to place due to their challenging behaviour as compared to a comparison group of individuals without challenging behaviour. Four years later, this increased cost had decreased but only to 49%, indicating the higher costs associated with caring for individuals with challenging behaviour. The cause of this disparity might be the increased use of psychology and psychiatry required by individuals with challenging behaviour as well as their greater occupation of NHS facilities which are less cost effective (Knapp, Comas-Herrera, Astin, Beecham & Pendaries, 2005). Thus, challenging behaviour in people with intellectual disabilities has significant economic implications for the NHS.

1.4 Interventions for challenging behaviour

The most common interventions available to individuals with intellectual disabilities and challenging behaviour are pharmacological treatments and behavioural

interventions based on operant theory and the communication hypothesis. Recent reviews of pharmacological interventions have cited some evidence for the use of several medications, such as Risperidone (Deb *et al*., 2008; Deb & Unwin, 2007), although an earlier systematic review of the literature concluded that the evidence regarding the effectiveness of antipsychotic medications generally was mixed, with no evidence that this form of medication helped or harmed individuals with an intellectual disability and challenging behaviour (Brylewski & Duggan, 1999). The cost effectiveness of these medications has also been questioned (Romeo, Knapp, Tyrer, Crawford & Oliver-Africano, 2009). These reviews are limited however by the methodology employed (Matson *et al.,* 2000).

Conversely, significant reductions in the frequency of challenging behaviour following behavioural intervention have been identified in both meta-analytic studies of behavioural interventions (e.g. Harvey, Boer, Meyer & Evans, 2009) and the empirical literature (e.g. Kahng, Iwata & Lewin, 2002). However, the resource intensive nature of behavioural interventions (Robertson *et al*., 2005) limits the number of individuals with challenging behaviour who are able to access them. This problem is highlighted by Ruddick and Oliver (in preparation) who indicated that children presenting with high levels of challenging behaviour were at least thirteen times more likely to require behavioural intervention, yet only twice as likely to have contact with a specialist health care professional. Bearing in mind that challenging behaviour is likely to become more ingrained in the behavioural repertoire given an individuals increased experience of operant reinforcement with age (Guess & Carr, 1991; Oliver, 1995) this disparity in the number of children requiring and actually receiving behavioural interventions might well increase the prevalence of challenging

behaviour demonstrated in this population. Given the inadequacy of current service provision, attention is now turning to the potential of early intervention for this population (Richman, 2008; Richman & Lindauer, 2005; Symons, Sperry, Dropik & Bodfish, 2005).

1.5 Early intervention

An early intervention strategy has been successfully implemented in a wide range of disciplines. Widely used within health care, early intervention has significant reduced mortality rates from breast cancer (Blanks, Moss, McGahan, Quinn & Babb, 2000) with individuals suffering from asthma (Holt & Sly, 2000) and obesity (de Onis, 2004) also benefiting from the strategy. In contrast, early intervention has had a lesser impact on psychological services, although this approach has proved particularly effective in the treatment of psychosis when employed during the prodromal phase and shortly after onset (Johannessen *et al.*, 2001; Harris *et al.,* 2005). Researchers have also reported long term gains in intellectual and adaptive functioning in autism as a result of early intervention (e.g. McEachlin, Smith & Lovaas, 1993; Remington *et al.,* 2007), illustrating the potential utility of the approach, although a recent review indicated that there is considerable variability in outcome at an individual level (Howlin, Magiati & Charman, 2009).

Families of young children with additional support needs receiving support from portage services have reportedly valued the support provided, although only a minority of those eligible for support actually receive it (Russell, 2007). Early intervention has also been conducted in the field of intellectual disability with some

success (e.g. Alexander, 1998), although not specifically targeted at individuals at risk of challenging behaviour.

Government policy has strongly advocated the use of early intervention, stating the importance of prevention in both the green and white papers 'Every Child Matters' (Department for Education and Skills, 2003) and 'Our Health, Our Care, Our Say' (Department of Health, 2006). Standard 8 of the National Service Framework (Department of Health, 2004) also recommends the provision of intervention at the earliest opportunity to ensure its success for children with disabilities and complex needs. The cost effectiveness of the strategy reported with regard to mental health care costs (McCrone, Dhanasiri & Knapp, 2006) also makes early intervention a particularly appealing strategy.

Thus, there is a great deal of support for the use of early interventions in terms of government policy and economic benefits. Early interventions have also been successfully implemented in other domains, although there is some mixed evidence with regard to its effectiveness in more closely allied fields, such as autism.

1.6 The future of early intervention for challenging behaviour in children with severe intellectual disabilities

Recently, the focus of attention has begun to turn towards preventative as opposed to reactive forms of intervention for challenging behaviour in individuals with intellectual disabilities. Illustrating this, Richman (2008) provides an in-depth review of the current literature regarding theoretical models of the development of self-injurious behaviour, as well as proposing future directions involving the

implementation of early intervention strategies. Within the review, Richman acknowledges that well established self-injury is harder to treat based on operant theory (e.g. Guess & Carr, 1991) and thus proposes that individuals displaying this behaviour should receive intervention as soon as possible. However, Richman also highlights, as others have previously, (Symons *et al.,* 2005) the paucity of empirical studies examining the early development of self-injury, in contrast to the wealth of studies examining well established cases of this behaviour, and thus a lack of understanding regarding how best to implement early intervention.

Nevertheless, some evidence has been provided for the use of functional communication training (FCT) as a preventative intervention for self-injury (Reeve & Carr, 2000). Given the evidence in support of the effective use of FCT for well established forms of self-injury, FCT thus appears to be an appropriate and potentially effective intervention for use in early intervention programmes, so that after the implementation of preventative FCT, an establishing operation for challenging behaviour, such as attention deprivation, would instead be a discriminative stimulus for appropriate communication. Thus, children with intellectual disabilities would be taught to request access to social consequences in their environment reducing the demonstration of challenging behaviour functioning as a form of communication. In order to increase the effectiveness of early intervention, Richman (2008) also proposes that families should be taught to be more responsive to the needs of their children.

As yet however, there is no evidence indicating how to best target the early intervention proposed by Richman (2008). Thus, research must now investigate a

method for identifying young children most at risk for challenging behaviour to allow optimal allocation of services and the subsequent prevention of self-injury through the early application of effective interventions, such as FCT.

Results from the research literature to date indicate that due to the high prevalence, apparent increase with age and problems associated with current service provision, early intervention is a potentially promising progression for people with intellectual disabilities and challenging behaviour. However, whilst characteristics associated with challenging behaviour have been identified, a reliable and valid method for quickly identifying those at highest risk of challenging behaviour has yet to be developed, so that it is currently unclear how to best target early intervention services.

1.7 An overview of subsequent chapters of the thesis

This thesis investigates risk markers for the challenging behaviour demonstrated by children with severe intellectual disabilities and the characteristics of those children identified as being at high risk, in order to assess the suitability of these individuals to the type of intervention likely to be central to early intervention strategies.

More specifically, the aim of the literature review presented in Chapter 2 is to examine the prevalence of challenging behaviour with age through statistical analysis of prevalence data for aggression and self-injury broken down by age band obtained by systematic review of the literature. The aim of the study described in Chapter 3 is to examine the child characteristics acting as putative risk markers for challenging behaviour in children with a severe intellectual disability through the development of a brief screen. The persistence of the putative risk markers identified in this chapter as

well as their ability to significantly predict the future presence of challenging behaviour will be examined in a follow up study described in Chapter 4. The concurrent and convergent validity of the brief screen developed will also be examined in Chapter 5. The final empirical chapter will aim to investigate the challenging, proto-imperative and affect behaviours demonstrated by children who show characteristics that predict the presence of challenging behaviour, in order to determine a potential function of this behaviour and the communicative repertoire available to these individuals.

Chapter 2

A Systematic Review of the Age Related Prevalence of Aggression and Self-injury in People with Intellectual Disability

2.1 Abstract

Background and aims: It is often assumed that the prevalence of challenging behaviour increases with age, although few studies have analysed systematically age related differences in prevalence. The aim of this chapter is to review and analyse published data regarding the age related prevalence of aggression and self-injury in people with intellectual disability.

Method: Studies including prevalence data for aggression and/or self-injury broken down by age band data, as well as the number of participants per age band were identified and relative risk analyses conducted to generate an index of age related change.

Results: Despite some conflicting results, the analysis conducted on the findings of studies considered to be the most methodologically robust indicated that the relative risk of self-injury, and to a lesser extent aggression, increased with age until mid-adulthood. The analysis based on two studies also indicated that the relative risk of self-injury significantly decreases in older adulthood, illustrating a curvilinear relationship. This might also be true for aggression, although this relationship is less clear.

Discussion: These conclusions have implications for the understanding of the development of different forms of challenging behaviour and the design of early

intervention strategies. Of most importance, these results allude to the need for more methodologically robust studies of the prevalence of challenging behaviour with age.

2.2 Introduction

Challenging behaviour has a detrimental impact on the lives of a significant minority of individuals with intellectual disabilities. Research has indicated that those showing challenging behaviour are significantly more likely to be socially excluded, given fewer opportunities to form relationships and gain employment (Murphy, 2009). The stress associated with challenging behaviour for families of individuals with intellectual disabilities is also well documented (e.g. Hastings, 2002; Gallagher *et al.,* 2008), as is the emotional impact of this behaviour on staff, which might be associated with the high turnover observed within services (Felce, Lowe & Beswick, 1993). Associations between challenging behaviour and the increased cost of services illustrate the economic costs associated with challenging behaviour (Knapp, Comas-Herrera, Astin, Beecham & Pendaries, 2005).

Although wide variation exists in the prevalence rates reported for challenging behaviour, it is clear that challenging behaviour is demonstrated by a significant minority of people with intellectual disabilities. With regard to challenging behaviour generally, research indicates a range of 10 to 17% of all people with intellectual disability (Emerson *et al.,* 2001a; Emerson & Bromley, 1995; Kiernan & Qureshi, 1993; Lowe *et al.,* 2007). Similar prevalence rates for self-injury have also been reported (e.g. Ando & Yoshimura, 1978; Ballinger 1971; Borthwick-Duffy, 1994; Eyman & Call, 1977; Jacobson, 1982; Kebbon & Windahl, 1986; Oliver, Murphy & Corbett, 1987; Saloviita, 2000), whilst the upper range of prevalence rates for

aggression appear to be slightly higher at 20% (range 2 to 20%; e.g. Cooper, 1998; Cooper, Smiley, Jackson *et al*., 2009; Crocker *et al.,* 2006; Harris, 1993; Sigafoos, Elkins, Kerr & Attwood, 1994). Differences in the prevalence rates of general challenging behaviour as opposed to specific forms is likely to be due to different approaches to the measurement of challenging behaviour, one defining behaviour in terms of impact and another topographically.

There have been few systematic analyses of age differences in prevalence rates of challenging behaviour. Age has long been considered to be related to the presence of challenging behaviour, with several methodologically robust studies reporting an increase in the prevalence of challenging behaviour with age. More specifically, a small number of studies indicate an increase in the prevalence of challenging behaviour until the mid 30's at which point prevalence begins to decline (e.g. Kiernan & Kiernan, 1994; Oliver, *et al.,* 1987). Many researchers however, have failed to identify any association between age and challenging behaviour (e.g. Fraser, Leudar, Gray & Campbell, 1986; Hillery & Mulcahy, 1996) whilst others report age related changes in the prevalence of challenging behaviour but are imprecise with regard to the age with which the prevalence of challenging behaviour begins to change (Collacott, Cooper, Branford & McGrother, 1998; Hemmings, Gravestock, Pickard & Bouras, 2006; Kiernan & Alborz, 1996; Maisto, Baumeister & Maisto, 1978).

Such disparity between the findings of studies investigating the prevalence of challenging behaviour with age in individuals with an intellectual disability might result from the divergent methodologies and samples employed. For example, the prevalence of challenging behaviour might depend on the form of the behaviour.

Comparisons of the prevalence of broadly defined challenging behaviour within age bands across different forms of challenging behaviour might therefore be unproductive. The definition used as the criterion for the presence of behaviour is also likely to have a significant effect on the results, so that the age related prevalence of severe self-injury causing tissue damage might be quite different to milder self-injury. Additionally, the size and origin of the sample will influence results. Studies including large total population samples are likely to provide the most generalisable results, although in order to provide robust age related prevalence of challenging behaviour data, relatively narrowly defined age bands are also required to allow useful comparisons.

Establishing the prevalence of challenging behaviour across age bands might inform models of the development of the behaviour. Whilst both biological and operant processes have been implicated in the development of challenging behaviour (Oliver, 1993), the ontogeny of specific forms of challenging behaviour are not well understood. Guess and Carr's (1991) stage model indicates that self-injury emerges from repetitive behaviour, thus this behaviour is proposed to have a specific developmental trajectory. However, very little research has been devoted to the development of other forms of challenging behaviour. From a clinical perspective, being aware of age related changes in the prevalence of challenging behaviour would not only enable services to plan effectively for the future needs of children with intellectual disabilities, but could potentially help services to target early intervention at different age bands before the prevalence of challenging behaviour begins to increase.

There are therefore, advantages to further investigating the prevalence of challenging behaviour with age. The aim of this chapter is to review and analyse data published in the research literature regarding the age related prevalence of aggression and self-injury in people with intellectual disability. These specific forms of challenging behaviour are reviewed due to their clinical significance and generally well defined nature. To generate an accurate and systematic review, the inclusion criteria for all studies will be the provision of prevalence of aggression and/or self-injury by age band data in addition to the number of participants in each age band so that these data might be analysed statistically. Studies will also be included if the number of participants per age band is not reported within the article, but can be obtained from the author(s). Whilst this will inevitably limit the number of studies included, this will enable a more robust assessment of the data.

Highlighting this issue, several frequently cited articles include data examining the prevalence of aggression and/or self-injury by age band, but the authors do not report the number of participants in each age band. Although these studies cannot be included in the review, the trends are worth noting. Both Oliver *et al.* (1987) and Borthwick-Duffy (1994) report the highest prevalence of self-injurious behaviour to be in the teenage years, whilst the highest prevalence rate reported by Rojahn (1986) was in a group of participants in their mid 20's. Conversely, Griffin *et al.* (1987) reported a decrease in the prevalence of self-injury in 14 to 22 year olds compared to younger individuals aged 4 to 14 years. With regard to aggression, Borthwick-Duffy also reported a slight increase in the prevalence of this behaviour in individuals after the age of 20, although this difference is not analysed statistically. Conclusions drawn from comparisons between the results of different studies should be tentative

however, as statistical significance of age related change in prevalence within studies is not evaluated. These results do nevertheless provide some indication of the kinds of trends demonstrated by published results not meeting criteria for inclusion in the study and allude to the need for a systematic review.

To summarise, the aim of this chapter is to conduct a systematic review of studies reporting the prevalence of aggression and self-injury by age band and thus to provide an evaluation of the association between age and two forms of challenging behaviour: self-injury and aggression.

2.3 Method

2.3.1 Search criteria

All peer reviewed published articles examining the relationship between depression and challenging behaviour 1967 and June 2011 were identified by a literature search using the search engine PsycINFO®. Table 2.1 lists the search terms that were employed. Both English and American spellings were included for all search terms.

Table 2.1: Terms employed in the literature search for studies reporting the prevalence of aggressive and self-injurious behaviour by age band

Search term	Variations
Aggression	Aggressive behavio*
Self-injury	Self-injurious behavio*, self-destruction, self-mutilation, auto mutilation
Challenging behavio*	Maladaptive behavio*, abnormal behavio*, problem behavio*, aberrant behavio*, externalising behavio*, behavio* disorder
Intellectual disability	Learning disability, mental retardat*, mental handicap*, developmental disabilit*
Age	Longitudinal, cross section*, prevalence, rate, time, aging, old

Whilst the prevalence rates of general challenging behaviour by age were not reviewed, search terms related to this behaviour were included to ensure no data regarding aggression or self-injury included as a subclass of challenging behaviour were overlooked. 'Intellectual disability' and variations of this term were included in order to limit the data reviewed to this population. The reference lists of all identified papers were also inspected to check for any omissions.

The inclusion criteria for studies were that they contained prevalence of aggression and/or self-injury by age band data in individuals with intellectual disability and information regarding the number of participants in each age band (studies without this information were included however if these data could be obtained from the author/s) so that raw data were available for statistical analysis. Within study statistical analysis of age related prevalence was adopted to overcome the difficulties of interpreting the results across studies using varying methodologies.

Fifteen studies, twelve regarding aggression and thirteen regarding self-injury (ten of the fifteen provided data regarding both aggression and self-injury), meeting these criteria were identified and included in this review. The sample and general methodology employed by each study are described in Tables 2.2 and 2.3, and illustrate the variability in the methodologies employed across studies.

Table 2.2: Methodology of twelve studies reporting the prevalence of aggression by age band

Study	Sample	Recruitment	Methodology	Measures of aggression	Measures of ID	Definition
Tavormina, Henggeler & Gayton (1976)	52 children aged 2-17 years. Most children assessed as being at 'trainable' range of disability or below in previous tests.	Mothers of children with ID living at home volunteered to participate	Unstructured Interview	93% agreement between raters coding aggression	No measure described	'Aggressive behaviour towards others.' Responses were grouped and two raters independently coded the target behaviours into categories.
Eyman & Call (1977)	6,870 individuals aged 0-13+ years. 57.3% mild-moderate, 21.3% severe, 21.3% profound ID.	Individuals with ID receiving services in America	Questionnaire	Items from the ABS (good reliability, no validity data)	No measure described	'Threatens or does physical violence'
Ando & Yohsimura (1978)	128 children aged 6-14 years (mean = 10.6). Children with ID, autistic and psychotic children excluded.	Students at a special school for children with ID and autism	Questionnaire completed by teachers and aides trained in use	Maladaptive behaviour scale (83% to 95% inter-rater reliability, no validity data)	ABS (good reliability, no validity data) and Suzuki-Binet (no reliability or validity data)	'Attack against other individuals is sometimes seen without overt external causes as well as in response to understandable causes'
Jacobson (1982)	30,578 individuals aged 0-65+ years. 19% mild, 22% moderate, 24% severe and 35% profound ID.	Individuals with ID living in a variety of settings	Population based survey. Data extracted from a database for the New York DDIS.	DDIS (no reliability or validity)	DDIS (no reliability or validity)	Aggression item records behaviour and frequency. No specific forms.

Study	Sample	Recruitment	Methodology	Measures of aggression	Measures of ID	Definition
Harris (1993)	901 individuals aged 8-85 years (mean = 34). More than 67.9% reported to have a severe ID, with ID ranging from mild to profound.	Population of one health district in the UK.	Interview	Bespoke interview (acceptable levels of reliability, no validity data)	No measure described	'Identified people who present serious problems…which may or may not result in injury to others… Behaviour rated for frequency (never to very often) and severity (no injury to very serious injury).'
Rojahn, Borthwick-Duffy & Jacobson (1993)	135,102, 1-45 years (mean = 28). 38% mild, 24% moderate, 18% severe, 20% profound.	All people with ID up to 45 years old receiving services in California and New York	Survey	CDER (satisfactory reliability and validity data) and the DDIS (no reliability or validity data).	CDER (satisfactory reliability and validity data) and the DDIS (no reliability or validity data).	'At least one violent episode causing physical injury had to have occurred within the past year'
Kobe, Mulick, Rash & Martin (1994)	203 'nonambulatory' individuals aged 6 months to 73 years (27.4 mean). Average developmental age equivalent between 1-29 months and 27-34 months depending on test used.	'Nonambulatory' persons residing in 2 intermediate care facilities for the 'mentally retarded'	Questionnaire, case note review and recent psychological evaluation	BPI (good reliability and validity).	Bayley scales of Infant Development (high reliability and good validity) and the Stanford Binet L-M (high validity but no reliability data).	No definition given

Study	Sample	Recruitment	Methodology	Measures of aggression	Measures of ID	Definition
Smith, Branford, Collacott, Cooper & McGrother (1996)	2,202 adults aged 18-93 years (mean = 37.7). 11.6% mild/borderline, 26% moderate, 31.5% severe, 30.9% profound ID.	Leicestershire learning disabilities register - interview key person involved with care of each registered adult with LD once every 5 years.	Questionnaire administered at interview	Bespoke questionnaire (low inter-rater reliability)	Bespoke questionnaire (low inter-rater reliability)	Based on frequency and severity. Severe challenging behaviour defined as behaviour of a severe nature or demonstrated three times per week.
Cooper (1998)	207 adults aged 20-65+ years	Leicestershire learning disabilities register. All people with ID aged 65+ years and a random sample of people less than 65 years of age.	Questionnaires and interviews	DAS (good reliability, no validity data).	VABS (good reliability and validity).	'Physical to people, significant due to severity or frequency (twice a month or more)… Not a sign of mental illness.' Excluding verbal aggression.
Deb, Thomas & Bright (2001)	101 individuals aged 16-64 years (mean = 37.7).	Randomly selected from a sample of people known to LD social services in a UK county.	Questionnaire and interview with patients and carers	Bespoke questionnaire (no reliability or validity) and the DAS (good reliability, no validity data).	Estimate of IQ obtained via questionnaire at interview (no reliability or validity)	'Aggression only included if rated severe and frequent (three times a week), less severe but frequent, severe but less frequent… No individual forms.'

Study	Sample	Recruitment	Methodology	Measures of aggression	Measures of ID	Definition
Crocker *et al.* (2006)	3,165 adults aged 18-60+ years (mean age = 40.63 years). 31.2% mild, 37.3% moderate, 18.9% severe and 12.6% profound.	Individuals receiving support from three learning disability services in Quebec	Questionnaire completed by educators who had known the client for 12 months	MOAS (high interater reliability and good validity but not in an ID sample).	Based on file and educator reporting (no reliability or validity)	'Acts displayed in the past twelve months ... defined as verbal and/or motor behaviour directed towards…others. It can be manifested directly or indirectly and can be more or less planned… Behaviours may not necessarily have led to injury but can potentially cause physical and psychological harm to… others and may present management difficulties.'
Tyrer *et al.* (2006)	3,062 adults aged 19-92. 23% mild, 20% moderate, 26% severe, 28% profound, 3% unknown ID.	Leicestershire LD register with interview data between 1993 and 2004	Interview and questionnaires	Questionnaire incorporating DAS (good reliability, no validity data).	No measure described	'Physically aggressive towards others during the last 12 months. Aggression present if it was severe and frequent (three times per week) or was severe but occurred less frequently or was considered less severe but occurred frequently.'

ABS = Adaptive Behaviour Scale, BPI = Behaviour Problems Inventory, CDER = Client Development Evaluation Report, DAS = Disability Assessment Schedule, DDIS = Developmental Disabilities Information Survey, ID = intellectual disability, LD = learning disability, MOAS = Modified Overt Aggression Scale, VABS = Vineland Adaptive Behavior Scale.

Table 2.3: Methodology of thirteen studies reporting the prevalence of self-injury by age band

Study	Sample	Recruitment	Methodology	Measures of self-injury	Measures of ID	Definition
Ballinger (1971)	626 individuals aged 0-68+ years. 31% mild/borderline, 23% moderate, 23% severe, 23% profound ID.	Patients 'mentally subnormal' hospital	Patient records and observations	Information from ward staff and observations	No measure described	'Painful or destructive act committed against their body in the last month whilst in hospital. Not accidents, tearing clothes, window breaking, swallowing dirt, nail biting or food refusal.'
Tavormina, Henggeler & Gayton (1976)	52 children aged 2-17 years. Most children assessed as being at 'trainable' range of disability or below in previous tests.	Mothers of children with ID living at home volunteered to participate	Unstructured Interview	93% agreement between raters coding aggression	No measure described	Self-mutilating behaviour. Responses were grouped and two raters independently coded the target behaviours into categories.
Eyman & Call (1977)	6,870 individuals aged 0-13+ years. 57.3% mild-moderate, 21.3% severe, 21.3% profound ID.	Individuals with ID receiving services in America	Questionnaire	Items from the ABS (good reliability, no validity data)	No measure described	'Does physical violence to self'
Ando & Yohsimura (1978)	128 children aged 6-14 years (mean = 10.6). Children with ID, autistic and psychotic children excluded.	Students at a special school for children with ID and autism	Questionnaire completed by teachers and aides trained in use	Maladaptive behaviour scale. 83% to 95% inter-rater reliability. No validity data.	ABS (good reliability, no validity data) and Suzuki-Binet (no reliability or validity data).	'Compulsive and conscious destructive behaviour against self.' No individual forms.

Study	Sample	Recruitment	Methodology	Measures of self-injury	Measures of ID	Definition
Jacobson (1982)	30,578 individuals aged 0-65+ years. 19% mild, 22% moderate, 24% severe and 35% profound ID.	Individuals with ID living in a variety of settings	Population based survey. Data extracted from database for the New York DDIS.	DDIS (no reliability or validity)	DDIS (no reliability or validity)	Self-injury item records behaviour and frequency. No forms.
Kebbon & Windahl (1986)	28,215 aged 1-82+ years. 24.7% mild, 34.6% moderate, 28.6% severe and 13.2% profound ID in SIB group.	Individuals in 22 counties (out of 25) in Sweden receiving services for ID during a 1 year census period	Questionnaire	Bespoke questionnaire (no reliability or validity data)	Judged by informants according to 4 categories corresponding to the ICD classification	'Must include an overt motor component. Frequency classified as behaviour observed daily, weekly, monthly or once/twice in three months.' No minimum intensity in terms of physical damage.
Rojahn *et al.* (1993)	135,102 aged 1-45 years (mean = 28). 38% mild, 24% moderate, 18% severe, 20% profound.	All people with ID up to 45 years old receiving services in California and New York	Survey	CDER (satisfactory reliability and validity data) and the DDIS (no reliability or validity data)	CDER (satisfactory reliability and validity data) and the DDIS (no reliability or validity data)	'Behavior occurs at least once a week and/or requires restraint as a preventative measure'

Study	Sample	Recruitment	Methodology	Measures of self-injury	Measures of ID	Definition
Kobe *et al.* (1994)	203 'nonambulatory' individuals aged 6 months to 73 years (27.4 mean). Average developmental age equivalent between 1-29 months and 27-34 months depending on test used.	'Nonambulatory' persons residing in 2 intermediate care facilities for the mentally retarded.	Questionnaire, case note review and recent psychological evaluation	BPI (good reliability and validity)	Bayley scales of Infant Development (high reliability and good validity) and the Stanford Binet L-M (high validity but no reliability data).	No definition given
Smith *et al.* (1996)	2,202 adults aged 18-93 years (mean = 37.7). 11.6% mild/borderline, 26% moderate, 31.5% severe, 30.9% profound ID.	Individuals with ID on the Leicestershire learning disabilities register	Questionnaire administered via interview	Bespoke questionnaire (low inter-rater reliability)	Bespoke questionnaire (low inter-rater reliability)	Based on frequency and severity. Severe challenging behaviour defined as behaviour of a severe nature or demonstrated 3 times per week.
Cooper (1998)	207 adults aged 20-65+ years	Leicestershire learning disabilities register. All people with ID aged 65+ and a random sample of people under 65 years.	Questionnaires and interviews	DAS (good reliability, no validity data).	VABS (good reliability and validity)	Results in negative consequences for the subject or occurs twice a month or more. Behaviour is not a response to a situation that other people in the same situation might often employ. Stereotypies and behaviour linked to mental illness excluded. No individual forms.

Study	Sample	Recruitment	Methodology	Measures of self-injury	Measures of ID	Definition
Saloviita (2000)	421 (mean = 32 years). 5% mild, 20% moderate, 29% severe, 43% profound and 3% unspecified ID.	Residents of an institution for people with ID	Questionnaires	ABS Part 2 (good reliability, no validity data)	Information from records and adaptive behaviour scale.	Type of SIB and frequency (occasional, frequent or not at all)
Deb *et al.* (2001)	101 individuals aged 16-64 years (mean = 37.7)	Randomly selected from a sample of people known to LD social services in a UK county	Questionnaire and interview with patients and carers	Bespoke questionnaire (no reliability or validity data) and the DAS (good reliability, no validity data).	Estimate of IQ obtained via questionnaire at interview	'Aggression only included if rated severe and frequent (three times a week), less severe but frequent, severe but less frequent… No individual forms.'
Crocker *et al.* (2006)	3,165 adults aged 18-60+ years (mean age = 40.63 years). 31.2% mild, 37.3% moderate, 18.9% severe and 12.6% profound.	Individuals receiving support from three learning disability services in Quebec	Questionnaire completed by educators who had known the client for 12 months	MOAS Self Aggression Subscale (high interater reliability and good validity but not in an ID sample)	Based on file and educator reporting (no reliability or validity)	"Verbal and/or motor behaviour directed towards oneself….It can be manifested directly or indirectly and can be more or less planned." Behaviours may not have led to injury but can potentially cause physical and psychological harm to self and may present management difficulties.

ABS = Adaptive Behaviour Scale, BPI = Behaviour Problems Inventory, CDER = Client Development Evaluation Report, DAS = Disability Assessment Schedule, DDIS = Developmental Disabilities Information Survey, ID = intellectual disability, LD = learning disability, MOAS = Modified Overt Aggression Scale, VABS = Vineland Adaptive Behavior Scale.

The reliability and validity of the measures used to assess both aggression and self-injury as well as severity of intellectual disability in each study were examined (where available) in order to appraise the quality of each, but not to exclude, given the paucity of papers providing prevalence of challenging behaviour by age band.

2.3.2 A summary of the methodologies employed by the studies identified

Of the fifteen studies included in the review, ten used total population samples, two used child only samples, two recruited from institutions and one used 'nonambulatory' participants only. Almost half of the studies (six) utilised questionnaires, two used interviews and two employed surveys. The remaining studies employed a combination of methods, including questionnaires, interviews, observations and reference to case notes. With regard to the measures of aggression and/or self-injury used, eight studies used measures which are well established within the literature whilst five used bespoke measures. One study used both established and bespoke measures.

2.3.3 Data analysis

For each study, relative risks were conducted to identify if a significant increase or decrease in the relative risk of aggression or self-injury was evident when comparing older age bands to the youngest age band in the study. Consequently, the youngest age band in each study was selected as the index group and other age groups in the same study were then used as a comparison to generate an index of age related change. 99% confidence intervals were used due to the number of relative risks calculated. Relative risks were deemed significant if both the upper and lower confidence intervals did not encompass a value of one. A significant relative risk greater than one indicates a

significantly greater likelihood of aggression or self-injury in an older age group, whilst a significant relative risk less than one indicates a significantly decreased relative risk of aggression or self-injury in an older age group. For the purpose of presentation, results were tabulated so that the rows of each table demonstrate increasing age in the right hand columns. Each individual cell in the comparison column equates (approximately) to one five year age band and individual cells are merged to denote age bands of multiples of five years.

2.4 Results

2.4.1 The prevalence of aggression by age

In order to investigate the prevalence of aggression by age, the prevalence of this behaviour by age band as described by the twelve studies identified were examined. These results are shown in Table 2.4.

Table 2.4: Prevalence of aggression % (n) by age bands (years) for the twelve studies identified meeting criteria. Each cell contains the italicised age band, as well as the % prevalence and number of participants in parentheses.

Study	Prevalence of aggression % (n) by age band (years)														
Tavormina *et al.* (1976)	*2-4*	*4-6*	*8-12*	*12-17*											
	0 (0)	35 (6)	15 (2)	33 (3)											
Eyman & Call (1977)	*0-12*				*13+*										
	28.5 (464)				27.5 (1442)										
Ando & Yohsimura (1978)	*6-9*	*11-14*													
	11.1 (5)	1.4 (1)													
Jacobson (1982)	*0-21*				*22+*										
	8.5 (669)				11.3 (2164)										
Harris (1993)	*5-9*	*10-14*	*15-19*	*20-24*	*25-29*	*30-34*	*35-39*	*40-44*	*45-49*	*50-54*	*55-59*	*60-64*	*65-69*	*70+*	
	10.6 (5)	14.9 (10)	20.2 (17)	24.7 (19)	16.5 (22)	11.9 (10)	28.9 (26)	18.6 (13)	21.8 (12)	6 (3)	22.9 (8)	11.8 (4)	16 (4)	12 (6)	
Rojahn *et al.* (1993)	*0-10*	*11-20*	*21-45*												
	7.09 (2095)	11.62 (2991)	14.13 (11274)												
Kobe *et al.* (1994)	*0-6*	*7-12*	*13-21*	*22-39*	*40-73*										
	0 (0)	24 (6)	22.2 (12)	26 (19)	33.3 (14)										
Smith *et al.* (1996)	*20-29*	*30-39*	*40-49*	*50-59*	*60+*										
	22.45 (183)	21.5 (116)	18.1 (73)	20.45 (48)	22.5 (47)										
Cooper (1998)	*20-64*	*65+*													
	6.8 (5)	5.2 (7)													
Deb *et al.* (2001)	*16-29*	*30-45*	*46-64*												
	31 (11)	21.5 (116)	14.7 (5)												
Crocker *et al.* (2006)	*18-29*	*30-39*	*40-49*	*50-59*	*60+*										
	26.8 (194)	24.5 (190)	28.5 (258)	15.1 (72)	22.4 (64)										
Tyrer *et al.* (2006)	*19*	*20-29*	*30-39*	*40-49*	*50-59*	*60-69*	*70+*								
	24 (57)	16 (105)	17 (122)	13 (82)	9 (38)	9 (23)	6 (9)								

Direct comparison of these results across studies is difficult due to variability in the samples employed. To rectify this problem, the studies were divided into three categories: those using a total population (subdivided into child and adult and adult only samples), child only and an otherwise specified sample. In the case of aggression, the latter category included one study using a 'nonambulatory' sample. Relative risks were calculated to compare the relative risk of aggression in each older age band compared to the youngest age band, the index group, in order to identify differences in the prevalence of aggression with increasing age in each study.

2.4.1.1 Total population studies of aggression

Table 2.5 shows the relative risks across age bands for each total population study, child and adult and adult only samples, examining the prevalence of aggression.

Table 2.5: Relative risks for the prevalence of aggression for each older comparison age group as compared to the index group for each total population study (child and adult samples above and adult samples only below the bold line). Each cell denotes a five year age band and cells are merged to signify multiple age bands. The index group is the first cell on the left of each row.

Paper	Index and comparison groups														
Eyman & Call (1977)	*0-12 Index*	*13+*													
		.97 (.86, 1.08)													
Jacobson (1982)	*0-21 Index*	***22+***													
		1.33 (1.19, 1.48)													
Harris (1993)		*5-9 Index*	*10-14*	*15-19*	*20-24*	*25-29*	*30-34*	*35-39*	*40-44*	*45-49*	*50-54*	*55-59*	*60-64*	*65-69*	*70+*
			1.4 (.37, 5.28)	1.9 (.56, 6.48)	2.32 (.69, 7.74)	1.55 (.47, 5.17)	1.12 (.3, 4.24)	2.72 (.84, 8.76)	1.75 (.49, 6.2)	2.05 (.57, 7.33)	.56 (.09, 3.45)	2.15 (.56, 8.32)	1.11 (.22, 5.65)	1.5 (.3, 7.51)	1.13 (.26, 4.91)
Rojahn *et al.* (1993)	*0-10 Index*	***11-20***			***21-45***										
		1.64 (1.53, 1.76)			**1.99 (1.88, 2.11)**										
Tyrer *et al.* (2006)		*19 Index*	***20-29***	*30-39*	***40-49***	***50-59***	***60-69***	***70+***							
			.66 (.46, .97)	.71 (.49, 1.02)	**.54 (.36,.81)**	**.38 (.23, .62)**	**.37 (.2, .67)**	**.26 (.11, .63)**							
Smith *et al.* (1996)			*20-29 Index*	*30-39*	*40-49*	*50-59*	*60+*								
				.96 (.73, 1.26)	.8 (.58, 1.11)	.92 (.63, 1.33)	1.01 (.7, 1.46)								
Cooper (1998)			*20-64+ Index*											*65+*	
														.77 (.18, 3.36)	
Deb *et al.* (2001)		*16-29 Index*	*30-45*	*46-64*											
			.74 (.25, 2.17)	.48 (.14, 1.68)											
Crocker *et al.* (2006)		*18-29 Index*	*30-39*	*40-49*	***50-59***	*60+*									
			.92 (.73, 1.15)	1.07 (.86, 1.31)	**.56 (.41, .78)**	.83 (.6, 1.15)									

Bold = $p < .01$

As demonstrated in Table 2.5, the relative risk of aggression increased significantly with age in two of the studies identified with total population samples employing child and adult participants. Illustrating this, the results of Jacobson's (1982) study indicated a significant increase in the relative risk of aggression in adults aged 22 years or over (RR = 1.33, CI = 1.19, 1.48) compared to individuals aged between 0 and 21 years. The results of Rojahn *et al.*'s (1993) study also indicate that compared to participants aged between 0 and 10 year olds, 11 to 20 (RR = 1.64, CI = 1.53, 1.76) and 21 to 45 (RR = 1.99, CI = 1.88, 2.11) year olds are at significantly greater relative risk of aggression.

Using an adult only sample, the results of Tyrer *et al.'s* (2006) study suggest a general decrease in the relative risk of aggression with increasing age, so that adults aged between 20 and 29, 40 and 49, 50 and 59, 60 and 69 and 70 years or more were all at significantly less relative risk (RR range = .26 to .66) of demonstrating aggression than individuals aged less than 19 years. The only exception to this was adults aged between 30 and 39 years, for whom the relative risk of aggression was not significantly different to those aged less than 19 years (relative risk = .71, CI = .49, 1.02). The results of Crocker *et al.* (2006) also indicated a significant decrease in the relative risk of aggression in 50 to 59 year olds (RR = .56, CI = .41, .78) as compared to 18 to 29 year olds, although there were no significant differences in the relative risk of aggression for the 30 to 39, 40 to 49 or 60 years or more age bands as compared to the index group.

2.4.1.1.1 The relative risk of aggression according to the results of total population studies using modified age bands

The results regarding the prevalence of aggression by age according to total population studies thus initially appear to be conflicting, although these discrepant results might be due to the divergent index groups employed by each study. Thus, in order to more accurately compare the results of each study, further relative risk analyses were conducted using modified index and older age bands selected to be as similar to the age bands used in comparative studies as possible.

Examining the total population studies employing child and adult samples, there appear to be discrepant results within these broadly similar samples. In contrast to the results of Rojahn *et al.* (1993) and Jacobson (1982), both Eyman and Call (1977) and Harris (1993) failed to detect any significant changes in the risk of aggression with age. Eyman and Call's results however might have been influenced by the very large older age band used masking any significant trends within this older group, although using far more narrow age bands, Harris also failed to identify any significant difference in the relative risk of aggression with age. Thus, in order to clarify these results, relative risk analyses were conducted using age bands from Harris' data made similar to those used by Rojahn *et al.* (further analysis to compare Harris' results to those of Jacobson were not conducted as the upper age band of this latter study was not accurately defined).

This analysis demonstrated that according to Harris' (1993) results, the relative risk of aggression did not differ significantly with age so that participants aged between 10 and 19 years (RR = 1.68, CI = .52, 5.47) and 20 and 44 years (RR = 1.86, CI = .61,

5.7) were at no greater relative risk of aggression than participants aged between 5 and 9 years, in contrast to Rojahn *et al.'s* (1993) results which indicated an increased relative risk of aggression in 11 to 20 (RR = 1.64) and 21 to 45 (RR = 1.99) year olds. Whilst the results based on Harris' modified age bands were not significant, they were however similar to the relative risks produced from Rojahn *et al.'s* results.

It might be hypothesised that the significantly reduced relative risk of aggression with age in two of the total population studies employing adult only samples was the result of the older index group utilised, so that the increase in the relative risk of aggression has already occurred in the index group and thus shows no significant difference to the older age groups. To test this hypothesis, the age bands utilised in Harris' (1993) study were again altered to match those utilised by Crocker *et al.* (2006) in order to examine whether the results gained are affected by the index group utilised (age groups were not altered in accordance with Tyrer *et al.*, 2006 or Cooper, 1998 due to the extremely narrow index group and large comparative older age group used in each study respectively). Using an index group of participants aged 20 to 29 years to compare to Crocker *et al.'s* findings, relative risk analysis indicated that participants aged between 30 and 39 (RR = 1.06, CI = .63, 1.79), 40 and 49 (RR = 1.02, CI = .57, 1.84), 50 and 59 (RR = .66, CI = .29, 1.49) and 60 years of more (RR = .66, CI = .31, 1.38) were at no greater relative risk of aggression than participants aged between 20 and 29 years. These results are similar to those of Crocker *et al.* except for the significantly reduced relative risk of aggression identified in 50 to 59 (RR = .56) year olds in Crocker *et al.'s* study, although again, whilst not reaching significance, the results gained from Harris' modified age bands were similar.

Since these modified age bands were the same as those employed by Smith *et al.* (1996), the results produced were also compared to those of Smith *et al*. This comparison indicated similarities between the data, so that there were no significant differences in the relative risk of aggression with age, although the relative risk figures were quite different. Finally, in order to examine how the use of the older index groups had affected the results of Deb *et al.* (2001), the age bands employed by Harris (1993) were further modified in order to accurately compare to the results of these two studies. Employing an index group of participants aged between 15 and 29 years, relative risk analysis indicated that participants aged between 30 and 44 years (RR = 1.02, CI = .65, 1.59) and 45 and 64 years (RR = .79, CI = .45, 1.36) were at no significantly different relative risk for aggression to the younger index group, results similar to Deb *et al.*, although the relative risk figures were quite different.

To summarise, the results of two total population studies of aggression employing child and adult samples indicated an increase in the relative risk of this behaviour with age. Using an older index group, two total population studies employing adult only samples indicated a decrease in the relative risk of aggression with age. Further relative risk analysis based on the results of Harris' (1993) modified age bands illustrated similar relative risks to these studies (although the results were not significant) indicating the potential influence of the older index group employed in these studies, although a real decrease in the prevalence of aggression in later life could also exist. Modifying Harris' age bands to fit those of Smith *et al.* (1996) and Deb *et al.* (2001) also indicated no significant differences in the relative risk of aggression with age.

2.4.1.2 Studies of aggression with child only samples

The relative risks across age bands for each study with a child only sample (Ando & Yoshimura, 1978; Tavormina *et al.,* 1996) examining the prevalence of aggression were calculated. The results of these analyses indicate that the relative risk of aggression does not significantly increase with age. Illustrating this, the results of Ando and Yoshimura showed that 11 to 14 year olds were at no greater relative risk of aggression than 6 to 9 year olds (RR = .13, CI = .01, 2.02), whilst Tavormina *et al.* also did not identify a significantly greater relative risk of aggression in 8 to 12 (RR = .44, CI = .07, 2.86) and 12 to 17 year olds (RR = .94, CI = .21, 4.16) as compared to an index group of children aged 4 to 6 years.

2.4.1.3 A study of aggression with an otherwise specified sample

The relative risks across age bands for a study employing a 'nonambulatory' sample (Kobe *et al.,* 2004), were calculated, investigating the risk of aggression in the youngest versus the older age bands. The results of this analysis indicated that in 'nonambulatory' individuals, there is no greater relative risk of aggression in individuals aged between 13 and 21 (RR = .93, CI = .3, 2.86), 22 and 29 (RR = 1.08, CI = .38, 3.1) or 40 and 73 years (RR = 1.37, CI = .47, 4.08) as compared to an index group of 7 to12 year olds.

2.4.2 The prevalence of self-injury by age

In order to investigate the prevalence of self-injury by age, the prevalence of this behaviour by age band as described by the thirteen studies identified were examined. These results are shown in Table 2.6.

Table 2.6: Prevalence of self-injury % (n) by age bands (years) for the thirteen studies identified meeting criteria. Each cell contains the italicised age band, as well as the % prevalence and number of participants in parentheses.

Study	Prevalence of self-injury % (n) by age band (years)									
Ballinger (1971)	*0-9*	*10-19*	*20-29*	*30-39*	*40-49*	*50-59*	*60-69*	*70+*		
	17.5 (7)	17.9 (18)	17.9 (27)	11.15 (13)	12.8 (14)	9.1 (6)	21.2 (7)	12.5 (1)		
Tavormina *et al.* (1976)	*2-4*	*4-6*	*8-12*	*12-17*						
	0 (0)	17.6 (3)	30.8 (4)	22.2 (2)						
Eyman & Call (1977)	*0-12*	*13+*								
	14.9 (243)	15.2 (797)								
Ando & Yoshimura (1978)	*6-9*	*11-14*								
	6.7 (3)	4.2 (3)								
Jacobson (1982)	0-21	22+								
	7.7 (551)	9.3 (1723)								
Kebbon & Windahl (1986)	*< 1*	*2-11*	*12-21*	*22-31*	*32-41*	*42-51*	*52-61*	*62-71*	*72-81*	*82+*
	0 (0)	4.8 (152)	20.5 (1360)	32.5 (1928)	22.1 (1073)	10.4 (303)	5.5 (131)	2.8 (43)	1.3 (8)	0 (0)
Rojahn *et al.* (1993)	*0-10*	*11-20*	*21-45*							
	7.1 (2100)	8.4 (2167)	9.05 (7212)							
Kobe *et al.* (1994)	*0-6*	*7-12*	*13-21*	*22-39*	*40-73*					
	20 (1)	32 (8)	35.2 (19)	58.9 (43)	61.9 (26)					
Smith *et al.* (1996)	*20-29*	*30-39*	*40-49*	*50-59*	*60+*					
	21.2 (173)	19.45 (105)	16.1 (65)	9.3 (22)	9.2 (19)					
Cooper (1998)	*20-64*	*65+*								
	2.7 (2)	3 (4)								
Saloviita (2000)	*0-17*	*18-34*	*35+*							
	34 (19)	47 (90)	36 (63)							
Deb *et al.* (2001)	*16-29*	*30-45*	*46-64*							
	20 (7)	35.5 (11)	17.6 (6)							
Crocker *et al.* (2006)	18-29	30-39	40-49	50-59	60+					
	22 (159)	28.4 (220)	26.6 (240)	20.2 (96)	19.9 (57)					

As for aggression, studies were divided into three categories: those using a total population (subdivided into child and adult and adult only samples), child only and an otherwise specified sample. The latter category included three studies, two using participants recruited from institutions and a third employing a 'nonambulatory' sample. Relative risks were calculated to compare the likelihood of self-injury in each older age band compared to the youngest age band and in order to identify any differences in the relative risk of self-injury with increasing age in each study.

2.4.2.1 Total population studies of self-injury

Table 2.7 shows the relative risks across age bands for each total population study, child and adult and adult only samples, examining the prevalence of self-injury.

Table 2.7: Relative risks for the prevalence of self-injury for each older comparison age group as compared to the index group for each total population study (child and adult samples above and adult samples only below the bold line). Each cell denotes a five year age band and cells are merged to signify multiple age bands. The index group is the first cell on the left of each row.

Study	Index and comparison groups							
Eyman & Call (1977)	*0-12 Index*	*13+*						
		1 (.97, 1.03)						
Jacobson (1982)	*0-21 Index*	***22+***						
		1.29 (1.14, 1.45)						
Kebbon & Windahl (1986)	*2-11 Index*	***12-21***	***22-31***	***32-41***	***42-51***	*52-61*	***62-71***	***72-81***
		4.27 (3.45, 5.29)	**6.18 (5.49, 8.36)**	**4.6 (3.71, 5.71)**	**2.17 (1.69, 2.77)**	1.15 (.85, 1.55)	**.59 (.38, .91)**	**.27 (.11, .68)**
Rojahn *et al.* (1993)	*0-10 Index*	***11-20***	***21-45***					
		1.19 (1.1, 1.28)	**1.27 (1.2, 1.35)**					
Smith *et al.* (1996)		*20-29 Index*	*30-39*	*40-49*	***50-59***	***60+***		
			.92 (.69, 1.22)	.76 (.54, 1.07)	**.43 (.24, .78)**	**.44 (.26, .77)**		
Cooper (1998)		*20-64 Index*				*65+*		
						1.09 (.12, 9.86)		
Deb *et al.* (2001)		*16-29 Index*	*30-45*	*46-64*				
			.74 (.25, 2.17)	.48 (.14, 1.68)				
Crocker *et al.* (2006)		18-29 *Index*	***30-39***	*40-49*	50-59	*60+*		
			1.29 (1.03, 1.63)	1.21 (.96, 1.52)	.92 (.68, 1.24)	.9 (.62, 1.29)		

Bold = $p < .01$

In contrast to aggression, four out of the eight total population studies investigating the prevalence of self-injury with age identified an increased likelihood of this behaviour with increased age. Illustrating this, the results of studies by Kebbon and Windahl (1986) and Rojahn *et al.* (1993) indicated that after the age of ten or eleven respectively, the likelihood of self-injury significantly increases with age up until 51 and 45 years respectively (RR range = 1.29 to 6.18). The results of Crocker *et al.* (2006) also indicated that compared to 18 to 29 year olds, 30 to 39 year olds were at significantly higher relative risk of self-injury (RR = 1.29, CI = 1.03, 1.63). The age band with the highest relative risk in each study varied between the teenage years to mid-adulthood (11 to 20 in Rojahn *et al.'s*, 22 to 31 in Kebbon & Windahl's and 30 to 39 in Crocker *et al.'s* study). Jacobson (1982) also illustrated an increased likelihood of self-injury in individuals aged 22 years and over as compared to individuals aged 21 years and younger (RR = 1.29, CI = 1.14, 1.45).

Additionally, further results by Kebbon and Windahl (1986) and those provided by Smith *et al.* (1996) indicate a decreased risk of self-injury in older adults with intellectual disabilities (RR range = .27 - .59). Whilst there is some discrepancy between studies with regard to the exact age of the start of this decline, individuals around 50 years of age and older appear to be significantly less likely to demonstrate self-injury (RR range = .27-.59). The remaining studies did not show any significant differences between age groups with regard to the likelihood of self-injury.

2.4.2.1.1 The relative risk of self-injury according to the results of total population studies using modified age bands

In order to compare more accurately the results of studies using both child and adult and adult only samples, the age bands used by Kebbon and Windahl (1986) were modified and made as comparable as possible to the age bands of Smith *et al.* (1996), Deb *et al.* (2001) and Crocker *et al.* (2006) so that further relative risk analyses could be conducted. Kebbon and Windhal's results were selected for modification due to the flexibility provided by the ten year age bands used. Age bands were not modified in accordance with Cooper (1998) as these were deemed too large to warrant useful analysis.

Using an index group of participants aged between 22 and 31 years of age, similar to that of Smith *et al.* (1996) and Crocker *et al.* (2006), relative risk analyses indicated that the relative risk of self-injury significantly decreased in participants aged between 32 and 41 (RR = .68, CI = .62, .74), 42 and 51 (RR = .28, CI = .37), 52 and 61 (RR = .17, CI = .14, .21) and 62 years or more (RR = .2, CI = .14, .28). These results broadly replicate the results of Smith *et al.* who also identified a significant reduction in the relative risk of self-injury in participants aged over 50, although not those aged between 30 and 49 years of age. These results are in contrast to those of Crocker *et al.* who identified a significant increase in the relative risk of self-injury in 30 to 39 year olds.

Using a younger index group similar to that of Deb *et al.* (2001), relative risk analyses indicated that participants aged between 32 and 41 (RR = .84, CI = .79, .91) and 42 and 61 (RR = .31, CI = .28, .36) were at significantly less relative risk of self-injury than

participants aged between 12 and 31 years, in contrast to Deb *et al.* who found no significant differences in the prevalence of self-injury with age.

Thus, despite some conflicting results, a general curvilinear relationship between self-injury and age has been identified in several total population studies, so that the risk of self-injury significantly increases with age up until a certain point when the relative risk begins to significantly decrease. The point at which the relative risk of self-injury begins to change is, however, unclear. Modification of the index group and older age bands used provided similar results to one (Smith *et al.,* 1996), but not the two other studies (Deb *et al.,* 2001; Crocker, *et al.,* 2006) using adult only samples.

2.4.2.2 Studies of self-injury with child only samples

The relative risks across age bands for the studies examining the prevalence of self-injury in child only samples (Tavormina *et al.,* 1976; Ando & Yoshimura, 1978) were calculated. The results of Tavormina *et al.'s* study indicated that there was no significant difference in the relative risk of self-injury with age so that participants aged between 8 and 12 years (RR = 1.74, CI = .31, 9.8) and 12 and 17 years (RR = 1.26, CI = .15, 10.3) were at no greater relative risk of aggression than participants aged between 4 and 6 years. Similarly, the results of Ando and Yoshimura's study indicated that children aged between 11 and 14 years were at no greater risk for demonstrating self-injury (RR = .63, CI = .08, 4.85) than children less than ten years of age. These results indicate that the likelihood of self-injury does not differ with age in children with intellectual disabilities.

2.4.2.3 Studies of self-injury with otherwise specified samples

Table 2.8 shows the relative risks across age bands for each study employing an otherwise specified sample examining the prevalence of self-injury.

Table 2.8: Relative risks for prevalence of self-injury for each older comparison age group as compared to the index group for each study with 'other' populations. Each cell denotes a five year age band and cells are merged to signify multiple age bands. The index group is the first cell on the left of each row.

Study	**Index and comparison groups**							
Ballinger (1971)	*0-9* *Index*	*10-19*	*20-29*	*30-39*	*40-49*	*50-59*	*60-69*	*70+*
		1.01 (.36, 2.86)	1 (.37, 2.69)	.63 (.21, 1.93)	.73 (.25, 2.19)	.52 (.14, 1.98)	1.21 (.33, 4.18)	.71 (.05, 9.34)
Kobe *et al.* (1994)	*0-6* *Index*	*7-12*	*13-21*	*22-39*	*40-73*			
		1.6 (.14, 18.12)	1.76 (17, 18.56)	2.95 (.29, 30.01)	3.1 (.3, 31.77)			
Saloviita (2000)	*0-17* *Index*		*18-34*	*35+*				
			1.39 (.83, 2.34)	1.07 (.62, 1.84)				

With no significant relative risks in the results of any of the studies demonstrated in Table 2.8, it appears that there is no increased likelihood of self-injury with age in a study with a 'nonambulatory' sample (Kobe *et al.,* 1994) or those recruited from institutions (Ballinger, 1971; Saloviita, 2000).

2.5 Discussion

The aim of this systematic review was to examine age related changes in the prevalence of aggression and self-injury in individuals with intellectual disability. Generating relative risk analyses from the results of the studies identified with prevalence of aggression and/or self-injury by age band data allowed an examination of trends in the results across many studies utilising varying samples and age bands. This within study analysis approach overcame difficulties in interpreting the results across studies using varying methodologies whilst tabulating these analyses provided a clear representation of the results across studies employing similar general methodologies. Whilst studies were not excluded on the basis of employing measures of challenging behaviour or intellectual disability with poor or no reported reliability or validity, the methodologies employed by each study were reported and the psychometrics of all measures used were assessed in order to examine the quality of the results produced by each study. By analysing and tabulating the relative risk analyses and examining the methodologies employed by each identified study, conclusions can be drawn based on an understanding of the strengths and limitations of each study.

Drawing conclusions based on the results of the studies identified is difficult due to the often diverse methodologies and samples employed. Clustering the studies based on

generally similar sample recruitment (i.e. total population, child only and otherwise specified samples) allowed any consensus between the results within clusters to be deciphered for both aggression and self-injury. With regard to aggression, studies utilising a total population sample appeared to show somewhat conflicting results. Illustrating this, the results of Jacobson's (1982) and Rojahn *et al.'s* (1993) studies employing child and adult samples indicated an increased risk of aggression with age. However, also employing a child and adult sample, both Eyman and Call (1977) and Harris (1993) failed to identify this trend, although these results might have been caused by the very large age band used in Eyman and Call's study, masking any significant trends within this older group and the relatively small sample compared to the number of age bands employed by Harris.

Utilising an adult only sample, Tyrer *et al.* (2006) reported a general decline in the prevalence of aggression with age, as demonstrated by the significant decrease in the relative risk of aggression in participants (a result partially supported by Crocker *et al.* 2006). However, further relative risk analysis using Harris' (1993) modified age bands, made to be similar to those of Crocker *et al.* (Harris' age bands could not be modified in accordance with Tyrer *et al.*'s due to the small index group utilised in this study) indicated that the results gained by this study might have been influenced by the older index group used. Indeed, close inspection of the relative risk analyses conducted show that despite not reaching significance, the relative risk results produced from Harris' modified aged bands were remarkably similar to those of Rojahn *et al.* (1993) and Crocker *et al.*, indicating that a lack of power caused by an inadequate sample size in Harris's study rather than the lack of age related change in aggression might account

for these non significant findings. This supposition is supported upon examination of the wide confidence intervals demonstrated in the analyses of Harris' findings.

Relative risk analysis based on the modified age bands of Harris (1993), when made comparable to those of Smith *et al.* (1996) and Deb *et al.* (2001) also showed similarities across findings, with no significant differences in the relative risk of aggression identified with increasing age. The relative risks produced however were not similar across studies. The methodologies of Smith *et al.* and Deb *et al.* might have influenced these results however, with both studies using bespoke measures of aggression with no or low reported levels of reliability. The sample employed by each study was also small in comparison to other total population studies (e.g. Jacobson, 1982; Rojahn *et al.,* 1993) particularly in the study of Deb *et al.*. It might also be surmised however that these results indicate that the decreased prevalence of aggression in later life demonstrated by theses studies is not caused by the older index group utilised, but instead illustrate a real decline in adulthood.

Thus, methodological weaknesses inherent in some of these total population studies must be considered so that conclusions can be drawn from these data based on an understanding of the limitations and strengths of each study. As discussed previously, the results of Smith *et al.* (1996) and Deb *et al.* (2001) are limited by the measures and samples used. Using a bespoke questionnaire with acceptable levels of reliability, Harris' (1993) study appears to be more methodologically robust, but is also limited by the relatively small sample employed, as is the case with Cooper (1998). A further limitation of Cooper's and both Eyman and Call's (1977) and Jacobson's (1982) studies is the large age bands employed, potentially masking any changing prevalence

of aggression within the age bands. Only including participants aged less than 45 years of age, Rojahn *et al.* (1993) also used, although smaller, larger than ideal age bands. Employing adequately large samples and established measures of aggression, the studies conducted by Crocker *et al.* (2006) and Tyrer *et al.* (2006) appear to be more methodologically robust. However, the use of an adult index group, and in Tyrer's case a very small index group, might alter the results gained so that they do not present a broad dataset with regard to the prevalence of aggression with age.

Thus, it is clear that none of the total population studies identified are free from threats to validity although, some are more methodologically robust than others. Whilst employing a relatively small sample and failing to describe the measure of intellectual disability employed, Harris (1993) utilised a reliable interview and useful age bands and produced relative risk values similar to that of studies with far larger samples. By recruiting a very large sample, the results of Rojahn *et al.* (1993) can also be considered to be generalisable. The use of at least one reliable, established measure of aggression and intellectual disability also adds weight to the accuracy of these results. In conclusion, the results of these studies indicate that, given a large sample, a general increase in the prevalence of aggression with age can be detected in total population samples, although it is unclear as to whether this increase continues beyond 45 years of age. Indeed, the results of several studies using adult only samples (e.g. Deb *et al.,* 2001; Tyrer *et al.,* 2006) indicate that the prevalence of aggression might decrease in later life, so that aggression might show a curvilinear relationship with age. Whilst some of the results using modified age bands indicated that the older index group used might account for these findings, real decreases in the prevalence of aggression in later life and the potential influence of healthy survivor effects cannot be ruled out.

With regard to the results of studies using child only (Ando & Yoshimura, 1978; Tavormina *et al.,* 1976) and 'nonambulatory' samples to examine the prevalence of aggression with age, the relative risk of aggression did not appear to change with age. These results might be taken to indicate differences in the prevalence of aggression with age depending on particular characteristics, such as mobility. It might also be deduced that age related changes in the prevalence of aggression with age commence in adulthood, a supposition tentatively supported by the results of the total population studies. However, these results should be considered in light of the relatively small samples used by these few studies.

Relative risk analysis based on the results of total population studies of self-injury also appeared to illustrate an increase in the prevalence of self-injury with age until mid-adulthood and the association between age and increased prevalence of self-injury was stronger than that observed for aggression. To illustrate, analysis of the results of studies by Kebbon and Windahl (1986) and Rojahn *et al.* (1993) indicated that the risk of self-injury significantly increases with age after eleven and ten years of age respectively until approximately 50 years of age, results also broadly supported by two further studies (Jacobson, 1982; Crocker *et al.*, 2006) utilising slightly older index groups.

A decreased risk of self-injury in older adults with intellectual disabilities was also found to commence in participants around the age of 50 years or more (Kebbon and Windahl, 1986; Smith *et al.,* 1996), illustrating a curvilinear relationship between self-injury and age. However, the remaining total population studies of self-injury did not identify any significant associations. As for aggression, these conflicting results are

likely to be due to the varied methodologies and samples used. For example, the discrepancy between the results of Kebbon and Windahl's and Smith *et al.'s* studies might be due to the less severely disabled sample employed by the latter.

In order to compare more accurately the results of studies using both child and adult and adult only samples, the age bands used by Kebbon and Windahl (1986) were modified and made as similar as possible to the results of Smith *et al.* (1996), Deb *et al.* (2001) and Crocker *et al.* (2006). The results of this analysis indicated that the differing results produced by total population studies employing adult only samples might be due to a factor other than the older index group used. Demonstrating this, the relative risk analysis conducted using Kebbon and Windahl's modified age bands illustrated a significant decrease in the relative risk of self-injury with age in participants older than 32, as compared to participants aged between 22 and 31. These results were in contrast to those of Crocker *et al.* whose results indicated a significant increase in the relative risk of self-injury in 30 to 39 year olds and Deb *et al.* whose results showed no significant difference in the relative risk of the behaviour with age. Similarities to the relative risks produced from Smith *et al.'s* data however were found in that both sets of analyses indicated a significantly decreased risk of self-injury in participants aged between approximately 50 and 60 years.

These discrepancies between the results of analyses based on Kebbon and Windahl's study (1986) and the three adult only studies might be due to the slight differences in index groups and older age bands used, as whilst age bands were made as similar as possible, small differences between the age bands across studies remained. The lack of psychometric data reported for the bespoke questionnaire used in Kebbon and

Windahl's study might also limit its usefulness as a comparative adult and child sample study. As previously discussed with regard to aggression, the poor reliability of the measures employed by Smith *et al.* (1996) and the small sample used by Deb *et al.* (2001) limit the accuracy of the data produced. Despite the large sample used, Jacobson's (1982) study is also flawed due to the lack of psychometric data reported for the measure of aggression used and infinitely large older age band, a limitation also true of Eyman & Call's (1977) study. Only including participants aged less than 45 years of age, Rojahn *et al.* (1993) also used smaller, but larger than ideal, age bands.

Despite these methodological limitations, the results of three out of the four total population studies using child and adult samples, as well as Crocker *et al.'s* (2006) study, indicated that the relative risk of self-injury increased with age, up until approximately 40 years. This consensus was reached despite the varying definition of self-injury employed, sample origin, specific measures used and age of the index group, indicating the robust nature of this finding. The presence of a curvilinear relationship between self-injury and age however is less clear. Identified by both Smith *et al.* (1996) and Kebbon and Windahl (1986), Crocker *et al.* however did not identify this trend despite the use of narrow age bands, a sample larger than that employed by Smith *et al.* and an established measure of aggression. The index group employed by this study also appeared to be unrelated to these results, although the modified age bands used did not match exactly those of Crocker *et al.* which might have affected the results.

As was also the case with aggression, the relative risk of self-injury did not appear to significantly differ with age in studies employing child only or otherwise specified

samples. These results might thus indicate that significant increases in the prevalence of self-injury begin later in early adulthood, although such broad conclusions cannot be based on the results of only two studies with limited samples. Similarly, both studies including participants recruited from institutions failed to identify any significant changes in the risk of self-injury with age. Whilst both studies appear to be methodologically robust, such findings might be the result of the small time window imposed in Ballinger's (1971) study. The lack of information provided by Saloviita (2000) with regard to the definition of self-injury also means that any conclusions based on the results of this study must be made carefully.

The results of this review indicate that the prevalence of challenging behaviour with age might depend on the form of challenging behaviour examined. Illustrating this, the relative risk of self-injury appears to significantly increase with age up until approximately 50 years, at which point the relative risk of self-injury begins to decrease significantly, indicating a curvilinear association between self-injury and age, although this relationship is proposed tentatively due to conflicting results and the small number of studies illustrating the relationship. The prevalence of aggression also appears to increase with age until mid-adulthood, although this relationship was found in relation to fewer studies. A decrease in the prevalence of aggression in later life might also be indicated by the results of this systematic review, although the use of older index groups in studies demonstrating this association might also be responsible for these results. Thus, the association between age related changes in challenging behaviour appear to be less clear in the case of aggression than self-injury.

Comparisons made between the results of this review and studies reporting age related prevalence of challenging behaviour data, but no age band participant numbers, indicate some similarities. For example, replicating the results of Oliver *et al.'s* (1987) total population study, Rojahn *et al.* (1993) also reported the highest prevalence rates of self-injury to be in 11 to 20 year olds. With regard to aggression, Borthwick-Duffy (1994) also reported a slight increase in the prevalence of this behaviour in individuals after the age of 20, although this difference is not analysed statistically. Conclusions from this systematic review and similarities with other studies outside of it however must be drawn tentatively due to the small number of studies identified with prevalence of aggression and/or self-injury by age band data and the methodological limitations inherent in many of these studies.

Whilst it is quite widely accepted by researchers working in the field of intellectual disabilities that challenging behaviour becomes more prevalent with increasing age, the results of this literature review indicate that this assumption might not be as infallible as originally thought. The difficulty in coming to such conclusions is caused by a paucity of methodologically robust studies employing large, generalisable samples aimed specifically at investigating challenging behaviour with age. Exemplifying this, much of the data examined in this literature review was collated from studies reporting prevalence of challenging behaviour by age data as a by-product of the main aims of the study. As discussed in the introduction, several studies providing age band data also had to be excluded from the analysis as they failed to report the number of participants per age band, thus the results of this systematic review are only based on a sample of the already published studies investigating prevalence of aggression and self-injury by age.

Basing conclusions regarding age related change of challenging behaviour in people with intellectual disabilities on the results of cross-sectional surveys is also problematic due to difficulties in separating out age related and cohort effects within this data. Differential mortality against the general population means that age-specific rates of challenging behaviour in people with intellectual disabilities have typically been found to peak in adolescence or young adulthood and then decline (Oliver *et al.,* 1987; Borthwick-Duffy, 1994). Additionally, early mortality is more common among people with particular genetic syndromes and more severe intellectual disabilities, both of which may be linked to the occurrence of challenging behaviour (Lesch & Nyhan, 1964; Shear, Nyhan, Kirman & Stern, 1971). Cohort effects might also exist, whereby differential mortality against the general population is changing due to increased longevity, thus, younger cohorts might differ from older ones (Janicki, Dalton, Henderson & Davidson, 1999). Potential interactions between these healthy survivor and cohort effects create difficulties in attributing age related effects in cross-sectional data.

Within the studies providing this age band data, there are also large differences in the methodologies and samples employed which has a significant effect on the results. Broadly encompassing individuals with intellectual disabilities, the average severity of intellectual disability of samples was not always clearly defined in the studies, partially due to the use of outdated terminology (e.g. Tavormina *et al.,* 1976) or measures without easily comparable scores (e.g. Kobe *et al.,* 1994). Several studies included in this systematic review utilised large, representative samples, however, a few were more limited, either in terms of the size or nature of the sample (e.g. Tavormina, *et al.*;

Ando & Yoshimura, 1978). Having recruited large, representative samples, the results of some studies were also hindered by the large age bands described, the use of which might conceal the changing prevalence of aggression or self-injury within more narrow age ranges (e.g. Cooper, 1998; Jacobson, 1982). What was also unclear in each study was the distribution of participants' age within the age bands, so that assumptions are made about the relative risk for a particular age band of participants, even though the average age of that group might not be the mean of the age band range.

With regard to the methodology, whilst some authors used explicit definitions of challenging behaviour, including criteria for length of time demonstrated, severity of behaviour and any exclusions (e.g. Ballinger, 1971), other used loose definitions, and did not stipulate exact criteria (e.g. Kobe *et al.,* 1994). Indeed, the criteria for inclusion of behaviour as self-injurious appeared to differ quite dramatically across studies, an issue illustrated by Saloviita (2000) who included provoked abuse from others, which might not be considered as self-injurious in more traditional definitions of the behaviour.

By taking into account these methodological limitations, the quality of each study can be interpreted and thus varying results reported understood. Whilst not all the results of the studies identified illustrated these trends, a few deemed to be methodologically robust appeared to show that age was more strongly associated with self-injury than aggression and that age potentially had a more robust curvilinear relationship with self-injury than aggression. Indeed, apparent decreases in the prevalence of aggression in later life might merely be the result of the older index group used as the basis of the statistical analysis of several studies. This has important implications on both a

theoretical and clinical level. In terms of research, the focus might now shift to the theoretical underpinnings of this discrepancy between forms of challenging behaviour, as well as conducting more methodologically robust studies aimed specifically at investigating the prevalence of various forms of challenging behaviour with age, as conclusions based on the prevalence of specific forms of challenging behaviour might change given the results of more methodologically robust studies. To date, much attention has been paid to the development of challenging behaviour generally, although comparatively less to the ontogeny of specific forms. Guess and Carr's (1991) stage model however does provide a detailed account for the development of self-injury, indicating its emergence from repetitive behaviours as a unique course and whilst the results of this literature review cannot support this model in any concrete fashion, they do suggest potentially different developmental progressions of different forms of challenging behaviour, as proposed in the model. This supposition is also supported by previous research which has also indicated that self-injury is related to health problems and pain (e.g. Carr & Owen-DeSchryver, 2007; de Lissovoy, 1962; Hart, Bax & Jenkins, 1984; Luzzani, Macchini, Valade, Milani & Selicorni, 2003) although these characteristics have not been widely associated with aggression.

More generally, these results also have wider implications for this field of research as a whole, indicating that assumptions, such as those based on the increased prevalence of challenging behaviour with age should be rigorously tested so that more accurate conclusions from existing research can be drawn. From a clinical perspective, understanding the prevalence of challenging behaviour with age is important in terms of service development and provision. For example, those providing services to young and middle-aged adults with intellectual disabilities need to be aware of the potentially

increased prevalence of self-injury and possibly aggression in this age group so that they might provide adequate intervention resources. Additionally, services for younger individuals with intellectual disabilities should remain alert to the onset of self-injury and aggression, the risk of which is likely, or in the case of aggression could, increase with age, so that services can prepare for the future needs of this population and early intervention can be effectively targeted.

2.6 Summary and implications

In Chapter 2, a systematic review of studies reporting the prevalence of aggression and self-injury by age band was conducted to provide an evaluation of the association between age and two forms of challenging behaviour: self-injury and aggression. Relative risk analyses indicated that both self-injury and aggression appear to increase with age until middle age, although this association may be stronger for self-injury than aggression. Additionally, self-injury appears to demonstrate a curvilinear relationship with age, so that self-injury appears to decrease after mid-adulthood. This might also be true for aggression, although the relationship appears to be less robust and might merely be an artefact of the older index age group used as the basis of these results.

These results indicate that prior assumptions regarding the association between challenging behaviour and age appear correct, but that more methodologically robust studies aimed specifically at investigating the prevalence of challenging behaviour with age are needed to confirm these conclusions. Research should also focus on the development of specific forms of challenging behaviour. These results also have implications for service development and provision, indicating that early intervention

targeted at younger children who appear to be at significantly lower relative risk of aggression and self-injury might help to prevent the development of challenging behaviour in individuals with intellectual disabilities.

For such early intervention services to be effective however, greater understanding needs to be developed regarding the child characteristics associated with challenging behaviour so that individuals at highest risk of challenging behaviour and those most in need of early intervention can be identified and prioritised. The aim of Chapter 3 is thus to identify these child characteristics or risk markers of challenging behaviour in young children with intellectual disabilities and to develop predictive models of challenging behaviour.

Chapter 3

Investigating Risk Markers for Severe Challenging Behaviour in Young Children with Severe Intellectual Disabilities

3.1 Abstract

Background and aims: Challenging behaviour has a significant impact on the lives of individuals with a severe intellectual disability and their families, with implications for service delivery and early intervention. This study investigated putative risk markers for challenging behaviour in children with a severe intellectual disability through the development of the Challenging Behaviour Screening Questionnaire (CBSQ).

Method: The CBSQ was completed by teachers regarding 629 participants aged between 2 and 12 years from schools for children with severe intellectual disabilities in the West Midlands.

Results: Analyses showed that children with 'probable ASD' and those demonstrating repetitive and restricted behaviours and interests (a composite of repetitive and restricted behaviour) and overactivity/impulsivity (a composite of overactive and impulsive behaviour) were at significantly greater relative risk of all forms of challenging behaviour. In terms of severe challenging behaviour, the relative risk of aggression and one or more forms of challenging behaviour was significantly increased by repetitive and restricted behaviours and interests and overactivity/impulsivity and if the child had been labelled as having 'probable ASD'. The presence of one or more health problems was also significantly correlated with the presence and severity of self-injurious behaviour.

Discussion: These findings are in accordance with previous research and indicate the possibility of identifying children at highest risk of developing severe challenging behaviour and thus those most in need of early intervention.

3.2 Introduction

Challenging behaviour has a significant impact on the lives of individuals with a severe intellectual disability and their families with implications for effective service delivery. Studies examining the prevalence of severe challenging behaviour in individuals with an intellectual disability have reported rates within the range of 5 to 17% (Borthwick-Duffy, 1994; Emerson & Bromley, 1995; Emerson *et al.,* 2001a; Lowe *et al.,* 2007; Qureshi & Alborz, 1992). The majority of research investigating the prevalence of challenging behaviour across the life span of individuals with an intellectual disability has also demonstrated an increase in the prevalence of this behaviour until early to mid-adulthood (e.g. Eyman & Call, 1977; Harris, 1993; Jacobson, 1982; Oliver, Murphy & Corbett, 1987; Sigafoos, Elkins, Kerr & Attwood, 1994). Indeed, the results of the literature review (section 2.4) indicated that the relative risk of both self-injury and aggression appears to increase with age until mid-adulthood, although this association may be stronger for self-injury than aggression. Additionally, self-injury appears to demonstrate a curvilinear relationship with age, so that self-injury appears to decrease after mid-adulthood. This might also be true for aggression, although the relationship appears to be less robust and might merely be an artefact of the older index age group used as the basis of these results. Research has also indicated that once established challenging behaviour is often persistent (Chadwick, Kusel, Cuddy & Taylor, 2004; Emerson *et al.,* 2001b; Kebbon &

Windahl, 1986; Murphy *et al*., 1993; Nottestad & Linaker, 2002; Schroeder, Schroeder, Smith, & Daldorf, 1978).

The prevalence and persistence of challenging behaviour indicates the seriousness of this issue for people with intellectual disabilities, particularly as research has also indicated that the quality of life of individuals demonstrating challenging behaviour is compromised (Emerson, 2001). Illustrating this, challenging behaviour is one of the most common reasons for placement in more restrictive facilities (Lakin, Hill, Hauber, Bruininks & Heal, 1983; Tausig, 1985) and is often distressing for families of individuals with challenging behaviour (Hastings & Brown, 2002; Qureshi, 1995). Additionally, the cost involved in service provision for individuals with challenging behaviour is problematic for the already financially stretched NHS (Knapp, Comas-Herrera, Astin, Beecham & Pendaries, 2005), because although a relatively small population compared to the intellectual disability population in general, individuals with intellectual disability and challenging behaviour require a disproportionate amount of resources (Mansell, 1992).

The paucity of effectively delivered interventions available to individuals with an intellectual disability and challenging behaviour does little to ease this situation. Whilst the behavioural interventions provided to reduce challenging behaviour have received robust empirical support (e.g. Kahng, Iwata & Lewin, 2002), these interventions are time consuming, costly, difficult to implement due to a lack of appropriately trained staff and are often only provided once the challenging behaviour has become so ingrained within the individuals' behavioural repertoire that modification is difficult (Murphy *et al.,* 1993; Robertson *et al.,* 2005). Whilst

pharmacological treatments are exempt from much of this criticism, the use of this form of intervention is somewhat controversial due to polypharmacy and associated side effects (Baumeister, Todd & Sevin, 1993).

Reviews of the use of drug interventions have generated mixed results regarding their effectiveness with some support for the use of specific medications (Deb & Unwin, 2007), although it is widely acknowledged that much of this evidence is based upon methodologically flawed research (Matson *et al.,* 2000). Conversely, in a systematic review of the literature, Brylewski and Duggan (1999) concluded that there was no evidence of whether antipsychotic medication helps or harms adults with intellectual disability and challenging behaviour.

The amalgamation of these factors alludes to the importance of an effective early intervention strategy, which has already been successfully utilised in various disciplines such as health (Blanks, Moss, McGahan, Quinn & Babb, 2000), education (NESS, 2005) and more specifically autism (Cohen, Amerine-Dickens & Smith, 2006; Eikeseth, Smith, Jahr & Eldevik, 2007; Lovaas, 1987; Remington *et al.,* 2007). Early intervention has also been strongly advocated in recent government policy (e.g. 'Every Child Matters,' Department for Education and Skills, 2003). It is hoped that by providing interventions for challenging behaviour when individuals with intellectual disabilities 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. It might also be proposed that the challenging behaviour demonstrated by younger children could be less resistant to interventions due to the lesser amount of operant

reinforcement experienced by younger individuals, thus enhancing the effectiveness of the intervention implemented.

To successfully execute early intervention strategies, those most at risk of developing challenging behaviour must be identified before the behaviour becomes too well established in their behavioural repertoire. The presence of putative risk markers (i.e. characteristics that predate the development of challenging behaviour and thus are potentially predictive of it) in children with a severe intellectual disability might be used to identify these children so that they can be prioritised for early intervention. Whilst no empirical studies have provided direct evidence for the development of the putative risk markers predating challenging behaviour, comparisons of separate studies investigating the development of challenging behaviour and the putative risk markers in isolation indicate this relationship.

Several studies investigating the prevalence of challenging behaviour in individuals with intellectual disabilities have illustrated acceleration in the proportions showing the behaviour and the severity of behaviour between the teenage years and mid to late 20's (Kebbon & Windahl, 1986; Oliver, Murphy & Corbett, 1987; Rojahn *et al.* 1993). In contrast, research investigating the development of behavioural characteristics that predict challenging behaviour such as repetitive, restricted, overactive and impulsive behaviour, has shown an earlier age of development. By the age of 2 to 3 years, many children with intellectual disabilities are already demonstrating stereotyped behaviour and interests (Lord, 1995; Berkson & Tupa, 2000; Young, Brewer & Pattison, 2003). A study examining the age of onset of ADHD has also shown that over 80% of participants with ADHD who met symptom

criteria had an age of onset of impairment less than 7 years (Applegate *et al.,* 1997). Many of these participants demonstrated symptoms of hyperactivity and impulsivity as young as 1 year of age, indicating that, like repetitive behaviours and interests, these behaviours develop earlier than the age at which the proportion of people showing challenging behaviour rises dramatically.

Numerous studies using a variety of methodologies and samples have examined various child characteristics that might be putative risk markers for challenging behaviour in individuals with intellectual disabilities. Chadwick, Piroth, Walker, Bernard and Taylor (2000) conducted one of the first studies to examine risk markers for challenging behaviour in children with a severe intellectual disability (114 participants aged between 4 and 11 years). These authors reported an association between severity of intellectual disability and destructive and self-injurious behaviour. Ambulant and younger children were also found to demonstrate significantly more destruction.

Investigating risk markers for self-injurious behaviour in children with autistic disorders (222 participants aged between 2 and 7 years), Baghdadli, Pascal, Grisis and Aussilloux (2003) identified three significant predictors of self-injury; greater delay in adaptive skill, more severe autism and presence of a perinatal condition. Younger children were also significantly more likely to demonstrate self-injury. However, due to the restricted nature of the sample, these findings cannot be easily generalised to children with intellectual disabilities, a problem rectified by a large scale meta-analysis conducted by McClintock, Hall and Oliver (2003). Examining data from 22 prevalence and cohort studies of challenging behaviour in individuals with an

intellectual disability, the meta-analysis broadly replicated previous findings and identified a significant positive association between severity of intellectual disability and the prevalence of self-injury and destruction. Self-injury was also significantly associated with poor communication skills. Finally, individuals with autism were also found to be at significantly higher risk of demonstrating aggression, destruction and self-injury. The potential overlap between these variables was not controlled for however, which, along with the difficulties in interpreting findings from studies utilising a range of methodologies and samples represent limitations of this meta-analysis.

In a large scale total population study of individuals with intellectual disabilities conducted by Holden and Gitlesen (2006) (904 participants, aged between 0 and 89 years) challenging behaviour occurred far more frequently among people with than without autism whilst self-injurious behaviour in particular was more common in individuals with a more severe degree of intellectual disability (although the opposite was true for aggression). More 'demanding' challenging behaviour was also significantly associated with age. Several putative risk markers, such as age and presence of autism have thus received much support in the literature. More recently however, studies examining the putative risk markers of challenging behaviour have focused on behavioural characteristics.

Illustrating this, Oliver *et al.* (in preparation) identified high frequency repetitive and ritualistic behaviour as predictors of the presence and severity of aggression, destruction and self-injury, a finding first reported in children with the rare genetic syndrome Cornelia de Lange Syndrome (Oliver, Sloneem, Hall & Arron, 2009) and

supported by a review (Petty & Oliver, 2005) and recent study (Arron, Oliver, Berg, Moss & Burbidge, in review). Also partially replicating this finding in a much smaller sample of children under the age of five, Petty *et al.* (in preparation) also showed a positive association between repetitive, overactive and impulsive behaviours and a number of challenging behaviours, indicating their roles as putative risk markers for challenging behaviour. A major strength of both Oliver *et al.'s* (in preparation) and Petty *et al.'s* study was the use of binary logistic regressions which controlled for the potential overlap between variables, which was a significant limitation in McClintock *et al.'s* (2003) meta-analysis. The identification of overactive and impulsive behaviour as putative risk markers has also been supported by the recently identified associations between self-injurious and aggressive behaviour and attention deficit hyperactivity disorder, also in individuals with intellectual disabilities (Cooper, Smiley, Allan *et al*., 2009; Cooper, Smiley, Jackson *et al*., 2009).

Additionally, Petty *et al*. (in preparation) reported a positive association between health problems and severity of self-injurious behaviour. Whilst this result was on the border of significance, a wealth of previous research has demonstrated a significant association between health and challenging behaviour, in both typically developing children and individuals with intellectual disabilities. Elevated rates of problem behaviour in association with physical illness in typically developing children are commonly reported (de Lissovoy, 1962; Hart, Bax & Jenkins, 1984). Carr and Owen-DeSchryver (2007) also identified health as a potential setting event for challenging behaviour, reporting higher frequency and intensity of problem behaviour on 'sick' than 'well' days in minimally verbal children with developmental disabilities. More severe pain and discomfort was also associated with more frequent and severe

challenging behaviour. An association between pain caused by gastro-oesophageal reflux and self-injury has also been reported in children with Cornelia de Lange Syndrome (Luzzani, Macchini, Valade, Milani & Selicorni, 2003).

Evidence for a causal link between physical illness and challenging behaviour is provided by several studies documenting a reduction in challenging behaviour following medical intervention (Ghaziuddin, Elkins, McNeeley & Ghaziuddin, 1993; Peine *et al.,* 1995). The prevalence of health problems in children with intellectual disabilities also appears to be particularly high (Berg, Arron, Burbidge, Moss & Oliver, 2007; Cooper, 1998), indicating health's viability as a risk marker for challenging behaviour.

However, some putative risk markers have received mixed evidence from the literature. For example, whilst McClintock *et al.* (2003) found that males were more likely to be aggressive, numerous other studies have failed to identify any association between gender and challenging behaviour (e.g. Baghdadli *et al.,* 2003; Chadwick *et al.,* 2000; Holden & Gitlesen, 2006). Similarly, several studies have failed to find significant associations between age and challenging behaviour (Einfeld & Tonge, 1996; Quine, 1986), despite younger individuals appearing to demonstrate more challenging behaviours in some studies (Baghdadli *et al.*; Chadwick *et al.*). The results of the literature review conducted in Chapter 2 indicate that the relationship between age and challenging behaviour might depend on the form of challenging behaviour, as well as the age of the sample (section 2.4). Numerous studies have also identified an association between severity of intellectual disability and challenging behaviour, indicating its role as a robust risk marker. However, several more recent

studies (with the exception of Chadwick *et al.*) investigating challenging behaviour in children with a severe intellectual disability have failed to identify such strong associations between severity of intellectual disability and challenging behaviour, with Petty *et al.* (in preparation) identifying no association and Oliver *et al*. (in preparation) detecting an association between severity of intellectual disability and self-injury only. These results might indicate that although severity of intellectual disability is a robust risk marker, it is less influential in individuals with a severe intellectual disability and thus those who are already at high risk.

Consequently, the aim of the present study is to examine those child characteristics acting as putative risk markers for challenging behaviour in children with a severe intellectual disability, in order to examine further child characteristics which have received mixed support in the literature and also to further investigate those only recently identified. An administrative definition of severe intellectual disability will be used in this study, so that participants are assumed to have a severe intellectual disability due to their attendance at severe learning disability schools. Whilst an administrative definition of severity of intellectual disability might lead to the inclusion of some more able participants, it is deemed sufficient and necessary considering the large population required to examine the putative risk markers and test their suitability for indicators of being at high risk in an early intervention context. Thus the sample employed in this study is assumed to comprise of children with a severe intellectual disability, although it is accepted that there might be some variation in severity of intellectual disability within the sample.

A secondary aim of this study is to investigate these risk markers through the development of a brief screen. Each of the studies examining child characteristics associated with challenging behaviour already discussed was limited through the use of long questionnaires to measure the putative risk markers and challenging behaviours. Whilst this is currently the easiest way of reliably and validly measuring these risk markers, such a methodology can result in difficulties when evaluating large samples. In order to benefit as many children as possible, early intervention programmes would need to screen large numbers of children with intellectual disabilities. This is far more likely given the required completion of a short and accessible screening tool, as opposed to several lengthy questionnaires.

Thus, as well as examining the ability of various child characteristics to predict the presence and severity of challenging behaviour, a further aim of this study is to develop a screen, the goal of which is to provide a robust and accessible measure of these putative risk markers and specific forms of challenging behaviour. Such a screen would prove valuable to clinicians and teachers as a way of predicting, with a known margin of error, those children in their care who are most likely to develop severe challenging behaviour. Such early identification would also enable practitioners to prevent the development of clinically significant challenging behaviour before it becomes ingrained within the child's behavioural repertoire, thus ultimately reducing the degree of challenging behaviour demonstrated by children with severe intellectual disabilities.

A final, and more theoretical, aim of this study is to develop predictive models of challenging behaviours which would control for the overlap between the putative risk

markers and thus examine whether different forms of challenging behaviour were predicted by different putative risk markers. As well as being interesting from a theoretical view point, further understanding the associations between particular risk markers and forms of challenging behaviour might prove clinically significant in the effective treatment of challenging behaviour in early intervention programmes.

The hypotheses for this study are that:

1. The presence of 'probable ASD' will significantly increase the relative risk of
    a. The presence of challenging behaviour
    b. The severity of challenging behaviour

    although the exact forms are not predicted.

    It is also hypothesised that the severity of intellectual disability will not be significantly associated with the presence or severity of challenging behaviour as participants might already be considered to be at high risk of challenging behaviour due to their severity of intellectual disability. A non significant association between age and challenging behaviour is also hypothesised due to the young age of participants. Due to the mixed evidence with regard to gender, predictions will not be made with regard to its associations with the presence or severity of challenging behaviour.

2. The presence of the behavioural variables repetitive, restricted, impulsive and overactive behaviour will significantly increase the relative risk of
    a. The presence of challenging behaviour
    b. The severity of challenging behaviour

    although the exact forms are not predicted.

3. The presence and severity of different forms of challenging behaviour will be significantly predicted by different models, for example, children with one or more health problems will demonstrate significantly more self-injury, but not aggression or destruction.

3.3 Method

3.3.1 Development of the Challenging Behaviour Screening Questionnaire (CBSQ)

The main design aim for the CBSQ was to keep its length to a minimum whilst reliably and validly measuring the putative risk markers. Questionnaires measuring each variable were chosen if they had been previously used 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.

3.3.1.1 Criteria for risk marker item inclusion in the CBSQ

Items measuring specific risk markers were selected for inclusion in the screen if they fitted the majority of the following criteria:

1. Reported associations between item construct and challenging behaviour
2. High prevalence of the measured construct in children with a severe intellectual disability
3. High inter item and total item reliability scores
4. Appropriateness for use in a population of children with a severe intellectual disability

5. Appropriateness for completion by teachers - The screen was designed to be completed by teachers as previous studies have received poor return rates from parents (Petty, 2006)

Two versions of the CBSQ were developed for participants aged less than 6 years (Appendix A) and those aged 6 years and older (Appendix B). Each version was exactly the same except for the measure of severity of intellectual disability used in each.

3.3.2 CBSQ Contents – Putative risk marker and challenging behaviour measures

Age

The sample was divided into two age groups using a median split (7 years and under and 8 years and over).

'Probable Autism Spectrum Disorder (ASD)'

Teachers were asked to state if a professional had ever said that the child was autistic, or had an autistic spectrum disorder, autistic like traits and/or features of autism. If the teacher endorsed any one of these variables, the child was scored as having 'probable ASD'.

Severity of Intellectual Disability

a) **Denver Developmental Screening Test II (DDST II; Frankenburg, Dodds, Archer, Shapiro & Bresnick, 1992) -** Twenty items from DDST II were used to assess developmental delay in children aged less than 6 years. Based on the

original Denver Developmental Screening Test (DDST) developed by Frankenburg & Dodds (1967), the test contains 125 items measuring personal-social, fine motor, gross motor and language skills. The mean percentage agreement for inter-rater and test-retest reliability reported by the authors was high (99.7% and 87.5% respectively). High correlations between the DDST II and other similar scales, such as the Revised Yale Development Schedule (.95; Frankenburg, Camp & Van Natta, 1971), also indicates good concurrent validity of the test. Content validity of the DDST II has not been reported, however, the screen is well established within the literature.

Items were chosen to represent all four subscales (five from each) and a range of ages (developmental age for each item chosen so that 90% of the original standardisation sample accomplished the milestone between 2.1 months and 3.7 years). For scoring, the DDST II items were arranged in order of lowest to highest developmental age required to achieve them and participants were given the score which matched the highest endorsed item. To calculate a Denver developmental quotient, each participant's Denver score was divided by their chronological age in months.

b) **The Wessex Behaviour Scale (Kushlick, Blunden & Cox, 1973) -** Three items from the Wessex Behaviour Scale were used as an estimate of intellectual disability and nine for level of sensory impairment, physical disability and adaptive behaviours for children aged between 6 and 12 years. The original Wessex Behaviour Scale measures physical disability, adaptive behaviour, communication and sensory impairment through nine subscales

with items scored on a 3 point scale whereby 1 indicates severe incapacity, 2 mild incapacity and 3 no incapacity. The data derived from these items can be categorised into the Social and Physical Incapacity (based on ratings of continence, mobility and behaviour problems) and the Speech Self-Help and Literacy subscales (based on ratings of an individuals ability to speak, read, write, count, wash, dress and feed themselves). Kushlick *et al.* (1973) reported a high degree of reliability for the scale, with good reliability scores found for all items, including incontinence (80%), mobility (92%), speech (82%), self-help (78%) and literacy (78%). Palmer and Jenkins (1982) also established good inter-rater reliability for the scale across both child and adult populations and residential and non-residential settings. The authors reported Kappa values of .62 (range = .54 to .72) for overall classification and a mean individual item reliability of .54 (range = .33 to .89).

The twelve items selected were all taken from the original twelve item Wessex incapacities subscale. Severity of intellectual disability was assessed using items regarding washing, dressing and feeding which were summed to produce the range 3 to 9, with a lower score representing a greater severity of intellectual disability. Also, one item was used to assess physical disability (scored between 1 and 3), two items for continence (scored between 1 and 3 for both wetting and soiling), two for literacy (scored on a range of 1 to 3 for reads and writes), one for numeracy (scored on a range of 1 to 3), one regarding speech (scored between 1 and 3) and two regarding sensory impairment (scored between 1 and 3 for both vision and hearing).

Overall Severity of Intellectual Disability Score

Disability percentile scores were calculated to generate comparable Denver and Wessex scores. Greater and lesser severity of intellectual disability groups were formed using median splits on these disability percentile data for both the under 6 and 6 years and older groups and then combined to form one high and low group for the whole sample.

Health Questionnaire (Hall, Arron, Sloneem & Oliver, 2008)

Six items from the Health Questionnaire were included in the screen to assess health problems in this sample. The questionnaire contains fifteen health conditions which are rated by caregivers for presence and severity over the previous month or the individual's life time. Hall *et al.* calculated the mean item level reliability kappa coefficients for the previous month and for lifetime as .76 (range = .32 to 1.00) and .72 (range = .32 to 1.00) respectively. Intra-class correlation co-efficient scores for the overall health problem score and total number of health problems occurring over the last month were .65 and .73 respectively. Those across the person's lifetime were .71 and .68 respectively.

The six items from the Health Questionnaire used for this screen referred to eye, ear, dental, digestive, skin and any other health or painful conditions. These items were used due to their reported association with challenging behaviour in the literature (de Lissovoy, 1963; Kravitz, 1964, Luzzani *et al.,* 2003; Oliver *et al.,* in preparation) and their high prevalence in children with an intellectual disability (Böhmer, Klinkenberg-Knol & Niezen-De Boer, 2002; van Schrojenstein Lantman-de Valk, Metsemakers, Haveman & Crebolder, 2000). Teachers rated the extent to which these conditions had

affected children in their class in the last month on a 4 point Likert scale ranging from 0 (never) to 3 (severe). A total health score was calculated through aggregating item responses with a possible range of 0 to 18. Two health groups were formed for later analysis; one or more health problems and no health problems.

Self-Help and Behaviour Rating Scale (Petty, 2006)

Two items from the behaviour and emotional difficulties section of the Self-Help and Behaviour Rating Scale were used to assess frequency and severity of repetitive and restricted behaviour. This scale is an adapted version of the Wessex Behaviour Scale (Kushlick, Blunden & Cox, 1973) and contains ten items referring to hyperactive, uncooperative, resistant, self-injurious, aggressive, destructive, repetitive, anxious, obsessional/ritualistic and problem behaviour. An acceptable level of inter-rater reliability for the frequency and management difficulty of self-injury, aggression and destruction was found (frequency = .51, .63 and .46 respectively; management difficulty = .44, .44 and .42 respectively). Test retest reliability data is not available for the Self-Help and Behaviour Rating Scale, although it is assumed that it would also be acceptable since test retest is normally higher than inter-rater reliability.

The two items included from this scale referred to repetitive movements and obsessions and rituals and were chosen due to their association with challenging behaviour. Items were scored on two 5 point scales; frequency, which ranges from 1 (never) to 5 (very often) and management difficulty, which ranges from 1 (not difficult to manage) to 5 (seriously difficult to manage). Children scoring 4 or 5 on frequency of repetitive and restricted behaviour formed the repetitive and restricted behaviour group, whilst those scoring 1, 2 or 3 were considered not to have the

putative risk marker. This composite was formed due to the highly significant association between repetitive and restricted behaviour (r = .65, $p < .001$). Severity of repetitive and restricted behaviour was not included in later analysis due to its highly significant correlation with frequency of these behaviours (r = .65, $p < .001$ and r = .64, $p < .001$ respectively). For brevity, repetitive and restricted behaviours and interests is abbreviated to RRBI in Figure 3.2.

Activity Questionnaire (Burbidge, 2005)

Four items from the Activity Questionnaire were used in order to assess overactive and impulsive behaviour. The eighteen item questionnaire contains three subscales: overactivity, impulsivity and impulsive speech. Two items were taken from both the overactive and impulsivity scales. No items from the impulsive speech subscale were used as impulsive speech was not considered to be a reliable measure of activity in a population, a large proportion of which were likely to be nonverbal. Items were selected based on their appropriateness for participants with a large age and mobility range and their level of inter-rater (ranging from .5 to .75) and test-retest (ranging from .72 to .81) reliability scores. All items selected were also significantly correlated with self-injurious behaviour, physical aggression and destruction in a population of children with severe intellectual disability.

The frequency of these behaviours was rated along a 5 point Likert scale which ranged between 0 (never/almost never) and 4 (always/almost all the time). The overactive and impulsive subscale scores, each ranging from 0 to 8, were derived from aggregating items 3 and 9, and 17 and 18 respectively. A total scale score was calculated by combining the two subscale scores (range = 0 to 16). These items all

had good inter-rater and test-retest reliability ranging from .5 to .81. Median splits were used to form two groups so that participants scoring 3 or above formed the overactivity/impulsivity group and those scoring 2 or below were considered not to have the putative risk marker. This composite was formed due to the highly significant correlation between overactivity and impulsivity ($r = .804$, $p < .001$). For brevity, overactivity/impulsivity is abbreviated to O/I in Figure 3.2.

Challenging Behaviour Questionnaire (CBQ; Hyman, Oliver & Hall, 2002) and Challenging Behaviour Interview (Part II) (CBI; Oliver *et al.,* 2003)

Three items to assess the presence of aggressive, destructive and self-injurious behaviour were taken from the Challenging Behaviour Questionnaire. Kappa values for the three forms of challenging behaviour were .85, .75 and .92 respectively. Presence of challenging behaviour groups were formed based on whether the teacher had reported that the child did or did not demonstrate each form of challenging behaviour. Endorsements were aggregated to provide information regarding the number of forms (0-3) of challenging behaviour displayed by each participant. Items 1, 2 and 13 from the Challenging Behaviour Interview were used to assess the severity of these challenging behaviours. According to the authors, these items have an inter-rater agreement of .78, .68 and .54 respectively. The internal consistency of these items is acceptable with correlation coefficients between the three items calculated as .39, .51 and .46 and item-total correlations of .56, .43 and .69. The total scale Alpha was also reduced if any of these items were removed. For each type of challenging behaviour, items 1, 2 and 13 were used to examine the longest episode of the behaviour, the need for physical restraint due to this behaviour and the frequency of this behaviour in the last month respectively, all based on caregiver report. These

items were all scored on a 5 point Likert scale. Item 2 was scored on a range of less than a minute (1) to more than an hour (5), item 3 was scored on a range of never (1) to at least once an hour (5) and item 13 was scored on a range of by this time next month (1) to in the next 15 minutes (5). Participants were classed as showing severe challenging behaviour if they scored within the top 20% of composite scores for the three questions from the CBI.

3.3.3 Reliability of the CBSQ

Inter-rater reliability data for particular CBSQ items was provided by raters both within the same and across two different environments. Various studies examining the inter-rater reliability of items included in the CBSQ contemporaneously indicated good inter-rater reliability for the severity of intellectual disability (as measured by the Wessex, .66) health (ranging from .35 to .83; Hall *et al.,* 2008), repetitive and restricted behaviours and interests (.48) and overactive (.54 and .65) and impulsive behaviour (.54 to .81; Burbidge, 2005) as well as the frequency of aggression (.63), destruction (.46) and self-injury (.51) (Petty, 2006) and the severity of these challenging behaviours (range = .5 to .72; Hyman *et al.,* 2002).

A study conducted in Essex provided inter-rater reliability for the CBSQ across different environments whereby parents and teachers completed the CBSQ regarding 54 children with a severe intellectual disability aged between 2 and 12 years. These data, analysed using Spearman's Rho correlations, indicated satisfactory reliability for each of the following variables: autism (.81), severity of intellectual disability (Wessex = .47; Denver = .48), health (ranging between .34 and .57) and repetitive and restricted behaviours and interests (.21). The correlation coefficient for

overactivity/impulsivity was low at .06. Research indicates however that this construct is often reported with low levels of inter-rater reliability across teachers and parents, with parents tending to rate overactivity and impulsivity higher than teachers (Amador-Campos, Forns-Santacana, Guardia-Olmos & Pero-Cebollero, 2006; Charach, Chen, Hogg-Johnson & Schachar, 2009; Papageorgiou, Kalyva, Dafoulis & Vostanis, 2008), as identified in this study.

3.3.4 CBSQ distribution

3.3.4.1 Recruitment

Schools were contacted and invited to participate if they were situated in Birmingham and catered for children with a severe intellectual disability and/or they were a severe learning disability school situated in the West Midlands who had already collaborated in previous research with the university. A second criterion for school recruitment was that the school provided education for children aged between 2 and 12 years.

3.3.4.2 Participants

Six hundred and twenty nine children (316 < 8 years, 305 $\geq$ 8 years old) attending fourteen schools for children with a severe intellectual disability were surveyed. The return rate of the screens was estimated at 85%. All participants were between the ages of 2 and 12 years (mean age = 7.33) and 62.5% of the sample was male. Just over a third of the sample (34.3%) was described by their teachers as having a genetic syndrome whilst 45.5% of the sample was labelled as having 'probable ASD'. The majority of the sample also had some speech (62.5%), normal vision (68.9%), normal sight (87.9%) and were ambulant (72.2%). Participants aged less than 6 years had a

mean Denver developmental quotient of 46.63 (range = 0 to 113.85). The categorical data provided for participants aged 6 years and over showed that 30.4% (134) participants comprised the most severe ID group (Wessex score = 3 - 4.5), 38% (167) the moderately severe ID group (Wessex score = 5-7) and 31.6% (139) the least severe ID group (Wessex score = 7.5-9). These categories were arbitrarily defined.

3.3.5 Procedure

Letters and information sheets were sent to parents of all children between the ages of 2 and 12 years in participating schools. Screens were completed regarding each child whose parents had not opted out of the study three weeks after receipt of a letter and information sheet. Participating schools were sent a screen for every eligible child in the school and screens were then returned to the university upon completion. Ethical approval for this study was obtained from the School of Psychology ethical review committee at the University of Birmingham.

3.3.6 Data analysis

Relative risk analyses (with 99.9% confidence intervals), a series of Spearman's Rho correlations and Mann Whitney U analyses were conducted to measure the associations between each putative risk marker and the presence and severity of challenging behaviour. Relative risks were deemed significant if the lower confidence interval was greater than one. 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 challenging behaviour binary logistic regressions were also conducted.

3.4 Results

3.4.1 Risk marker and challenging behaviour prevalence rates

40.1% (252) of the total sample demonstrated one or more forms of challenging behaviour (20.7% (130) one form, 13.7% (86) two forms and 5.7% (36) three forms). Of these, aggression was the most common, demonstrated by 32.7% (204), whilst 17.2% (107) demonstrated self-injury and 15.8% (99) demonstrated destruction. 5.9% (37), 3.3% (20), 3.3% (21) and 8.1% (51) showed severe aggression, self-injury, destruction and one or more forms of severe challenging behaviour respectively. (See Table 3.1 for further prevalence figures).

Table 3.1: Prevalence rates (% and number of participants) of putative risk markers and challenging behaviours

Variable	Total % (n)	7 & under % (n)	8 & over % (n)
Male	62.5 (374)	60.5 (181)	64.2 (188)
'Probable ASD'	45.5 (216)	39.1 (95)	52.4 (119)
'High' severity of intellectual disability	50.5 (307)	43.5 (130)	57 (172)
One or more health problems	38.2 (232)	38.9 (119)	37.5 (110)
Repetitive behaviour	24.7 (145)	20.4 (59)	23.4 (67)
Restricted behaviour	22 (128)	20.4 (59)	23.4 (67)
Impulsive behaviour	57.3 (331)	55.6 (160)	58.9 (166)
Overactive behaviour	42 (231)	39.3 (106)	43.8 (119)
Aggression	32.7 (204)	28.5 (89)	37.2 (113)
Severe aggression	5.9 (37)	6.1 (19)	5.9 (18)
Destruction	15.8 (99)	16.2 (51)	15.5 (47)
Severe destruction	3.3 (21)	3.8 (12)	3 (9)
Self-injury	17.2 (107)	16.5 (52)	18.4 (55)
Severe self-injury	3.2 (20)	2.2 (7)	4.4 (13)
One or more forms of challenging behaviour	40.1 (252)	37.3 (118)	43 (131)
One or more forms of severe challenging behaviour	8.1 (51)	7.6 (24)	8.9 (27)

3.4.2 Overlap between variables

Prior to examining the relative risk of challenging behaviour given the putative risk markers, the relationship between the individual putative risk markers and forms of challenging behaviour was examined using relative risk analysis in order to identify potential variable overlap. As demonstrated in Table 3.2, this analysis showed that

many of the putative risk markers were significantly associated and thus it is likely that if these variables were entered into the analysis as they were, the results would reflect an interaction between putative risk markers rather than their independent influence.

Table 3.2: Relative risk analyses illustrating overlap between variables

Confounds	Relative Risk	Lower CI	Upper CI
Sex x autism	.49*	.37	.65
Autism x repetitive behaviour	3.82*	2.57	5.67
Aggression x destruction	8.63*	5.38	13.82
Aggression x self-injury	3.91*	2.71	5.64
Destruction x self-injury	3.88*	2.82	5.34
Repetitive x restricted behaviour	8.41*	5.92	11.94
Impulsivity x overactivity	6.26*	4.28	9.15

CI = 95%, * = $p < .001$

Two methods were utilised to resolve this issue. Firstly, to control for some of the overlap between four of the putative risk markers, two composites were formed for the analysis, so that repetitive and restricted behaviour formed the composite repetitive and restricted behaviours and interests whilst overactivity and impulsivity formed a composite of the same name, overactivity/impulsivity.

Secondly, the relative risk results indicated that it is likely that children demonstrating challenging behaviour will demonstrate more than one form. Thus, if a form of challenging behaviour was entered into the relative risk analysis, the results might not indicate the risk of demonstrating that one form alone. To assess the relative risk of demonstrating each of the forms of challenging behaviour independently, relative risk analyses were therefore conducted whereby one group demonstrating only one form

of challenging behaviour was compared to the remaining participants in the sample who did not demonstrate this form of challenging behaviour, but might demonstrate another form or no forms of challenging behaviour, as demonstrated in Table 3.3.

Table 3.3: Relative risk analyses for risk markers and independent challenging behaviours

Putative Risk Marker	Aggression RR (CI)	Destruction RR (CI)	Self-injury RR (CI)
Sex	.7 (.35, 1.39)	.71 (.09, 5.96)	1 (.28, 3.59)
Age	1.36 (.73, 2.52)	.52 (.06, 4.59)	.89 (.27, 2.94)
'Probable ASD'	1.44 (.7, 2.98)	4.22 (.36, 4.98)	.74 (.19, 2.9)
Severity of intellectual disability	1.8 (.94, 3.42)	.98 (.14, 6.83)	.42 (.11, 1.68)
Health	.76 (.39, 1.48)	.7 (.08, 5.82)	2.05 (.5, 6.96)
Repetitive and restricted behaviour and interests	1.34 (.71, 2.53)	4.65 (.41, 53.18)	2.09 (.52, 8.43)
Overactivity/ impulsivity	**2.87 (1.28, 6.42)***	-	1.3 (.33, 5.08)

CI = 99.9%, * = $p < .001$, - = incalculable due to an empty cell

These data indicate that whilst children with overactivity/impulsivity were at greater relative risk of showing aggression alone, none of the other risk markers were significantly associated with the other independent forms of challenging behaviour. However, the reduced group size in this analysis might have been responsible for these results. Thus, despite the risk of overlap between risk markers, relative risk analyses were conducted comparing groups demonstrating one form of challenging behaviour (but not necessarily only that form) with a group who did not.

3.4.3 Presence of challenging behaviour

3.4.3.1 Association between the broad variables and the presence of challenging behaviour

To test hypothesis 1a, the relative risk of challenging behaviour given the presence of 'probable ASD' as well as gender and severity of intellectual disability was examined across the total sample and two median split age groups (7 years and under and 8 years and over) (See Table 3.4).

Table 3.4: Relative risk of challenging behaviour given the broad variables for the total, 7 years and under and 8 years and over samples

Putative risk marker	Challenging behaviour	Total sample RR (CI)	7 & under RR (CI)	8 & over RR (CI)
Sex	**Aggression**	.74 (.5, 1.11)	.83 (.46, 1.52)	.68 (.4, 1.19)
	Destruction	.81 (.43, 1.51)	.75 (.31, 1.79)	.89 (.36, 2.2)
	Self-injury	.82 (.45, 1.48)	.86 (.37, 1.98)	.77 (.33, 1.8)
	One or more Forms	.78 (.55, 1.1)	.85 (.52, 1.39)	.72 (.45, 1.17)
'Probable ASD'	**Aggression**	**2.08* (1.34, 3.22)**	**2.22* (1.16, 4.23)**	**1.83* (1.01, 3.34)**
	Destruction	**3.04* (1.44, 6.41)**	**3.12* (1.18, 8.23)**	2.97 (.91, 9.71)
	Self-injury	**2.21* (1.16, 4.2)**	1.5 (.62, 3.61)	**3.53* (1.19, 10.41)**
	One or more forms	**1.95* (1.35, 2.81)**	**1.85* (1.12, 3.07)**	**1.97* (1.14, 3.4)**
Severity of intellectual disability	**Aggression**	1.19 (.83, 1.71)	1.35 (.76, 2.39)	1 (.63, 1.6)
	Destruction	.84 (.47, 1.51)	.96 (.41, 2.22)	.78 (.34, 1.78)
	Self-injury	.78 (.44, 1.39)	.81 (.33, 1.98)	.72 (.34, 1.54)
	One or more forms	1.06 (.78, 1.45)	1.1 (.69, 1.77)	1 (.66, 1.51)
Health	**Aggression**	1.04 (.72, 1.49)	.62 (.33, 1.17)	1.49 (.95, 2.35)
	Destruction	1.15 (.65, 2.05)	.79 (.34, 1.85)	1.75 (.77, 3.99)
	Self-injury	**1.81* (1.05, 3.13)**	1.45 (.66, 3.16)	2.27* (1.06, 4.87)
	One or more forms	1.13 (.83, 1.53)	.83 (.51, 1.34)	1.49 (1, 2.22)

CI = 99.9%, * = $p < .001$

The results of this analysis showed that the relative risk of presenting one or more forms of challenging behaviour and aggression in particular was increased by the presence of 'probable ASD' in all three samples, although the associations with self-injury and destruction were not significant in the 7 years and under and 8 years and over samples respectively. Differences in results across age groups also indicated the potential importance of age as a putative risk marker for challenging behaviour.

However, when entered into the relative risk analysis as a factor, age was not significantly associated with aggression (RR = 1.3, CI = .91, 1.87), destruction (RR = .95, CI = .54, 1.7), self-injury (RR = 1.11, CI = .65, 1.92) or one or more forms of challenging behaviour (RR = 1.15, CI = .85, 1.56), with no significant difference in the relative risk of these behaviours in participants aged 7 years and under and 8 years and over, as predicted.

Also supporting hypothesis 1a, the relative risk of challenging behaviour across samples was not significantly different given severity of intellectual disability, as predicted, or gender, whilst the presence of one or more health problems did increase the relative risk of self-injury almost two-fold in the total and 8 years and over sample. Mann Whitney analysis indicated that skin problems were the only health complaint to be associated with challenging behaviour in the total sample, with participants showing self-injury demonstrating significantly more severe skin problems (U = 21254, p <.001). However, this result might reflect the skin damage caused by self-injury rather than demonstrating the potential causal role of skin problems.

These results indicate that unlike gender, severity of intellectual disability and age, 'probable ASD' is a significant putative risk marker for challenging behaviour across age groups, supporting hypothesis 1a. Although the differences in the relative risks for self-injury and destruction across age groups indicate that age is not a risk marker in its own right, the significance of 'probable ASD' as a risk marker might change with age. Finally, the presence of one or more health problems was also associated with self-injury alone, supporting hypothesis 3.

3.4.3.2 Association between the behavioural variables and the presence of challenging behaviour

Hypothesis 2a was examined by investigating the relative risk of challenging behaviour given the presence of the putative behavioural risk markers using relative risk analysis (see Table 3.5).

Table 3.5: Relative risk of challenging behaviour given the putative behavioural risk markers for the total, 7 years and under and 8 years and over samples

Putative risk marker	Challenging Behaviour	Total sample RR (CI)	7 & under RR (CI)	8 & over RR (CI)
Repetitive and restricted behaviour and interests	**Aggression**	2.69* (1.77, 4.1)	2.65* (1.39, 5.04)	2.63* (1.5, 4.63)
	Destruction	4.8* (2.22, 10.38)	3.97* (1.51, 10.44)	6.2* (1.67, 23)
	Self-injury	4.87* (2.31, 10.3)	4.33* (1.59, 11.82)	5.42* (1.75, 16.82)
	One or more forms	2.71* (1.89, 3.9)	2.66* (1.58, 4.49)	2.67* (1.6, 4.46)
Overactivity/ impulsivity	**Aggression**	3.92* (2.29, 6.71)	5.55* (2.19, 14.03)	3.09* (1.61, 5.96)
	Destruction	7.61* (2.65, 21.81)	9.9* (2.06, 47.63)	5.95* (1.43, 24.74)
	Self-injury	3.51* (1.64, 7.54)	3.41* (1.22, 9.56)	3.81* (1.22, 11.88)
	One or more Forms	3.61* (2.29, 5.71)	4.34* (2.14, 8.78)	3.09* (1.7, 5.63)

CI = 99.9%, * = $p < .001$

The results demonstrate that the putative risk markers showed a much stronger association with each form of challenging behaviour than the broad variables as participants with repetitive and restricted behaviours and interests and overactivity/impulsivity were at a significantly greater relative risk of each form of challenging behaviour across all three samples. The relative risk of destruction was almost ten times greater given the presence of overactivity/impulsivity, indicating the

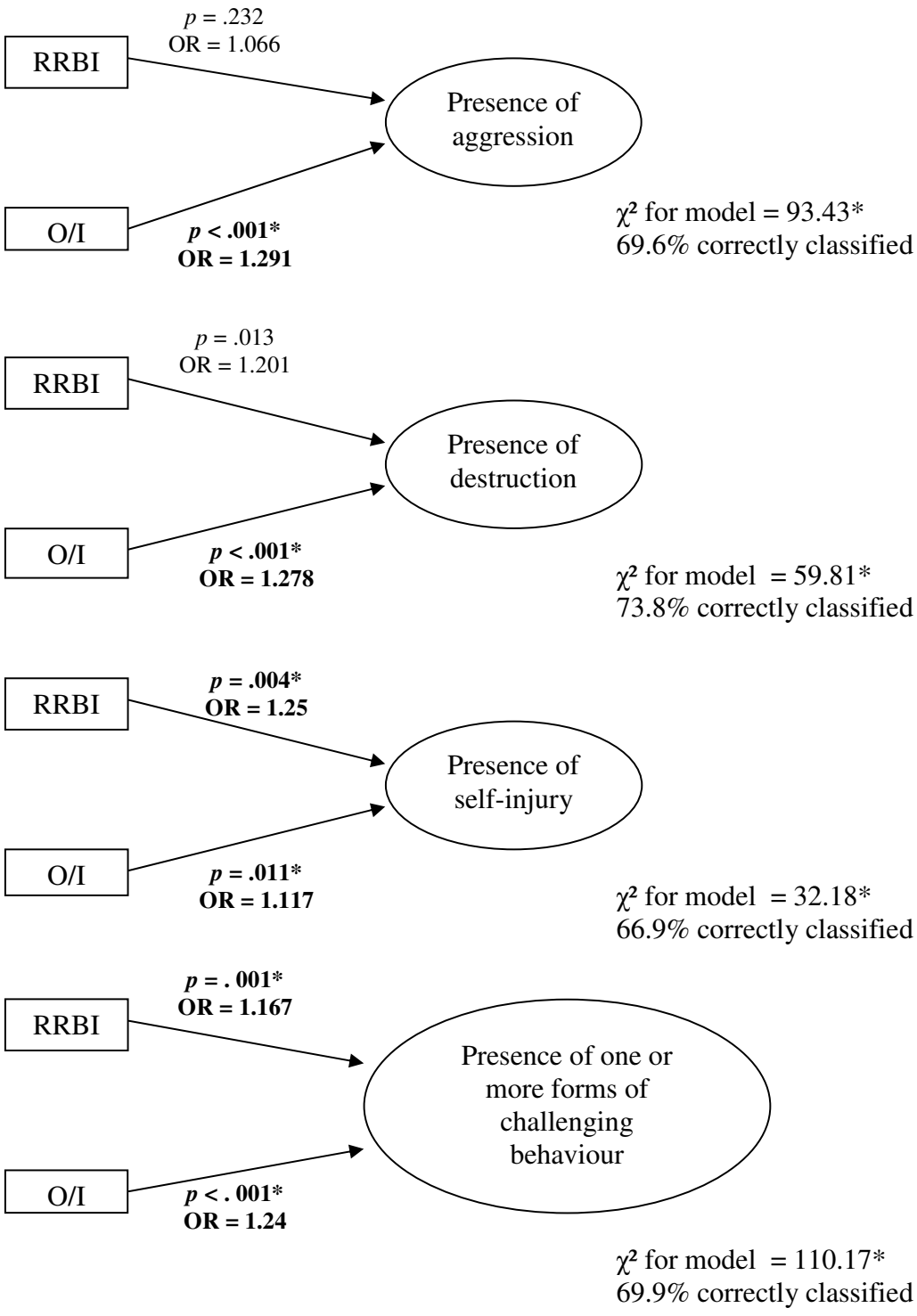
importance of this variable as a putative risk marker for challenging behaviour. The relative risk of self-injury given the presence of repetitive and restricted behaviours and interests was also high across age groups. These results indicate the importance of both repetitive and restricted behaviours and interests and overactivity/impulsivity for the presence of challenging behaviour. The similarity in results across age groups indicates the robust nature of the risk markers across age and provides support for hypothesis 2a.

3.4.3.3 Predictive models of the presence of challenging behaviour

In order to control for the overlap between variables, to produce predictive models of challenging behaviour and to test hypothesis 3a, a series of binary logistic regressions was conducted. If the relative risk of a form of challenging behaviour was significantly greater in participants with a broad or behavioural characteristic, this was entered into the regression analysis as a predictor variable. However, 'probable ASD' was not entered into this analysis due to its overlap with repetitive behaviour. Health was also excluded from the analysis as its influence on challenging behaviour was felt to be of a very different nature to the remaining variables which might have affected the significance of the other individual predictors in the model. Due to the similarities in relative risk given the behavioural putative risk markers across age groups, predictive models were based on the whole sample.

The results of these analyses, as demonstrated by Figure 3.1, show that each of the models significantly predicted each form of challenging behaviour, supporting hypothesis 2a. However, different forms of challenging behaviour were predicted by slightly different risk markers, supporting hypothesis 3. For example, whilst one or

more forms of challenging behaviour were predicted by both overactivity/impulsivity and repetitive and restricted behaviours and interests, none of the specific forms of challenging behaviour were significantly predicted by both composite risk markers.



***p* <. 0125**

Figure 3.1: Binary logistic regressions predicting the presence of challenging behaviour

These results indicated that aggression, destruction and self injury were significantly predicted by overactivity/impulsivity whilst self-injury was also predicted by

repetitive and restricted behaviours and interests. However, it must be noted that repetitive and restricted behaviours and interests was on the border of significance for destruction. Additionally, whilst not acting as a significant predictor of destruction, RRBI clearly contributed to the model for destruction, with an odds ratio of 1.201.

3.4.4 Severity of challenging behaviour

3.4.4.1 Association between the broad variables and severe challenging behaviour

In order to test hypothesis 1b, the relative risk of severe challenging behaviour given the presence of 'probable ASD' as well as gender and severity of intellectual disability was examined across the total sample and two age groups (see Table 3.6).

Table 3.6: Relative risk of severe challenging behaviour given broad variables for the total, 7 years and under and 8 years and over samples

Risk Marker	Severe challenging behaviour	Total sample RR (CI)	7 & under RR (CI)	8 & over RR (CI)
Sex	**Aggression**	.90 (.3, 2.65)	1.2 (.29, 4.98)	.59 (.1, 3.41)
	Destruction	.84 (.2, 3.43)	.77 (.12, 4.98)	.89 (.1, 7.72)
	Self-injury	.76 (.17, 3.43)	.61 (.05, 7.91)	.88 (.14, 5.67)
	One or more forms	.88 (.36, 2.16)	.99 (.28, 3.52)	.79 (.22, 2.8)
Autism	**Aggression**	**5.24* (1.16, 23.69)**	4.66 (.61, 35.32)	5.84 (.58, 59.36)
	Destruction	6.02 (.56, 65.35)	3.12 (.22, 44.26)	-
	Self-injury	2.64 (.51, 13.73)	1.56 (.07, 33.78)	2.72 (.36, 20.58)
	One or more forms	**3.48* (1.09, 11.15)**	2.8 (.52, 15.05)	3.89 (.73, 20.72)
Severity ID	**Aggression**	.87 (.32, 2.37)	1.29 (.31, 5.3)	.6 (.14, 2.5)
	Destruction	.65 (.16, 2.61)	.48 (.06, 3.83)	.94 (.12, 7.27)
	Self-injury	.35 (.07, 1.74)	.26 (.01, 7.62)	.34 (.05, 2.1)
	One or more forms	.64 (.27, 1.55)	.52 (.12, 2.22)	.71 (.23, 2.2)
Health Problems	**Aggression**	2.35 (.86, 6.41)	1.12 (.28, 4.51)	**5.82* (1.05, 32.43)**
	Destruction	1.48 (.39, 5.6)	.79 (.12, 5.11)	3.36 (.39, 29.11)
	Self-injury	**4.89* (1.01, 23.72)**	3.94 (.3, 51.38)	5.63 (.76, 41.8)
	One or more forms	1.96 (.85, 4.51)	1.12 (.33, 3.84)	3.31 (.99, 11.09)

*** *p* <.001, CI = 99.9%**

As demonstrated by Table 3.6, analogous with the presence of challenging behaviour, being male or having a more severe level of intellectual disability did not increase the risk of severe challenging behaviour across samples. However, having one or more health problems did increase the relative risk of severe self-injury in the whole sample

and aggression in children aged 8 years and over. More specifically, skin problems were also significantly more severe in participants with severe self-injury (U = 3820, p <.001) and unlike the presence of self-injury, digestive (U = 3794, p <.001) and any other health problems (U = 3077, p < .001) were also significantly more severe in participants with severe self-injury. Participants with one or more forms of severe challenging behaviour also had significantly more severe skin problems (U = 11279, p <.001). The presence of 'probable ASD' also increased the relative risk of both severe aggression and one or more severe forms of challenging behaviour, but not severe destruction or severe self-injury.

On the whole, the trends were similar across samples, indicating that age was not associated with severity of challenging behaviour. Supporting this, the relative risk of severe aggression (RR = .97, CI = .36, 2.61), destruction (RR = .78, CI = .2, 2.98), self-injury (RR = 1.96, CI = .47, 8.22) and one or more forms of severe challenging behaviour (RR = 1.17, CI = .51, 2.69) was not significantly different in participants aged 7 years and under or 8 years and over.

Therefore, supporting hypothesis 1b, 'probable ASD' did significantly increase the risk of severe challenging behaviour but only aggression specifically, whilst severity of intellectual disability and age did not predict severity of challenging behaviour. Gender also showed no significant association with challenging behaviour. Supporting hypothesis 3b, severe self-injury was the only form of challenging behaviour to be significantly associated with health problems in the total sample.

3.4.4.2 Association between the behavioural variables and severe challenging behaviour

Hypothesis 2b was examined by investigating the relative risk of challenging behaviour given the presence of the behavioural risk markers. The risk of severe challenging behaviour given the presence of repetitive and restricted behaviours and interests and overactivity/impulsivity varied considerably depending on the form of challenging behaviour under examination. The results of these analyses are shown in Table 3.7.

Table 3.7: Relative risk of severe challenging behaviour given the putative behavioural risk markers in the total, 7 years and under and 8 years and over samples

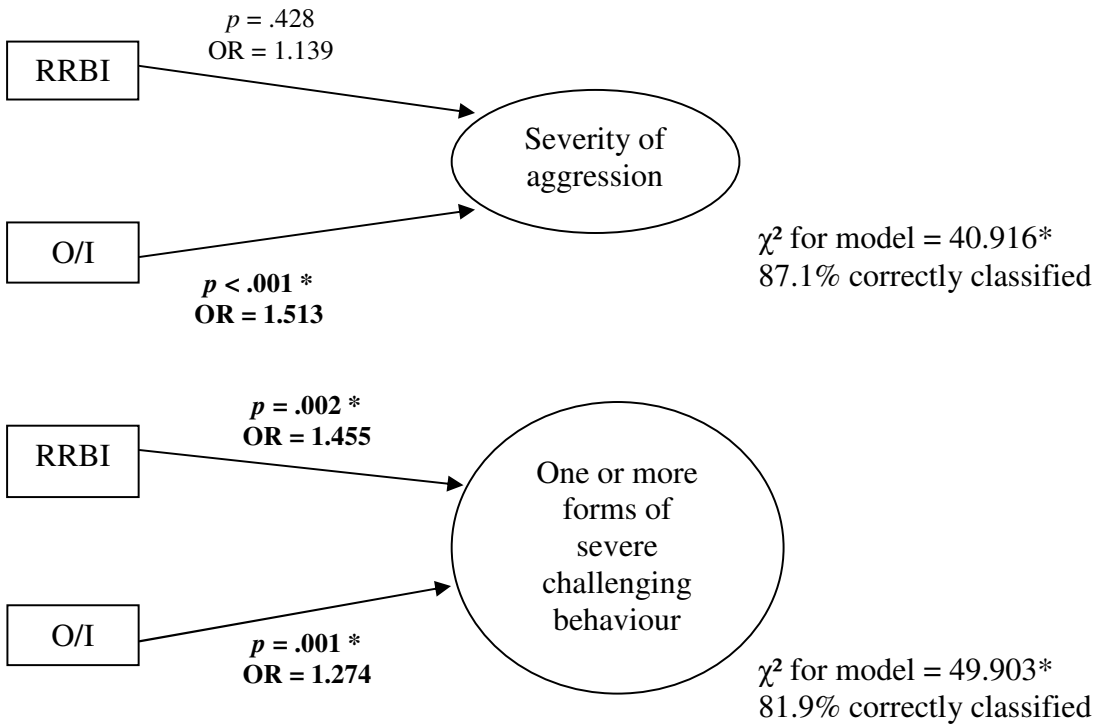
Putative risk markers	Severe challenging behaviour	Total sample RR (CI)	7 & under RR (CI)	8 & over RR (CI)
Repetitive and restricted behaviours and interests	**Aggression**	**11.51* (1.79, 73.86)**	9.28 (.92, 93.74)	15.66 (.66, 374.45)
	Destruction	19.83 (.83, 476.57)	12.25 (.48, 314.78)	-
	Self-injury	4.09 (.72, 23.27)	2.71 (.19, 38.62)	4.93 (.46, 52.83)
	One or more forms	**11.67* (2.34, 58.06)**	**12.83* (1.32, 124.64)**	**10.35* (1.08, 99.62)**
Overactivity/impulsivity	**Aggression**	**6.39* (1.26, 32.36)**	15.25 (.65, 360.18)	3.65 (.53, 25.31)
	Destruction	14.5 (.61, 346.39)	9.47 (.38, 236.01)	-
	Self-injury	3.7 (.53, 26.54)	4.4 (.15, 128.57)	3.45 (.32, 37.75)
	One or more forms	**4.87* (1.41, 16.79)**	6.08 (.93, 39.64)	4.14 (.8, 21.54)

*** = $p < .001$, CI = 99.9%**

As illustrated by Table 3.7, repetitive and restricted behaviours and interests significantly increased the relative risk of severe aggression and one or more severe forms of challenging behaviour by over eleven times. Overactivity/impulsivity also increased the relative risk of these same two forms of severe challenging behaviour. However, neither behavioural putative risk marker significantly increased the relative risk of severe destruction or severe self-injurious behaviour, so that predictive models of challenging behaviour could only be examined for these forms. Thus, hypothesis 2b was only partially supported.

3.4.4.3 Predictive models of severe challenging behaviour

As with the presence of challenging behaviour, binary logistic regressions were conducted in order to control for the overlap between variables, to produce predictive models of challenging behaviour and to test hypothesis 3b. Binary logistic regressions were only conducted if the previous relative risk analyses had shown significant associations between the behaviour and the putative risk marker. This analysis was based on the total sample due to the similarities in the relative risk analysis across groups. The results of these analyses are shown in Figure 3.2.



*** = *p* < .025**

Figure 3.2: Binary logistic regressions predicting the presence of severe challenging behaviour

The results of the binary logistic regressions demonstrated that models with a significant fit were produced for both severe aggression and one or more forms of severe challenging behaviour. Both putative behavioural risk markers significantly predicted the severity of one or more forms of challenging behaviour, although only overactivity/impulsivity significantly predicted the severity of aggression. Thus, hypothesis 3b was not met for the severity of challenging behaviour as although different forms of challenging behaviour were predicted using different models, not all forms could be significantly predicted.

3.5 Discussion

The primary aim of this study was to examine those child characteristics acting as putative risk markers for challenging behaviour in children with a severe intellectual disability. By employing a large, representative sample and a reliable brief measure of both challenging behaviour and the putative risk markers, the results of this study can be considered accurate and easily generalised to the population of children with severe intellectual disabilities as a whole.

The results of this study indicate that particular child characteristics are predictive of challenging behaviour at one point in time, implying that we are one step closer to identifying children with severe intellectual disability at high risk of challenging behaviour. More specifically, the relative risk analyses demonstrated that participants with 'probable ASD' were at significantly greater risk of all forms of challenging behaviour, replicating previous research (Baghdadli *et al.,* 2003; Holden & Gitlesen, 2006; McClintock *et al.,* 2003). Children with one or more health problems were also at a significantly greater risk of showing self-injury, although further investigation showed that skin problems were the only health condition to be significantly associated with this behaviour. The putative behavioural risk markers repetitive and restricted behaviours and interests and overactivity/impulsivity were also significantly associated with all forms of challenging behaviour so that children showing these behaviours were all at significantly greater relative risk of showing challenging behaviour.

The largest increase in risk for a behaviour given the presence of a putative behavioural marker was destruction, the relative risk of which was almost eight times

greater given the presence of overactivity/impulsivity, indicating the importance of this putative risk marker. The predictive models produced for each form of challenging behaviour were also significant with both repetitive and restricted behaviours and interests and overactivity/impulsivity significantly predicting one or more forms of challenging behaviour and having various associations with the specific forms of challenging behaviour. These findings provide further evidence for the preliminary findings of Oliver *et al.* (in preparation) and Petty *et al.* (in preparation) who originally indicated the role of repetitive and restricted behaviours and interests and overactivity/impulsivity as risk markers for challenging behaviour.

The results of the analysis regarding severe challenging behaviour were similar to that of presence so that children with 'probable ASD' were at significantly greater risk of one or more forms of severe challenging behaviour, but only aggression specifically. Replicating the findings of Petty *et al*. (in preparation) children with one or more health problems were also at significantly greater risk of severe self-injury. Further analysis showed that skin, digestive problems and any other health problems were all significantly associated with severe self-injury. Unlike the presence of challenging behaviour, the putative behavioural risk markers only increased the risk of severe aggression and one or more forms of severe challenging behaviour. However, these relative risks were very high, with the relative risk of severe aggression increased by over eleven times given the presence of repetitive and restricted behaviours and interests. Although, once entered into the binary logistic regression, only overactivity/impulsivity was able to significantly predict both severe aggression and one or more forms of challenging behaviour, with repetitive and restricted behaviours

and interests only significantly predicting one or more forms of severe challenging behaviour.

The sometimes discrepant results across age groups also indicated that age might act as a risk marker for the presence and severity of challenging behaviour. However, entering age into the relative risk analyses did not produce any significant results, indicating that rather than acting as a risk marker, age might affect the significance of the putative behavioural risk markers. In accordance with the work of Oliver *et al.* (in preparation), severity of intellectual disability also failed to show a significant association with challenging behaviour in a sample of children with severe intellectual disabilities which might have been the result of the severe level of intellectual disability of the sample or the way in which severity of intellectual disability was measured. The findings of this study also suggest that gender is not a risk marker for challenging behaviour, in support of some (Baghdadli *et al.,* 2003; Chadwick *et al.,* 2000; Holden & Gitlesen, 2006) but not all (McClintock *et al.,* 2003) previous research.

The increased relative risk of the presence and severity of self-injury given the presence of one or more health problems indicated the significance of health and, by implication, pain as a putative risk marker for self-injury, supporting previous research (Carr & Owen-DeSchryver, 2007; Ghaziuddin, Elkins, McNeeley & Ghaziuddin, 1993; Luzzani *et al.,* 2003; Peine *et al.,* 1995).

The association between health, pain and self-injury can be explained with reference to three mechanisms. Firstly, self-injury might function to directly remove the painful

or uncomfortable stimulus, so that, for example, an individual with an irritating skin condition might scratch the site of pain in order to relieve an itching sensation. According to the gate control theory of pain (Melzack & Wall, 1965), individuals might also engage in self-injurious behaviour in order to relieve the pain experienced at another body site. Finally, the presence of a painful health condition might increase the perceived aversiveness of a task, leading to challenging behaviour. As is the case when working with many individuals with an intellectual disability, a demand imposed by an adult might result in some form of challenging behaviour, in order to escape the demand (Iwata *et al.,* 1994). However, if a child has health problems, the likelihood of this behaviour is even greater as the individual perceives the demand as more aversive and thus is more likely to react in a challenging manner to this demand. This supposition is supported by previous evidence identifying an interaction between menstrual pain and increased demand aversiveness associated with challenging behaviour (Carr, Smith, Giacin, Whelan & Pancari, 2003).

Thus, these three mechanisms explain how health can lead to the introduction of challenging behaviour into the behavioural repertoire. To understand the maintenance of this behaviour, consideration should also be given to operant theory. For example, when an individual engages in self-injurious behaviour in order to remove a painful stimulus, they experience pain relief and negative reinforcement which increases the likelihood of them repeating the behaviour in the future. This behaviour is also likely to elicit adult concern and attention which in turn also positively reinforces the behaviour (Oliver, 1993, 1995), so that eventually, children engage in self-injurious behaviour in order to gain attention as well as relieve pain and through mutual reinforcement the adult attends to the child so that the they will stop self-injuring,

which the adult finds reinforcing. Similarly, if an individual is experiencing pain, this setting event functions as an establishing operation, increasing the aversiveness of the task. The resulting challenging behaviour is aversive to the individual's carer, who consequently removes the task to reduce the behaviour, which is negatively reinforcing for both the individual displaying the behaviour and the carer (Carr *et al.,* 2003).

Thus these results are important as they illustrate a way in which a behaviour can enter into an individuals behavioural repertoire and develop into a more serious challenging behaviour. Given the already established high prevalence of health problems in individuals with an intellectual disability (Berg *et al*., 2007), the identification and treatment of health problems in children with severe intellectual disabilities should now be considered a priority in the attempt to prevent self-injury in this population.

The association between repetitive and challenging behaviour could also be explained in a similar way to health, in that it introduces the behaviour into the behavioural repertoire. Research has shown that the demonstration of repetitive behaviour is common in children with intellectual disabilities (Chadwick *et al*., 2004; Thompson & Reid, 2002) and that if subjected to social reinforcement, this behaviour can evolve to become self-injurious. This is in accordance with Guess and Carr's model (1991) whereby self-injury develops from repetitive behaviour via operant processes.

The association between impulsive, overactive and repetitive behaviour and the presence and severity of challenging behaviour also implies a potential role for

compromised behavioural inhibition in challenging behaviour in this population. This supposition is supported by contemporary neuropsychological models of ADHD and autism proposed by Barkley (1997a, 1997b) and Turner (1997, 1999) respectively which have indicated an association between repetitive behaviour, impulsivity, hyperactivity and poor inhibition. This could also complement operant models (e.g. Oliver, 1995; Oliver, Hall & Murphy, 2005) of challenging behaviour in which challenging behaviour is evoked under stimulus conditions and reinforced so that the challenging behaviour becomes a learned response. Children with compromised behavioural inhibition might find it even harder to inhibit this learned response and thus might show a greater prevalence of challenging behaviour, such as aggression. Therefore, repetitive behaviour might act as a risk marker in a number of ways; introducing a behaviour into the repertoire which can be shaped by operant processes into self-injury and by indicating an underlying inhibition deficit which makes it harder for children to inhibit a learned response, like aggression.

Supporting the influence of biological factors on challenging behaviour, recent studies have provided evidence for the effect of the monoamine oxidase A (MAOA) promoter gene on the presence of challenging behaviour. Illustrating this, May *et al.* (2009) reported a significantly higher prevalence of the short allele MAOA polymorphism in individuals with intellectual disabilities and challenging behaviour than individuals with or without intellectual disabilities and no challenging behaviour. Associations between polymorphisms in the MAOA gene and aggression and problem behaviour have already been established in the general population (Caspi *et al.,* 2002; Brunner, Nelen, Breakefield, Ropers & Oost, 1993), thus whilst some studies have failed to identify this association (Kim-Cohen *et al.,* 2006, Taylor & Kim-Cohen, 2007),

evidence is accumulating in support of biological vulnerabilities to challenging behaviour which, if confirmed, could prove vital to early intervention strategies. The influence of such biological factors on challenging behaviour thus requires further examination.

Whilst these results are promising in the context of early intervention and the identification of children in need of it, several limitations of this research must be highlighted. Firstly, the risk markers identified in this study only serve to predict challenging behaviour at one point in time. Considering the dramatically increased risk of challenging behaviour given the presence of particular putative behavioural risk markers, it is likely that these risk markers will continue to predict challenging behaviour over time, although longitudinal research will be required to test this hypothesis.

Secondly, whilst reflecting the cautious nature with which the data was interpreted, the use of Bonferroni corrections for the binary logistic regressions does affect the risk markers found to significantly predict the presence of challenging behaviour. For example, only overactivity/impulsivity was found to significantly predict the presence of destruction, although the likelihood of destruction given RRBI and overactivity/impulsivity was very similar (odds ratios were 1.201 and 1.278 respectively). Indeed, the conventional 0.05 critical region acting as the baseline for the Bonferroni correction is merely an arbitrary convention. Hence, it would be reasonable to assume that there is little difference in the predictive power between two risk markers with similar odds ratios. Therefore, whilst the use of Bonferroni correction in this case represents an attempt at conservative interpretation and an

avoidance of Type 1 error, had such a strict approach not been utilised, both risk markers would have been found to significantly predict destruction and thus altered the model produced.

Due to the overlap in forms of challenging behaviour demonstrated by the participants, it was also difficult to disentangle the risk markers predictive of only one type of challenging behaviour. Yet, the ability of the putative behavioural risk markers to predict only severe aggression and one or more forms of severe challenging behaviours and not severe destruction or severe self-injury also indicates the importance of different predictive models for individual forms of severe challenging behaviour. These results might indicate the different developmental pathways for different forms of challenging behaviour, as already proposed by Guess and Carr (1991) who developed a self-injury specific stage model illustrating the unique development of self-injury from repetitive behaviour. Further research however is required to broaden our understanding of the development of other specific forms of challenging behaviour.

Thus a number of avenues for future research have arisen as a result of this study, although arguably the most important conclusion to be drawn from the findings is that the identification of children at high risk of challenging behaviour based on the presence of particular putative risk markers at one point in time is possible. Also of great importance is the fact that these results, which broadly replicate previous findings, were obtained using a short and accessible screen and whilst they will require replication within a longitudinal study, it appears that early intervention is a strategy worth considering in light of the results of this study and previous research.

3.6. Summary and implications

In Chapter 3, a cohort questionnaire study was conducted in order to investigate associations between putative risk markers and challenging behaviour in a population of children with severe intellectual disabilities using the CBSQ. Replicating the findings of previous studies, the results of this chapter illustrated the increased risk of all forms of challenging behaviour associated with the presence of autism as well as two behavioural risk markers, repetitive and restricted behaviours and interests (a composite of repetitive and restricted behaviour) and overactivity/impulsivity (a composite of overactive and impulsive behaviour). In terms of severe challenging behaviour, the relative risk of aggression and one or more forms of challenging behaviour was significantly increased by repetitive and restricted behaviours and interests and overactivity/impulsivity and if the child had been labelled as having 'probable ASD'. A significant relationship between health and self-injurious behaviour was also identified. Predictive models derived from the results of binary logistic regressions highlighted differential relationships between challenging behaviour and the putative risk markers depending on the exact form examined, although such conclusions must be considered in light of the highly conservative statistical methods used.

Replicating the results of previous studies using a condensed and accessible measure represents a significant progression towards the implementation of early intervention strategies, indicating the possibility of identifying children at highest risk of developing severe challenging behaviour and thus those most in need of early intervention at one point in time. These results also illustrate the possible role of compromised behavioural inhibition in challenging behaviour in this population.

The study described in Chapter 4 builds upon these results by examining the association between the same putative risk markers and challenging behaviour investigated in Chapter 3 in an 18 month follow up study in order to determine the ability of these risk markers to predict challenging behaviour over time.

Chapter 4

Risk Markers for the Remission, Incidence and Persistence of Challenging Behaviour in Young Children with Severe Intellectual Disabilities

4.1 Abstract

Background and aims: The combination of increasing prevalence with age, persistence and associated negative consequences of challenging behaviour in people with severe intellectual disabilities, as well as the paucity of effective interventions, indicate the need for targeted early intervention. The aim of this study was to examine the capacity of four putative risk markers to predict the presence of challenging behaviour 18 months later, and particularly, the remission, incidence and persistence of challenging behaviour.

Method: A follow up study was conducted with 417 participants with severe intellectual disabilities recruited as part of the screen study conducted 18 months previously (Chapter 3). The methodology employed also replicated that used in the screen study.

Results: Replicating the results of previous studies, the persistence of challenging behaviour was high (28.5% for one or more forms). The composite behavioural risk markers, restricted and repetitive behaviours and interests (comprising repetitive and obsessive behaviour) and overactivity/impulsivity (comprising overactivity and impulsivity), measured at the screen significantly predicted the presence of challenging behaviour over this 18 month period, although both risk markers were more successful in predicting the persistence than either the remission or incidence of challenging behaviour. Concern expressed by teachers at the screen was also

significantly higher for participants who persisted to show self-injury at the follow up. However, health did not significantly predict self-injury as it did at the screen.

Discussion: These results indicate the ability of the behavioural risk markers to predict the persistence of challenging behaviour over time, indicating their potential utility as indicators of risk for challenging behaviour in children with severe intellectual disabilities.

4.2 Introduction

Prevalence rates of challenging behaviour in people with intellectual disabilities are within the range of 5 to17% (Borthwick-Duffy, 1994; Emerson & Bromley, 1995; Emerson *et al.,* 2001a; Lowe *et al.,* 2007; Qureshi & Alborz, 1992). The negative consequences of challenging behaviour for people with an intellectual disability are well documented and include physical harm to themselves and others (Konarski, Sutton & Humman, 1997; Nissen & Haveman, 1997) and exclusion from an array of services (Kiernan & Qureshi, 1993). Additionally, challenging behaviour is a major source of distress for families, particularly if associated with the need for residential care outside of the family home (e.g. McIntyre, Blacher & Baker, 2002), and is expensive for those providing services to people with intellectual disabilities (Knapp, Comas-Herrera, Astin, Beecham & Pendaries, 2005). According to previous research, the prevalence of challenging behaviour increases with age up until early adulthood (e.g. Eyman & Call, 1977; Harris, 1993; Jacobson, 1982; Oliver, Murphy & Corbett, 1987; Sigafoos, Elkins, Kerr & Attwood, 1994), although self-injurious behaviour might be more prone to changes in prevalence with age than aggression (section 2.5). Challenging behaviour also appears to be highly persistent once established (Kebbon & Windahl, 1986; Murphy *et al*., 1993; Schroeder, Schroeder, Smith & Daldorf,

1978), with reported rates varying between 57 and 90% (Chadwick *et al*., 2004; Emerson, Robertson, Fowler, Letchford & Jones, 1996; Emerson *et al.,* 2001b; Kiernan & Alborz, 1996; Nottestad & Linaker, 2002).

Despite the common use of major neuroleptics (Kalachnik, 1999) a systematic review concluded that there is no robust evidence that antipsychotic medication decreases challenging behaviour (Brylewski & Duggan, 1999) and their use is associated with detrimental side effects (Baumeister, Todd & Sevin, 1998) and is not cost effective (Romeo, Knapp, Tyrer, Crawford & Oliver-Africano, 2009). Indeed, some medications have been found to be less effective than placebos for the amelioration of aggression (Tyrer *et al.,* 2008). More recent reviews have cited some evidence for the use of several medications, such as Risperidone and Lithium, although methodological flaws in these studies invite cautious interpretation (Deb *et al*., 2008; Deb & Unwin, 2007). Conversely, behavioural interventions, whilst resource intensive (Robertson *et al*., 2005), have been found in meta-analytic studies to successfully reduce the frequency of challenging behaviour (e.g. Harvey, Boer, Meyer & Evans, 2009) and have broad support in the empirical literature (e.g. Kahng, Iwata & Lewin, 2002).

Because of the high prevalence and persistence of challenging behaviour and the paucity of effective and economically viable interventions, attention has now turned to the appropriateness of early intervention strategies (Richman, 2008; Richman & Lindauer, 2005; Symons, Sperry, Dropik & Bodfish, 2005). Having already achieved successful results in other areas, including health (Blanks, Moss, McGahan, Quinn & Babb, 2000) and education (NESS, 2005), this strategy appears to be particularly

appropriate and thus has the potential to significantly reduce challenging behaviour in people with intellectual disabilities. A major difficulty facing services for challenging behaviour is that by the age of referral, the behaviour is more forceful and thus more dangerous. By intervening early, behavioural interventions would be less difficult to implement as the challenging behaviour would be easier to manage. Empirical evidence now exists to support established models of the development of self-injury (Guess & Carr, 1991; Oliver 1995), illustrating a direct relationship between the increased frequency of early self-injury and social reinforcement (Oliver, Hall & Murphy, 2005). Thus, it might be presumed that when intervening early, challenging behaviour might be less resistant to behavioural intervention as the behaviour is less well established in the repertoire.

It therefore appears that early intervention strategies are appropriate and have the potential to reduce the future prevalence of challenging behaviour in people with intellectual disabilities. However, in order to implement this strategy efficiently, services would need to know which children are at the highest risk of challenging behaviour. Previous research indicates several child characteristics which are associated with and precede the presence of challenging behaviour and might therefore predict its future presence. Chadwick, Piroth, Walker, Bernard and Taylor (2000) identified severity of intellectual disability as a risk marker via an association between destructive and self-injurious behaviour and lower level of intellectual functioning. This finding was replicated in three further studies, which also identified the importance of autism, as well as presence of a perinatal condition, lower chronological age (Baghdadli, Pascal, Grisis & Aussilloux, 2003) and communicative ability (Holden & Gitlesen, 2006; McClintock, Hall & Oliver, 2003). More recently,

two longitudinal studies have identified significant associations between self-injury and communicative ability (Danquah *et al*., 2009) and autism (Baghdadli *et al*., 2008). Indeed, the association between challenging behaviour and severity of intellectual disability is well established in the literature (e.g. Griffin, Williams, Stark, Altmeyer & Mason, 1986; Maisto *et al.,* 1978, Schroeder *et al.,* 1978). Whilst investigating the early development of self-injurious behaviour, Murphy, Hall, Oliver & Kissi-Debra (1999) also discovered an association between an increase in self-injurious behaviour and teachers' concern regarding this behaviour 18 months previously, illustrating that variables external to individuals might also be important.

With severity of intellectual disability already established as a robust risk marker, more recent studies have focused on behavioural characteristics associated with challenging behaviour in children already at high risk within this already higher risk group. Illustrating this, Oliver *et al*. (in preparation) reported associations between repetitive, ritualistic and challenging behaviours, a finding replicated in children with the rare genetic syndrome Cornelia de Lange Syndrome (Oliver, Sloneem, Hall & Arron, 2009) and supported by a review (Petty & Oliver, 2005) and recent study (Arron, Oliver, Berg, Moss & Burbidge, in review). This is particularly significant due to the high reported prevalence rates of repetitive behaviours in people with intellectual disabilities (Chadwick *et al*., 2004; Jones, 1999; Thompson & Reid, 2002). A more recent study also found that overactive and impulsive behaviours significantly predicted both the presence and severity of challenging behaviour (Petty *et al*., in preparation). By replicating the analysis employed by Oliver *et al.*, a major strength of both this and Oliver *et al.'s* study was the use of binary logistic regression utilised to control for the potential overlap between variables. Substantiating the

importance of impulsivity and overactivity, two recent studies have also identified associations between self-injurious and aggressive behaviour with attention deficit hyperactivity disorder (Cooper, Smiley, Allan *et al*., 2009; Cooper, Smiley, Jackson *et al*., 2009).

Whilst these studies utilised a range of samples and methodologies, they all involved the completion of long questionnaires that might not be appropriate when aiming to screen large populations of children, as would be required for an early intervention strategy, due to the burden placed upon those completing the questionnaires. The screen study described in Chapter 3 addressed this issue through the development of a short and accessible screening tool, designed for completion by teachers. The results of this study replicated previous findings (e.g. Oliver *et al.,* in preparation; Petty *et al*., in preparation) whilst developing predictive models for both the presence and severity of challenging behaviour. Replicating the statistical analysis employed by Oliver *et al*. (in preparation) and Petty *et al*. (in preparation), the potential confound between variables was also controlled for using a series of binary logistic regressions. This analysis demonstrated that repetitive and restricted behaviours and interests (a composite of both repetitive and restricted behaviours) predicted the presence of self-injury, whilst aggression and destruction were predicted by overactivity/impulsivity (a composite of overactive and impulsive behaviours), illustrating differences in the putative risk markers associated with different forms of challenging behaviour.

The presence of these putative behavioural risk markers in children with a severe intellectual disability also, arguably, implies a role of compromised behavioural inhibition in challenging behaviour in this population. This builds on the work of

Barkley (1997a, 1997b) and Turner (1997, 1999) who identified an association between repetitive behaviour in autism and impulsivity and hyperactivity in ADHD with poor inhibition (section 3.5). The relative risk of self-injury was also significantly increased by the presence of health problems during the previous month, although this putative risk marker was not included in the predictive model due to its more transient nature.

However, similar to previous studies investigating putative risk markers (e.g. Chadwick *et al*., 2000; Baghdadli *et al*., 2003; but with the exception of Baghdadli *et al.,* 2008; Danquah *et al*., 2009) the study described in Chapter 3 only examined the child characteristics associated with challenging behaviour at one point in time. Whilst the results of such studies provide insight into the child characteristics associated with challenging behaviour, they do not evaluate whether these associations will remain over time, information important to the successful implementation of early intervention programmes. Longitudinal evidence for the putative risk markers identified in Chapter 3 is essential for laying the successful foundations of early intervention programmes.

The aim of this study is to investigate the persistence of the putative risk markers (repetitive and restricted behaviours and interests, overactivity/impulsivity and health) identified in Chapter 3 (section 3.4.3.2) and to examine whether they predict the future presence of challenging behaviour. In order to test this, a follow up study will be conducted, employing the sample recruited for the original screening study (section 3.3.3.2) and utilising the same screening measure, the Challenging Behaviour Screening Questionnaire (CBSQ). As described in Chapter 3 (section 3.2), an

administrative definition of severe intellectual disability was used in this study, so that participants were assumed to have a severe intellectual disability due to their attendance at schools for children with severe intellectual disability. Whilst an administrative definition of severity of intellectual disability might lead to the inclusion of some more able participants, it was deemed sufficient and necessary considering the large population required to examine the putative risk markers and test their suitability for indicators of high risk in an early intervention context. Thus, the sample employed in both studies (Chapters 3 and 4) are assumed to comprise of children with a severe intellectual disability, although it is accepted that there might be some variation in severity of intellectual disability within the sample.

The primary focus of this paper in terms of putative risk markers of challenging behaviour will be on the behavioural markers, restricted and repetitive behaviours and interests and overactivity/impulsivity. Although the role of health will be examined, less emphasis is placed on this due to the more limited associations between health and only one form of challenging behaviour, self-injury, as illustrated in Chapter 3 (section 3.4.3.1) and potential bidirectional effects. Additionally, whilst severity of intellectual disability has been reported to be a robust risk marker for challenging behaviour in children with intellectual disabilities, recent studies conducted by Oliver *et al*. (in preparation), and that reported in Chapter 3 (section 3.4.3.1) have failed to identify a significant association between this risk marker and challenging behaviour within the group of children with a more severe intellectual disability. Whilst this is not the case for all studies (e.g. Chadwick *et al*., 2000) it is assumed that children with severe intellectual disabilities are already at high risk of challenging behaviour and thus this study will focus on the behavioural characteristics of this group of children.

Therefore, the potential of severity of intellectual disability as a risk marker within this group will not be examined, although its influence on the ability of the putative risk markers to predict the presence of challenging behaviour will be investigated. Examination of the less well established putative risk marker, teacher concern, will also be conducted in order to potentially replicate the findings of Murphy *et al.* (1999).

As well as providing potential longitudinal evidence for these putative risk markers, a second broad aim of this study is to make an initial estimate regarding the number of children with these risk markers who would require effective intervention in order to significantly reduce the frequency of challenging behaviour in this population. Given the evidence in support of the operant processes underlying the development of challenging behaviour (e.g. Oliver, Hall & Murphy, 2005) and the success of behavioural interventions within early intervention programmes, as illustrated in autism (Lovaas, 1987; Eldevik, Hastings, Hughes, Jahr, Eikeseth, *et al.,* under review) such interventions would seem appropriate. However, perhaps more important than the nature of the intervention at this point is the identification of those at risk and thus examining whether pursuing early intervention on the basis of these putative risk markers is appropriate.

In contrast to these more pragmatic aims, a third and more theoretically driven aim of this paper is to test the predictive models of challenging behaviour developed in the screen study (section 3.4.3.3). These models indicated that some forms of challenging behaviour were predicted by different risk markers, so that destruction and aggression were both predicted by overactivity/impulsivity and self-injury by repetitive and

restricted behaviours and interests. By model building at follow up, the longitudinal significance of these models will be examined.

In summary the aims of this study are to:

a) Examine the persistence and increased severity of challenging behaviour in young children with severe intellectual disability between an initial screen and an 18 month follow up.
b) Test whether the putative risk markers (repetitive and restricted behaviours and interests, overactivity/impulsivity, health and concern) can predict the presence of three forms of challenging behaviour (aggression, destruction and self-injury) 18 months later, and more specifically the remission, incidence and persistence of these challenging behaviours. Numbers needed to treat will also be generated based on these results. The effect of severity of intellectual disability on the ability of the putative risk markers to predict the remission, incidence and persistence of challenging behaviour will also be examined.
c) Test the theoretical models of challenging behaviour formulated at screen (section 3.4.3.3) whilst controlling for potential confound of variables.

Hypotheses

a) It was proposed in Chapter 3 (section 3.5) that the presence of repetitive, impulsive and overactive behaviour in children with a severe intellectual disability might indicate compromised behavioural inhibition in this population. Given this proposition, it is hypothesised that these behaviours will persist in the behavioural repertoire of participants due to an underlying neuropsychological vulnerability. Consequently, these putative risk markers

will continue to predict presence, and particularly the persistence of challenging behaviour.

b) It is hypothesised that the models of challenging behaviour developed using binary logistic regressions and formulated at screen (section 3.4.3.3) will be significant at follow up, based on the predicted persistence of both the putative risk markers and challenging behaviour. Thus, the composite behavioural risk markers repetitive and restricted behaviours and interests and overactivity/impulsivity will continue to predict the persistence of challenging behaviour 18 months later.

4.3 Method

4.3.1 Measures

The Challenging Behaviour Screening Questionnaire (CBSQ; section 3.3.2)

Developed to assess risk of challenging behaviour, the CBSQ was used as a measure of both the putative risk markers and three forms of challenging behaviour. Including items related to severity of intellectual disability, health, and repetitive, obsessive, impulsive, overactive, aggressive, destructive and self-injurious behaviours, as well as more basic demographic information, the CBSQ is a four page questionnaire specifically designed to be completed by teachers of children with a severe intellectual disability. Both versions of the CBSQ (under 6 years and 6 years and over), which differ only in the way in which severity of intellectual disability is measured, were utilised in this study in order to measure this construct in a sample with a relatively large age range. Further details regarding the development of the

CBSQ and its inter-rater reliability are provided in Chapter 3 (sections 3.3.1 and 3.3.3 respectively). Also presented in Chapter 5 is an examination of the validity of the CBSQ.

Variables included within the CBSQ

The variables utilised were those used and formed at the screen; 'probable autism spectrum disorder (ASD)', severity of intellectual disability, health, repetitive and restricted behaviours and interests, overactivity/impulsivity and challenging behaviour. (See Chapter 3, section 3.3.2, for more detailed descriptions of the variables).

'Probable Autism Spectrum Disorder (ASD)': Teachers were asked to state if a professional had ever said that the participant was autistic, had an autistic spectrum disorder, autistic like traits and/or features of autism. If the teacher endorsed any of these variables, the child was scored as having 'probable ASD'. This variable was used to provide demographic data for the sample as opposed to being examined as a putative risk marker due to the overlap between this variable and the composite repetitive and restricted behaviours interests.

Severity of Intellectual Disability: Severity of intellectual disability was measured using twenty items from the Denver Developmental Screening Test II (DDST II; Frankenburg, Dodds, Archer, Shapiro & Bresnick, 1992) for children less than 6 years of age and ten items from the Wessex Behaviour Scale (Kushlick, Blunden & Cox, 1973) for children aged 6 years and older. Developmental percentile scores were calculated for children aged less than 6 and 6 years or more using the Denver

developmental quotients and Wessex scores respectively. Greater and lesser severity of intellectual disability groups were formed using median splits on these disability percentile data for both the under 6 and 6 years and older groups and then combined to form one high and low group for the whole sample.

Health: Six items from the Health Questionnaire developed by Hall, Arron, Sloneem and Oliver (2008) were included in the screen to assess health problems during the previous month. Based on the results from these items, groups of participants with or without health problems were formed comprising participants with one or more health problems compared to those with none respectively.

Restricted and Repetitive Behaviours and Interests: Two items from the behaviour and emotional difficulties section of the Self-Help and Behaviour Rating Scale (Petty, 2006) were used to assess frequency and severity of repetitive and restricted behaviour. Scores from the items measuring the frequency of each behaviour were combined to form the composite repetitive and restricted behaviours and interests. Participants scoring 4 or 5 on frequency of repetitive and restricted behaviour formed the repetitive and restricted behaviours and interests group whilst those scoring 1, 2 or 3 were considered not to have the putative risk marker. For brevity, repetitive and restricted behaviours and interests is abbreviated to RRBI in some tables and figures.

Overactivity/Impulsivity: Four items from the Activity Questionnaire (Burbidge, 2005) were used in order to assess overactive and impulsive behaviour (two each). Scores from these four items were combined to form the overactivity/impulsivity composite and two groups were formed using a median split, so that those scoring 3

or above formed the overactivity/impulsivity group and those scoring 2 or below were considered not to have the putative risk marker. For brevity, overactivity/impulsivity is abbreviated to O/I in some tables and figures.

Challenging Behaviour: Three items to assess the presence of aggressive, destructive and self-injurious behaviour were taken from the Challenging Behaviour Questionnaire (CBQ; Hyman, Oliver & Hall, 2002). Three items from the Challenging Behaviour Interview (CBI; Oliver, McClintock, Hall, Smith *et al*., 2003) were used to assess the severity of these challenging behaviours. The variable "one or more forms of challenging behaviour" was formed based on the presence of aggression, self-injury and/or destruction. Presence of challenging behaviour groups were formed based on whether the teacher had reported that the child did or did not demonstrate each form of challenging behaviour. Participants were categorised as showing severe challenging behaviour if they scored within the top 20% of severity scores from the CBSQ at screen. Participants also formed four challenging behaviour groups; participants who had not demonstrated challenging behaviour at either the screen or follow up, participants who had challenging behaviour at screen but not follow up (remission), follow up but not screen (incidence) and at both screen and follow up (persistence). These resemble groups formed by Cooper, Smiley and Allan *et al.,* (2009) for the investigation of self-injury.

4.3.2 Recruitment

The headteacher of every school which participated in the screening study (fourteen severe learning disability schools across the West Midlands; see Chapter 3, section 3.3.4) was contacted fifteen months following completion of the screen study and

asked to help trace the location of every child from their school who originally took part.

4.3.3 Participants

Of the original 629 participants, eight had died, two were in hospital and thus not attending school, one could not be traced, four were in the process of moving schools and three had left the UK by the time of the 18 month follow up, reducing the sample to 611 participants. Of the 611 questionnaires sent out, 128 (21%) were not returned. Many of these came from two schools (59 participants) that declined to participate in the follow up study due to time constraints. A further sixteen (2.6%) questionnaires were completed too late to be included in the study (later than the 18 month and 3 week cut off, see section 4.3.4) and another 50 (8.2%) questionnaires had too much missing data to warrant inclusion (over 25% of items missing), leaving a follow up sample of 417 participants, 66.3% of the original sample.

4.3.3.1 Demographics

Basic demographics for the follow up sample are presented in Table 4.1.

Table 4.1: Prevalence (% and number of participants) of putative risk markers and challenging behaviours

Variable	Follow up sample % (n)
Health problems	47.6 (189)
Repetitive and restricted behaviours and interests	54.4 (217)
Overactivity/impulsivity	46.5 (185)
Aggression	33.7 (140)
Destruction	22.3 (92)
Self-injury	17.1 (71)
One or more forms of challenging behaviour	42.8 (177)

To ensure that the follow up sample was representative of the original screen sample and not biased by the attrition of 212 participants, a series of Mann Whitney U and χ^2 analyses were conducted to detect possible significant differences between participants included in the follow up (417) and those from the original sample who were not included. This analysis revealed that the follow up participants were significantly younger (median = 7, IQR = 4) (U = 33559, $p < .001$) than the participants who were not included (median = 8, IQR = 4). The follow up participants also had a significantly more severe intellectual disability (median = 0, IQR = 1) (U = 35886, $p = .002$) than the participants who were not included (median = 1, IQR = 1). Concern regarding the destruction demonstrated by the follow up group (median = 2, IQR = 2) was also significantly less (U = 663.5, $p = .004$) than for the remaining participants (median = 3, IQR = 2).

There were no significant differences between groups with regard to the putative risk markers; repetitive and restricted behaviours and interests, overactivity/impulsivity, health, concern regarding aggression and concern regarding self-injury (Appendix C shows the median and inter-quartile range differences for the putative risk markers between participants who were and were not included in the follow up sample). Similarly, no significant differences between these groups in terms of frequency or severity of challenging behaviour were found (Appendix D shows the Chi Square results for differences in frequency or severity of challenging behaviour between groups who were and were not included in the follow up sample), except for aggression, the frequency of which was greater in the follow up sample, than participants who were not included (χ^2 (1, N = 30) = 3.27, $p < .05$).

In summary, there were no significant differences in the challenging behaviour or putative behavioural risk markers (with the exception of concern regarding destruction) demonstrated between participants in the follow up sample or those from the original sample who were not included, indicating that the follow up participants were a representative sample of the original participants. However, this is not to say that they are significantly similar as the tests used are more sensitive as a test of difference than similarity.

Ranging in age from 4 to 14 years, the mean age of the follow up sample was 8.57 (SD = 2.56). Teachers reported that 83 (20%) participants had a genetic syndrome, the most common of which reported was Down Syndrome (n = 20, 4.8%). This is far less than would be predicted, indicating the inaccuracy of these data.

4.3.4 Procedure

One month before the follow up questionnaires were due to be completed, the location of all the participants had been traced and one questionnaire per participant was sent to the headteacher of each participant's school. If a participant had moved schools, the headteacher of their new school was contacted and invited to participate in the screening study and sent a questionnaire per participant two weeks before the questionnaire was due for completion. This involved contacting a further 24 schools.

Participants' teachers were encouraged to complete the questionnaire as close to the 18 month follow up as possible. The shortest time between completion of the questionnaires at screen and then follow up was 17 months and 1 week, whilst the longest was 18 months and 3 weeks. Parental consent for the follow up study had already been obtained at the same time as the original screening study, although in a few cases whereby the participants' guardian had changed between the screen and follow up, consent was obtained from the new guardian before the questionnaire was sent to the participant's teacher. Ethical approval for this study was obtained from the School of Psychology ethical review committee at the University of Birmingham.

4.3.5 Data Analysis

When examining basic group comparisons, parametric tests were used unless the data were not normally distributed. Bonferonni corrected Cramer's phi tests were conducted in order to examine the persistence of both the putative risk markers and challenging behaviour. Relative risk analysis was also conducted to examine the ability of the putative risk markers to predict the presence of challenging behaviour 18 months later, and particularly the remission, incidence and persistence of challenging

behaviour. In order to ascertain how many participants would require effective intervention in order to produce a successful outcome in one participant, i.e. significant reduction in the frequency of challenging behaviour, numbers needed to treat analyses were conducted. Because of the large number of relative risk and numbers needed to treat tests conducted, 99.9% confidence intervals (p <.001) were used. Finally, a series of binary logistic regressions was also conducted to control for the potentially confounding overlap between variables in the relative risk analysis and to test the theoretical predictive models developed at the screen (section 3.4.3.3) for the presence of challenging behaviour. Bonferonni corrections were applied to the Alpha levels for these analyses.

4.4 Results

4.4.1 Remission, incidence and persistence of the presence and severity of challenging behaviour

In order to examine the remission, incidence and persistence of the presence and severity of challenging behaviour, the percentage of the sample who showed challenging behaviour and severe challenging behaviour at the screen but not the follow up study, the follow up but not the screen study and both the screen and follow up study respectively was calculated (Table 4.2). Cramer's phi analysis was also conducted in order to statistically assess the persistence of challenging behaviour.

Table 4.2: Percentage and number of participants in remission, incidence and persistence and no behaviour groups and analysis examining the persistence of challenging behaviour between the screen and follow up studies (left of the bold line). Remission and persistence of challenging behaviour in participants showing the behaviour at screen (right of the bold line).

Challenging behaviour	No behaviour at either stage	Remission	Incidence	Persistence	*P* (1 tailed)	Remission in participants with behaviour at screen	Persistence in participants with behaviour at screen
Aggression	57.04 (235)	9.47 (39)	12.38 (51)	21.12 (87)	**<.001**	30.95 (39)	69.05 (87)
Destruction	70.32 (289)	7.3 (30)	12.65 (52)	9.73 (40)	**<.001**	42.86 (30)	57.14 (40)
Self-injury	76.16 (313)	7.06 (29)	7.06 (29)	9.73 (40)	**<.001**	42.03 (29)	57.97 (40)
One or more forms	46.62 (193)	10.6 (44)	14.49 (60)	28.5 (117)	**<.001**	27.33 (44)	72.67 117
Severe aggression	89.02 (365)	3.66 (15)	5.37 (22)	1.95 (8)	**<.001**	65.22 (15)	34.78 (8)
Severe destruction	92.93 (381)	3.17 (13)	3.42 (14)	.49 (2)	0.03	86.67 (13)	13.33 (2)
Severe self-injury	95.35 (390)	1.96 (8)	1.47 (6)	1.22 (5)	**<.001**	61.54 (8)	38.46 (5)
One or more severe forms	84.39 (346)	4.63 (19)	7.07 (29)	3.9 (16)	**<.001**	54.29 (19)	45.71 (16)

Significance = $p < .015$

The Cramer's phi analysis shown in Table 4.2 indicates that, as predicted, challenging behaviour was highly stable across the 18 month follow up period, with the only significant change in the demonstration of challenging behaviour between screen and follow up being severe destruction. Although, for all forms of challenging behaviour, the majority of participants either continued to demonstrate the behaviour or still did not show it, indicating the general stability of challenging behaviour. Of the forms of challenging behaviour, aggression was the most persistent, with over 21% of participants demonstrating aggression at screen and continuing to demonstrate the behaviour at follow up. With regard to challenging behaviour as a whole, over 300 participants (over 75%) either never showed challenging behaviour or were persistent in their demonstration of challenging behaviour.

4.4.1.1 One year incidence and persistence of challenging behaviour at follow up in participants demonstrating the behaviour at screen

Incidence during 1 year and the persistence of challenging behaviour at follow up in participants demonstrating the behaviour at the screen study was calculated (Table 4.3).

Table 4.3: One year incidence and persistence of challenging behaviour in participants demonstrating challenging behaviour at screen

Challenging behaviour	1 year incidence (%)	Persistence in challenging behaviour group only (%) n
Aggression	8.25	69 (87)
Destruction	8.43	57.1 (40)
Self-injury	4.71	58 (40)
One or more forms	9.66	72.7 (117)
Severe aggression	3.58	34.8 (8)
Severe destruction	2.28	13.3 (2)
Severe self-injury	.98	38.5 (5)
One or more severe forms	4.71	45.7 (16)

The results as illustrated in Table 4.3 indicate the high persistence and incidence of challenging behaviour in this group, with an incidence rate of almost 10% and nearly three quarters of the participants with one or more forms of challenging behaviour at screen continuing to demonstrate this behaviour at follow up. In terms of specific forms, aggression had the highest incidence and persistence. The incidence and persistence of severe challenging behaviour however was lower.

4.4.2 Association between challenging behaviour at the screen study and severity of challenging behaviour at follow up

Associations between the presence of challenging behaviour at screen and severity of challenging behaviour at follow up were investigated. Mann Whitney U analysis demonstrated that participants showing aggression at screen demonstrated significantly more severe aggression at follow up (median = 12; U = 1687, $p < .05$)

than children who started to show aggression after the screen (median = 8), although there were no significant differences in severity of destruction (U = 930.5, p = .47) or self-injury (U = 505, p = .36) between participants who did or did not demonstrate these behaviours at screen. A series of Spearman's correlations also indicated significant associations for severity of aggression (r = .256, p = .017) and self-injury (r = .424, p = .007), but not destruction (r = 0, p = .999) between the screening and follow up stages. These analyses indicate that participants with aggression at screen demonstrated significantly more severe aggression at follow up than those who did not demonstrate aggression at screen. Participants with severe aggression or self-injury at screen were also more likely to continue to evidence the same level of severity of these behaviours at follow up.

4.4.3 Persistence of the putative risk markers

Cramer's phi analysis was used in order to examine the persistence of the putative risk markers between the screen and follow up studies. This analysis showed that, as predicted, the majority of putative risk markers were persistent with no significant differences in frequency of the variables overactivity/impulsivity, severity of intellectual disability and concern regarding aggression, destruction and self-injury (Table 4.4).

Table 4.4: Percentage and number of participants with a putative risk marker at screen but not follow up, follow up but not screen, both screen and follow up and at neither screen nor follow up and Cramer's phi analysis examining the persistence of the putative risk markers between the screen and follow up

Putative risk marker	Screen - absent Follow up - absent	Screen - present Follow up - absent	Screen - absent Follow up - present	Screen - present Follow up - present	*p* (1 tailed)
Repetitive and restricted behaviours and interests	34.97 (128)	10.38 (38)	18.03 (66)	36.61 (134)	**<.001**
Overactivity/ impulsivity	31.88 (110)	17.68 (61)	12.75 (44)	37.68 (130)	**<.001**
Health	40.79 (155)	12.11 (46)	21.05 (80)	26.05 (99)	**<.001**
Severity of intellectual disability	43.54 (172)	9.87 (39)	10.38 (41)	36.2 (143)	**<.001**
Concern regarding aggression	68.84 (274)	9.3 (37)	12.56 (50)	9.3 (37)	**<.001**
Concern regarding destruction	92.14 (375)	3.43 (14)	3.69 (15)	.74 (3)	**<.001**
Concern regarding self-injury	91.58 (370)	2.72 (11)	2.72 (11)	2.97 (12)	**<.001**

Bold = $p < .007$

Each putative risk marker was significantly persistent across the 18 month follow up period.

4.4.4 Ability of the putative behavioural risk markers to significantly predict the presence of challenging behaviour at follow up

The ability of the putative behavioural risk markers, repetitive and restricted behaviours and interests and overactivity/impulsivity at the screen study, to significantly predict the presence of challenging behaviour at follow up was examined using relative risk analyses (Figure 4.1).

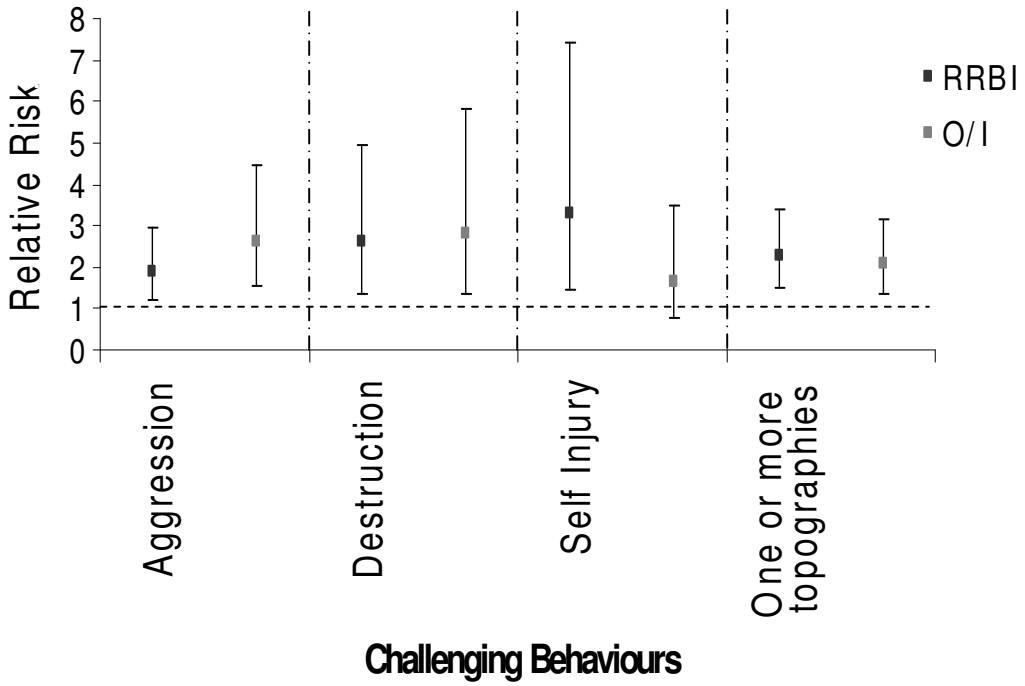


Figure 4.1: Relative risk of challenging behaviour at follow up given the presence of RRBI (repetitive and restricted behaviours and interests) and O/I (overactivity/impulsivity) at screen

Figure 4.1 shows that, as predicted, the relative risk of the presence of each form of challenging behaviour at follow up given the presence of the behavioural risk markers at screen was significantly increased, with the exception of self-injury, the relative

risk of which was not increased by the presence of overactivity/impulsivity. Whilst there was some variation in the extent to which each putative behavioural risk marker increased the relative risk of each form of challenging behaviour, the majority of the analyses generated relative risk results around two, indicating that the risk of each behaviour was approximately doubled given repetitive and restricted behaviours and interests or overactivity/impulsivity.

4.4.5 Numbers needed to treat challenging behaviour at follow up

Numbers needed to treat analyses were also conducted to identify the number of participants who would need to be treated effectively to significantly reduce the frequency of challenging behaviour in one participant given the presence of repetitive and restricted behaviours and interests and overactivity/impulsivity at screen (Table 4.5).

Table 4.5: Numbers needed to treat challenging behaviour given the presence of repetitive and restricted behaviours and interests and overactivity/impulsivity at screen

Putative Risk Marker at Screen	Challenging Behaviour At Follow Up	Numbers Needed to Treat (CI)
Repetitive and restricted behaviours and interests	Aggression	**4.66 (2.75, 15.3)***
	Destruction	**4.58 (2.84, 11.9)***
	Self-injury	**5.22 (3.21, 13.98)***
	One or more Forms	**2.9 (2.03, 5.12)***
Overactivity/impulsivity	Aggression	**3.19 (2.17, 5.97)***
	Destruction	**4.46 (2.8, 10.91)***
	Self-injury	11.47 (-4.75, 27.69)
	One or more Forms	**3.17 (2.13, 6.22)***

*** = significant at 99.9% confidence intervals**

These analyses indicated that, for each challenging behaviour, the risk of which was significantly increased by the putative risk markers, less than six children would need to be treated and the most clinically significant results were produced for participants showing any, rather than a particular form of, challenging behaviour. Illustrating this, the lowest numbers needed to treat one or more forms of challenging behaviour were less than three for repetitive and restricted behaviours and interests.

4.4.6 Ability of the putative behavioural risk markers to significantly predict the severity of challenging behaviour at follow up

The ability of the putative behavioural risk markers, repetitive and restricted behaviours and interests and overactivity/impulsivity at the screen, to significantly predict the severity of challenging behaviour at follow up could not be analysed using relative risk analysis as high and low severity of challenging behaviour groups could not be formed due to the small number of children who demonstrated severe challenging behaviour. Instead, Mann Whitney U analyses were conducted in order to investigate any significant differences between the severity of challenging behaviour at follow up demonstrated by participants with or without the putative behavioural risk markers at screen (Table 4.6).

Table 4.6: Mean, standard deviation and Mann Whitney U scores for severe challenging behaviour in participants with and without the putative risk marker at screen

Putative risk markers	Challenging Behaviour	No risk marker at screen	Risk marker at screen	U	*p*
Repetitive and restricted behaviours and interests	**Severe Aggression**	3.96 (10.62)	9.18 (18.89)	13956.5	<.001*
	Severe Destruction	1.73 (6.12)	7.56 (18.3)	13578.5	<.001*
	Severe self-injury	1.05 (4.5)	5.57 (16.35)	14544.5	<.001*
	One or more severe forms	6.67 (17.83)	22.27 (42.3)	11190	<.001*
Overactivity/ Impulsivity	**Severe Aggression**	2.01 (5.84)	10.49 (19.35)	10546	<.001*
	Severe Destruction	2.42 (9.71)	6.8 (16.71)	12175.5	<.001*
	Severe self-injury	2.09 (8.13)	4.36 (14.55)	14625.5	.027
	One or more severe forms	6.65 (16.16)	21.46 (41.3)	10405	<.001*

*** = *p* < .013**

The results of this analysis demonstrated that participants with repetitive and restricted behaviours and interests or overactivity/impulsivity at screen demonstrated significantly more severe challenging behaviour at follow up. The only exception to this was that there was no significant difference in the severity of self-injury between participants with or without overactivity/impulsivity at screen.

4.4.7 The ability of the putative risk markers to predict the remission, incidence and persistence of challenging behaviour

Relative risk analyses were also conducted to investigate whether repetitive and restricted behaviours and interests and overactivity/impulsivity were significant predictors of the remission, incidence or persistence of challenging behaviour. Each of the three groups was compared to the remaining participants who showed no challenging behaviour at either time to generate relative risk indices (Figure 4.2).

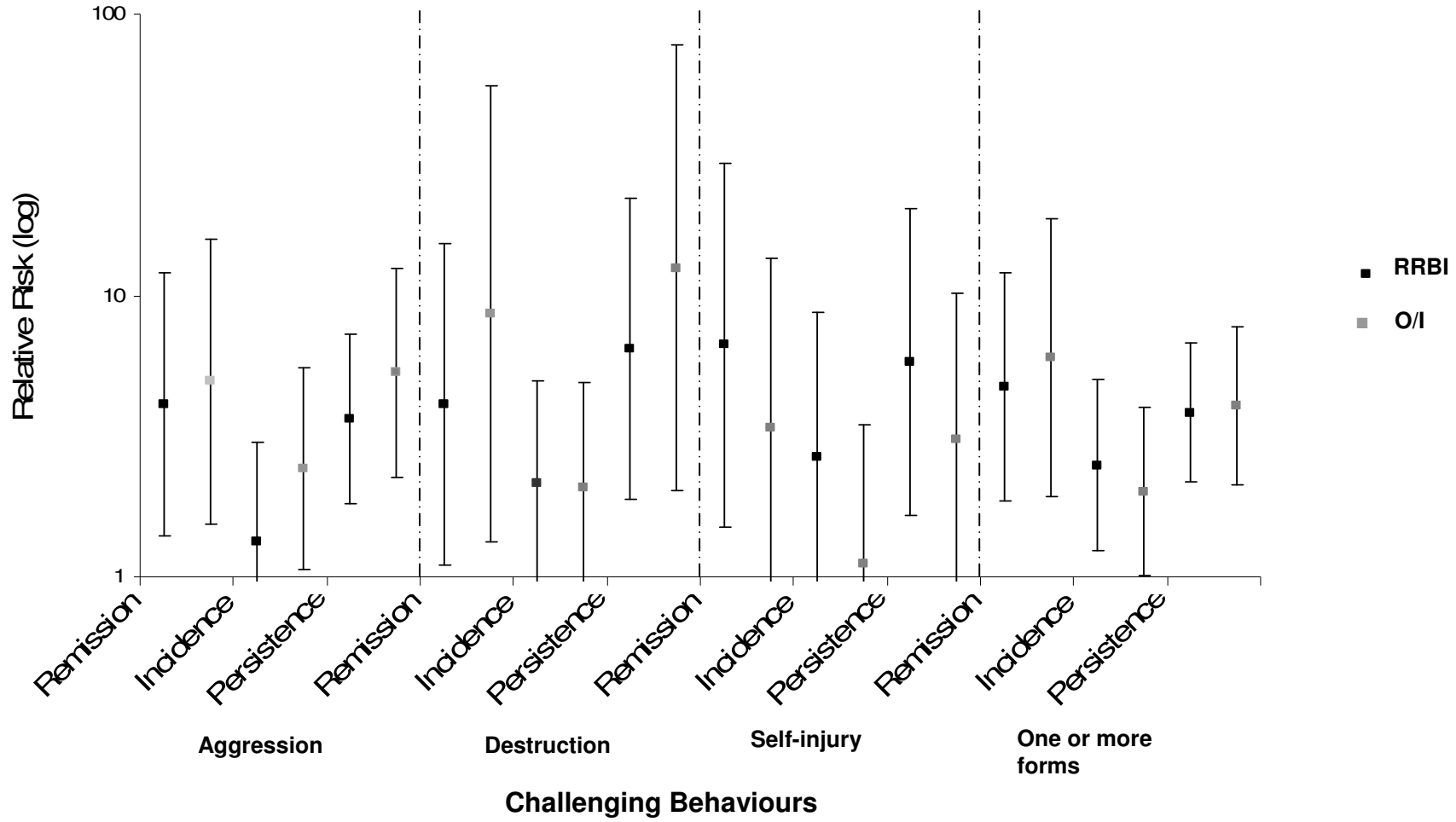


Figure 4.2: Relative risk of remission, incidence and persistence of challenging behaviour given the presence of repetitive and restricted behaviours and interests and overactivity/impulsivity at screen

As demonstrated in Figure 4.2, the putative behavioural risk markers repetitive and restricted behaviours and interests and overactivity/impulsivity significantly predicted the persistence and remission of challenging behaviour. For example, the relative risk of persistent destruction given repetitive and restricted behaviours and interests was 12.51, so that participants with this putative behavioural risk marker were over twelve times more likely to demonstrate destruction than participants without the risk marker. Only the association between self-injury and overactivity/impulsivity failed to reach significance. The ability of both repetitive and restricted behaviours and interests and overactivity/impulsivity to predict the incidence of challenging behaviour however was weaker, with five of the relative risk indices generated revealing associations which were not significant (repetitive and restricted behaviours and interests and incidence of aggression, destruction and self-injury and overactivity/impulsivity and incidence of destruction and self-injury). These results indicate that there are differences in the associations between the putative risk markers and challenging behaviour so that repetitive and restricted behaviours and interests appear to predict more forms of challenging behaviour than overactivity/impulsivity.

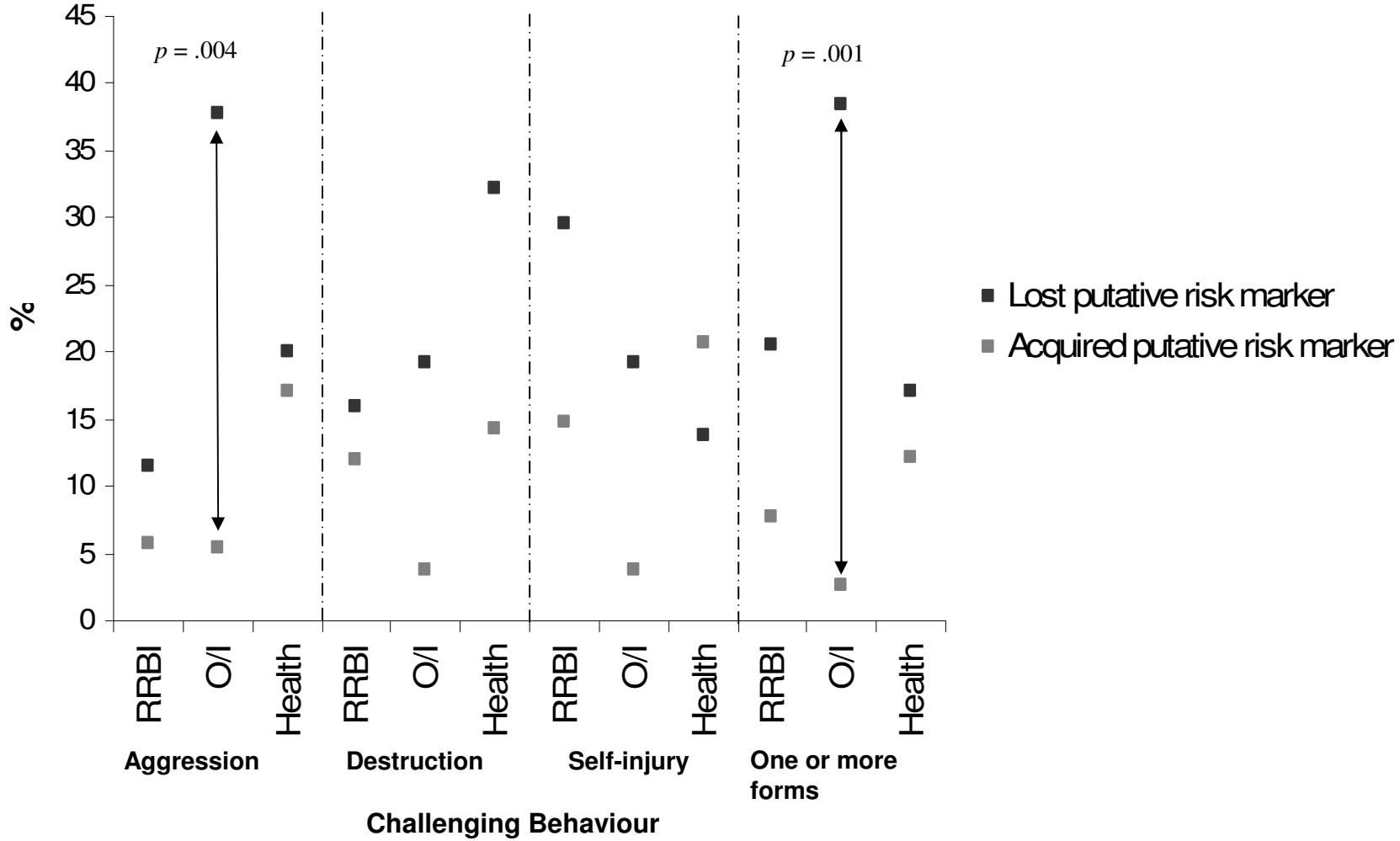
The remaining relative risks however were significant, indicating that these putative behavioural risk markers are generally robust, significant predictors of the remission and persistence of challenging behaviour. Some of the relative risks were particularly high, as illustrated by the relative risk of destruction given overactivity/impulsivity, which was significantly increased more than twelve fold. Whilst the relative risks were not necessarily greater for any compared to a specific form of challenging behaviour given these putative risk markers, one or more forms were better predicted by repetitive and restricted behaviours and interests and overactivity/impulsivity, with

significant relative risks for transient (remission and incidence) and persistent challenging behaviour.

4.4.7.1 Change in the putative risk markers in participants in remission and incidence

To provide further insight into the nature of remission and incidence in this population, a series of Cramer's phi analyses was conducted. The aim of this analysis was to decipher whether the behavioural characteristics of these participants had changed since the screen. To test this, Cramer's phi analysis compared the repetitive and restricted behaviours and interests, overactivity/impulsivity and health problems at screen and follow up of participants with challenging behaviour at remission and incidence for all forms. The results of this analysis are shown in Figure 4.3.

a)


45
40
35
30
25
20
15
10
5
0
%
p = .004
p = .001
RRBI
O/I
Health
Aggression
Destruction
Self-injury
One or more forms
Challenging Behaviour
Lost putative risk marker
Acquired putative risk marker

b)

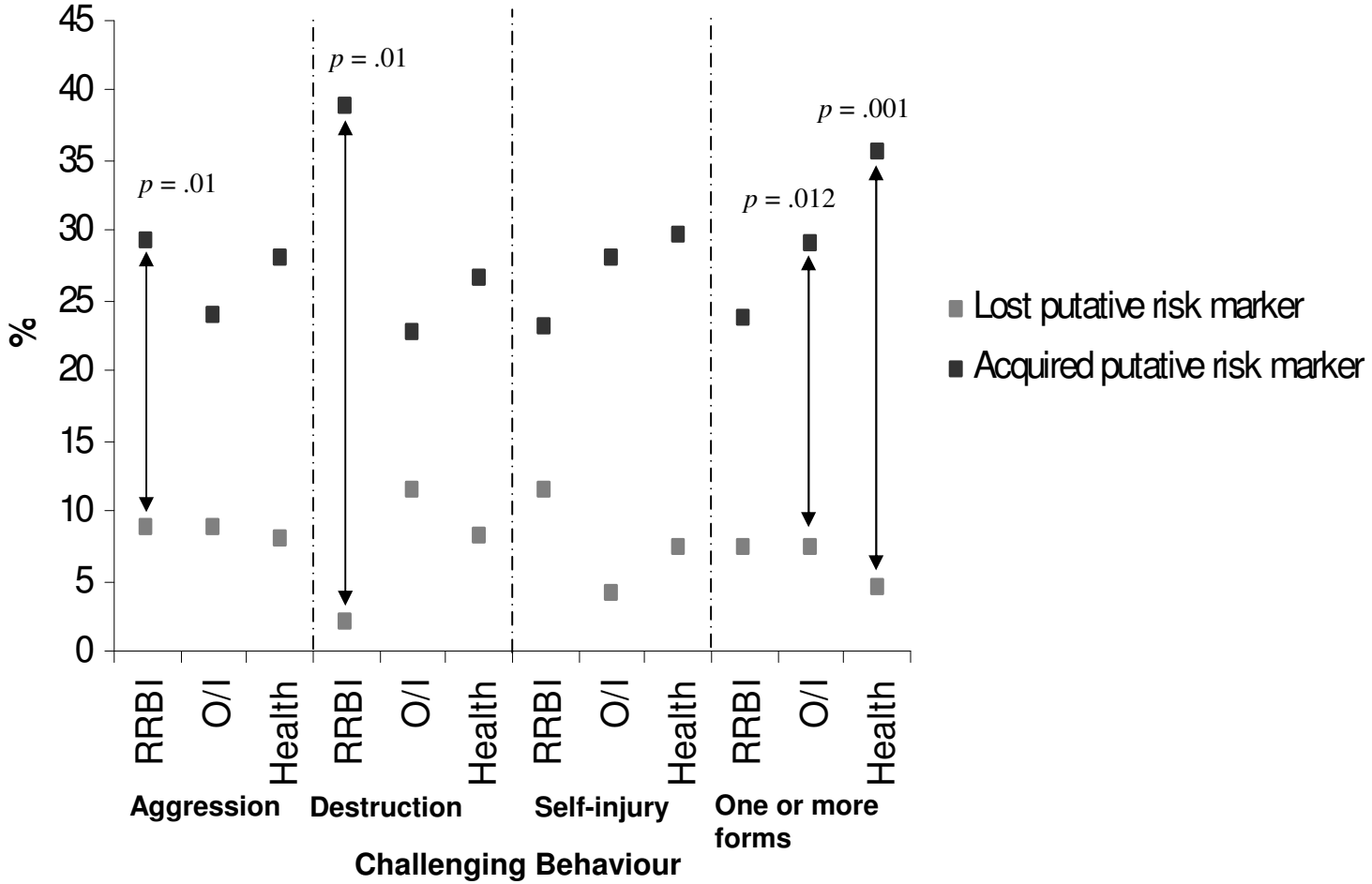


Figure 4.3a and 4.3b: Percentage of participants with challenging behaviour in remission (a) and incidence (b) who lost or acquired the putative risk markers

As demonstrated by Figure 4.3a, for participants in remission for aggression, there were significant differences in overactivity/impulsivity, so that significantly more participants lost the putative behavioural risk marker than acquired it. Similarly, there were significant changes in the overactivity/impulsivity shown by participants in remission for one or more challenging behaviours, so that significantly more participants acquired than lost the putative behavioural risk marker. There were no significant changes in the putative risk markers shown for the remission of destruction or self-injury, and health or repetitive and restricted behaviours and interests did not differ significantly for aggression or one or more forms of challenging behaviour. Despite these non significant differences, with the exception of health and self-injury, there was a general trend of participants losing their putative risk marker between screen and follow up, with fewer participants acquiring a putative behavioural risk marker during this time period.

Figure 4.3b demonstrated that for the incidence of aggression, there were significant changes in the demonstration of repetitive and restricted behaviours and interests, so that significantly fewer participants lost their putative behavioural risk marker than acquired it. Similarly, for the incidence of destruction, significantly fewer participants lost their repetitive and restricted behaviours and interests than participants who acquired them. There were also significant differences in the demonstration of overactivity/impulsivity and health for the incidence of one or more forms of challenging behaviour, so that significantly fewer participants lost these putative risk markers than acquired them. There were no significant changes in the putative risk markers shown for the remission of self-injury, and health or repetitive and restricted behaviours and interests did not differ significantly for aggression or destruction, or

repetitive and restricted behaviours and interests for one or more forms of challenging behaviour. In contrast to the remission data already presented (Figure 4.3a), the results illustrated in Figure 4.3b followed a general trend of participants acquiring a putative risk marker during the 18 month follow up, with fewer participants losing their putative risk marker.

These results indicate that there is a relationship between the remission and incidence of least some forms of challenging behaviour and the putative risk markers and that the relationship depends on the form of challenging behaviour. For the majority of challenging behaviours in remission or incidence however, there was no significant change in the putative risk markers shown at screen or follow up.

4.4.8 Numbers needed to treat the remission, incidence and persistence of challenging behaviour

Numbers needed to treat analyses were also conducted with regard to the remission, incidence and persistence of challenging behaviour given the presence of repetitive and restricted behaviours and interests and overactivity/impulsivity at screen (Table 4.7).

Table 4.7: Numbers needed to treat the remission, incidence and persistence of challenging behaviour given repetitive and restricted behaviours and interests and overactivity/impulsivity

Challenging behaviour	Risk marker at screen	Group	Number needed to treat (CI)
Aggression	Repetitive and restricted behaviours and interests	Remission	**4.89 (2.79, 19.75)***
		Incidence	17.69 (-4.57, 9.46)
		Persistence	**2.94 (2.01, 5.48)***
	Overactivity/ impulsivity	Remission	**4.15 (2.5, 12.21)***
		Incidence	**5.51 (2.85, 83)***
		Persistence	**2.43 (1.77, 3.88)***
Destruction	Repetitive and restricted behaviours and interests	Remission	**7.94 (4.1-123.22)***
		Incidence	7.95 (-3.8, 85.5)
		Persistence	**4.49 (2.84, 10.65)***
	Overactivity/ impulsivity	Remission	**6.2 (3.64, 20.68)***
		Incidence	8.58 (-3.95, 50.49)
		Persistence	**4.11 (2.74, 8.22)***
Self-injury	Repetitive and restricted behaviour and interests	Remission	**6.54 (3.78, 24.17)***
		Incidence	11.5 (-5.09, 44.85)
		Persistence	**5.58 (3.36, 16.42)***
	Overactivity/ impulsivity	Remission	9.79 (-4.87, 1268.88)
		Incidence	106.56 (-8.73, 10.44)
		Persistence	8.62 (-4.41, 189.38)
One or more forms	Repetitive and restricted behaviour and interests	Remission	**3.02 (1.89, 7.51)***
		Incidence	**3.98 (2.19, 21.87)***
		Persistence	**2.03 (1.53, 3.03)***
	Overactivity/ impulsivity	Remission	**2.89 (1.87, 6.33)***
		Incidence	5 (-2.47, 189.24)
		Persistence	**2.01 (1.52, 3)***

* = significant at 99.9% confidence intervals

The results illustrated in Table 4.7 indicate that fewer participants would require effective treatment in order to produce a significant reduction in challenging behaviour when measured as presence of any forms, and without considering exact forms of challenging behaviour. Indeed, as few as two participants with overactivity/impulsivity demonstrating one or more forms of challenging behaviour at both screen and follow up would require effective intervention for one of the participants to show a significant reduction in challenging behaviour. However, even the highest number needed to treat was less than eight, demonstrating the clinical significance of repetitive and restricted behaviours and interests and overactivity/impulsivity for challenging behaviour in children with severe intellectual disabilities.

4.4.9 Health as a putative risk marker

The relative risk of each form of challenging behaviour given the presence of health problems at screen was also calculated in order to examine the ability of this putative risk marker to predict challenging behaviour over time. In contrast to repetitive and restricted behaviours and interests and overactivity/impulsivity, health did not significantly increase the relative risk of any form of challenging behaviour (Appendix E).

4.4.10 The relative risk of challenging behaviour given the putative risk markers across severity of intellectual disability

To detect whether the relative risk of challenging behaviour given the behavioural putative risk markers differed significantly according to severity of intellectual disability, the relative risk of the remission, incidence and persistence of one or more

forms of challenging behaviour given repetitive and restricted behaviours and interests and overactivity/impulsivity was conducted using low and high severity of intellectual disability groups (formed using a median split). The results of this analysis are demonstrated in Table 4.8.

Table 4.8: Relative risk of remission, incidence and persistence of one or more forms of challenging behaviour given the presence of repetitive and restricted behaviours and interests and overactivity/impulsivity for participants with a low compared to a high degree of intellectual disability

Putative risk marker at screen	Challenging behaviour group	Severity of intellectual disability	Relative Risk (CI)	Numbers needed to treat (CI)
Repetitive and restricted behaviours and interests	**Remission**	**High**	**4.75 (1.16, 19.37)***	**3.38 (1.73, 71.67)***
		Low	**4 (1.12, 14.3)***	**3.14 (1.64, 37.39)***
	Incidence	**High**	2.25 (.8, 6.34)	4.8 (-1.99, 11.71)
		Low	2.44 (.92, 6.52)	3.84 (.79, 26.02)
	Persistence	**High**	**3.98 (1.85, 8.56)***	**1.9 (1.35, 3.23)***
		Low	**3.43 (1.47, 8.04)***	**2.28 (1.44, 5.42)***
Overactivity/ impulsivity	**Remission**	**High**	**6.88 (1.28, 36,83)***	**2.81 (1.47, 32.18)***
		Low	**9.58 (1.05, 87.21)***	**2.68 (1.63, 7.48)***
	Incidence	**High**	1.82 (.64, 5.17)	5.68 (-1.91. 5.87)
		Low	2.38 (.8, 7.08)	4.27 (.97, 25.6)
	Persistence	**High**	**3.76 (1.82, 7.75)***	**1.77 (1.27, 2.89)***
		Low	**5.51 (1.42, 21.42)***	**2.17 (1.44, 4.42)***

* = significant at 99.9% confidence intervals

As demonstrated by the results in Table 4.8, there was no significant increase in the relative risk of incidence of one or more forms of challenging behaviour given repetitive and restricted behaviour and interests or overactivity/impulsivity for either group. The relative risk of remission and persistence of this behaviour was also significantly increased given the putative risk markers for both groups. These results indicate the importance of these putative risk markers for the whole sample, irrespective of severity of intellectual disability.

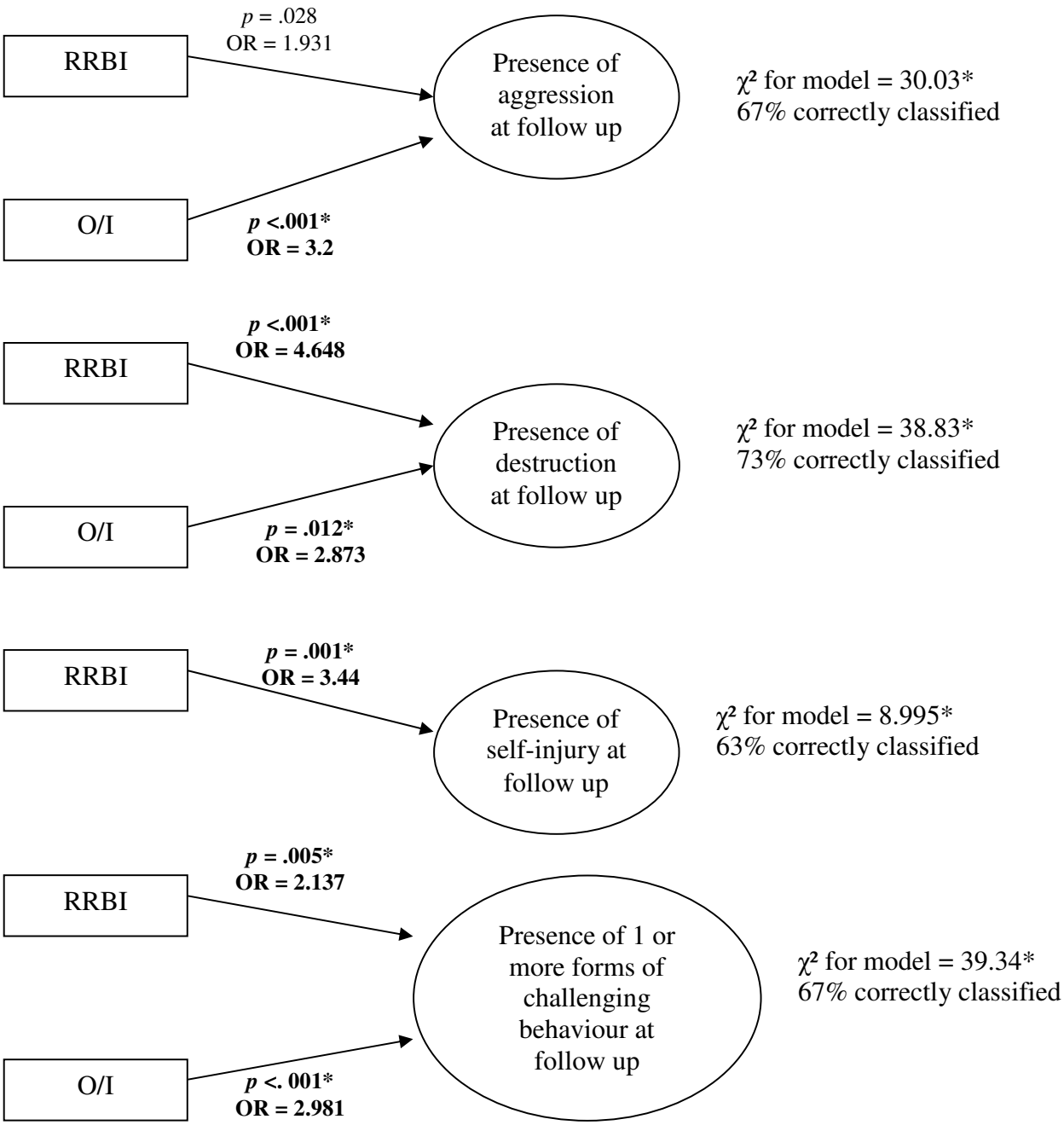
4.4.11 Concern as a predictor of the presence of challenging behaviour

In order to identify significant differences in the degree of concern expressed by teachers at screen regarding children who did and did not demonstrate challenging behaviour at follow up, a series of t-tests was conducted. These analyses indicated that participants who persisted to show self-injury had greater teacher concern at screen (mean = 1.36, SD = 1.57) than participants who remitted (mean = .13, SD = .52) ($t(402) = 11.55$, $p < .001$). There were no significant differences in the degree of concern teachers expressed regarding children who did or did not show aggression ($t(1) = 1.579$, $p > .05$) or destruction ($t(1) = .757$, $p > .05$) at follow up. These analyses indicate the significance of concern as a risk marker for self-injury but not aggression or destruction over an 18 month follow up period.

4.4.12 A predictive model of challenging behaviour

In order to control for the overlap between variables and thus a potential confound, and to test the predictive model of challenging behaviour generated at screen (section 3.4.3.3), a series of binary logistic regressions was conducted, examining associations between repetitive and restricted behaviours and interests and overactivity/impulsivity

at screen and the presence of challenging behaviour at follow up. If the relative risk of a form of challenging behaviour was significantly greater in participants with the putative behavioural risk markers, it was entered into the regression analysis as a predictor variable. The results of this analysis are demonstrated in Figure 4.4.



*** = p < .0125**

Figure 4.4: Binary logistic regression models predicting the presence of challenging behaviour

As demonstrated in Figure 4.4, each of the models significantly predicted each form of challenging behaviour, as well as the presence of one or more forms of challenging

behaviour. However, different forms of challenging behaviour were predicted by different risk markers. For example, whilst one or more forms of challenging behaviour and destruction specifically were predicted by both overactivity/impulsivity and repetitive and restricted behaviours and interests, aggression and self-injury were significantly predicted by just overactivity/impulsivity and repetitive and restricted behaviours and interests respectively. Additionally, whilst RRBI did not significantly predict the presence of aggression, it clearly contributed to the model, with an odds ration of 1.931.

4.5 Discussion

The primary aim of this study was to test the ability of the putative risk markers to significantly predict the presence, and more specifically, the remission, incidence and persistence of three forms of challenging behaviour; aggression, destruction and self-injury. As well as examining the persistence of both challenging behaviour and the putative risk markers, a second aim was to test the models of challenging behaviour formulated at screen (section 3.4.3.3), which indicated different associations between the putative risk markers and challenging behaviour depending on the form of behaviour.

A major strength of the study was the size of the follow up sample. Tracing children to 36 schools across the UK enabled a sample to be obtained which was almost two thirds of that recruited at screen. Statistical analysis also revealed that there were few significant differences in the putative risk markers or challenging behaviour demonstrated by participants who were or were not included in the sample. The strict criteria by which data were excluded from the study, including a small 6 week time

window and the amount of missing data, ensured that the results gained were a true reflection of an 18 month follow up, using a similar sample for all analyses. The achievement of these results using a far more accessible and brief measure indicates a real progression in this research.

As predicted and supporting previous findings (e.g. Emerson *et al*., 2001a), challenging behaviour was found to be highly persistent between the screen and follow up studies. Persistence rates of challenging behaviour within participants who demonstrated the behaviour at screen were also high and comparable to those reported in some previous studies (e.g. Emerson *et al.,* 2001b), although they were also a little lower than those reported in others (e.g. Kiernan & Alborz, 1996; Nottestad & Linaker, 2002). The persistence of severe challenging behaviour was also lower than in previous studies (e.g. Emerson et *al.,* 1996; Murphy *et al.,* 1993), although this was a likely result of the strict criteria of severity of intellectual disability employed in this study. In contrast, incidence rates of self-injury and aggression were considerably higher than those published by Cooper, Smiley & Allan *et al.,* (2009) and Cooper, Smilely & Jackson *et al.,* (2009) respectively, probably due to the older age group (aged 16 years and over) employed in Cooper *et al.'s* studies.

With the exception of health, which was unsurprising given the transient nature of and short time period with which the construct was measured, the putative risk markers (repetitive and restricted behaviours and interests and overactivity/impulsivity) were also highly persistent, indicating that, if also predictive of challenging behaviour at follow up, they would be suitable as indicators of risk for persistence in early intervention programmes.

Broadly replicating the relative risk results of the screen study, (section 3.4.3.2) whereby both repetitive and restricted behaviours and interests and overactivity/impulsivity significantly predicted the presence of aggression, destruction and self-injury, both putative behavioural risk markers were found to significantly predict the presence of all three forms of challenging behaviour at follow up, with only overactivity/impulsivity and self-injury failing to significantly increase the risk of self-injury. Numbers needed to treat analysis further highlighted the significance of overactivity/impulsivity and repetitive and restricted behaviours and interests to individuals with a severe intellectual disability and challenging behaviour, indicating that fewer than six participants would require effective intervention to reduce challenging behaviour in one participant if these putative risk markers were used as identification for being at high risk for presence of challenging behaviour in eighteen months time. The number needed to treat decreased further when the putative risk markers were used to predict any as opposed to a particular form of challenging behaviour.

Thus, if these putative risk markers were adopted to identify children at risk, fewer children would require effective intervention to decrease levels of challenging behaviour generally, as opposed to any specific form. This is important clinically for children with intellectual disabilities, many of whom often demonstrate more than one form of challenging behaviour (e.g. Emerson *et al*., 2001b). Additionally, considering the number needed to treat depression with cognitive behavioural therapy, (one of the most widely used and effective interventions for depression according to the Department of Health's 2001 document, Treatment Choice in Psychological Therapies and Counselling Evidence-Based Clinical Practice Guideline) which is

reported in the literature to be approximately six (Paykel, *et al*., 1999), the identification of these putative risk markers appears to be a comparatively effective strategy.

Both putative behavioural risk markers significantly predicted the remission and persistence of challenging behaviour, although again, overactivity/impulsivity was not significantly associated with self-injury. These putative risk markers also predicted the incidence of challenging behaviour less consistently, with repetitive and restricted behaviours and interests showing no significant associations with incidence of aggression, destruction or self-injury and overactivity/impulsivity failing to significantly increase the relative risk of incidence of destruction. The lack of association between overactivity/impulsivity at screen and the incidence of aggression and destruction at follow up was explained by the significantly higher number of participants acquiring than losing the overactivity/impulsivity putative risk marker. However, none of the remaining non significant associations between the putative risk markers at screen and challenging behaviours at follow up could be explained by significant changes in the demonstration of risk markers over this time.

Therefore, it might be that the incidence of aggression and destruction is caused by a mechanism which also brings about the increase in overactive and impulsive behaviour demonstrated by participants. This supposition is supported by the results of a recent study, which identified temporal associations between repetitive and self-injurious behaviour (Petty, Oliver & Allen, 2009). Such temporal associations between the putative behavioural risk markers and challenging behaviour allude to the potential presence and function of some sort of common mechanism, responsible for

the presence of both types of behaviour. If this were proven to be the case, it would indicate that challenging behaviours such as self-injury might not emerge from prior putative risk markers like repetitive and restricted behaviours and interests, as indicated by Guess and Carr (1991), but that these behaviours might co-exist.

In contrast, the non significant associations between the putative risk markers at screen and the remission or incidence of challenging behaviour at follow up which are not associated with change in the prevalence of the putative behavioural risk markers might indicate participants who demonstrate challenging behaviour intermittently, and just happen to be demonstrating, or not, challenging behaviour in the same time window as the follow up study.

In line with the results of the screen study (section 3.4.3.1) which indicated that increased severity of intellectual disability did not increase the relative risk of challenging behaviour, the results of this follow up study indicated that the relative risk of the remission, incidence and persistence of challenging behaviour given the putative risk markers was not influenced by severity of intellectual disability. There were no significant differences in the relative risk of any form of challenging behaviour given the putative risk markers for individuals with low as opposed to high severity of intellectual disability. This indicates that whilst severity of intellectual disability appears to be a robust risk marker of challenging behaviour, it becomes less important in a sample of children with a severe intellectual disability who are already at high risk.

The results of the original screening study identified health as a putative risk marker for the presence and severity of self-injury. At follow up however, health failed to significantly predict the presence, be that remission, incidence or persistence of self-injurious behaviour. This might be expected given the previously discussed transient nature of and short time period with which health problems were measured. These findings do not however minimise the importance of health problems as a risk factor due to their clear association with self-injury at screen and within the wider research literature (e.g. Luzzani, Macchini, Valade, Milani & Selicorni, 2003), as well as the high prevalence of health problems in individuals with an intellectual disability (Berg, Arron, Burbidge, Moss & Oliver, 2007).

With regard to concern, the results showed participants who persisted to show self-injury had higher teacher concern ratings at screen than participants who remitted, although this was not the case for destruction or aggression. This suggests that the concern expressed by teachers regarding self-injury is indicative of the persistence of self-injury. This replicates exactly previous findings whereby teachers' degree of concern significantly predicted increases in self-injury 18 months later (Murphy, Hall, Oliver & Kissi-Debra, 1999) and was in fact the only variable in this particular study to significantly predict increases in self-injury. Teachers spend a considerable amount of time with children with severe intellectual disabilities and often have much experience managing challenging behaviour and observing its development. The combination of these factors might enable teachers to become accurate judges of which children are likely to develop persistent challenging behaviour. Perhaps more probable though, is that teachers who are concerned about the self-injury displayed by a child with a severe intellectual disability are more likely to reinforce its presence, a

supposition predicted by operant models of the behaviour and also proposed by Murphy *et al.*. Thus, the identification of concern as a putative risk marker for self-injury is significant not only due its potential inclusion in early intervention services, but because it also highlights the importance of variables external to the child to challenging behaviour and the significant role of operant reinforcement.

Finally, by controlling for the overlap between the putative risk markers, the series of binary logistic regressions conducted provided support for the models generated at screen (section 3.4.3.3). As previously identified, both repetitive and restricted behaviours and interests and overactivity/impulsivity significantly predicted the presence of one or more forms of challenging behaviour, whilst aggression and self-injury were predicted by only overactivity/impulsivity and repetitive and restricted behaviours and interests respectively. In a deviation from the original models though, both repetitive and restricted behaviours and interests and overactivity/impulsivity both significantly predicted the presence of destruction, as opposed to overactivity/impulsivity alone. Had a less conservative approach been utilised however, RRBI would also have significantly predicted the presence of aggression.

By providing data in line with the theoretical models formulated at screen (section 3.4.3.3), the proposed role of compromised behavioural inhibition in challenging behaviour in this population (as discussed in section 3.5) is strengthened. Based on the work of Barkley (1997a, 1997b) and Turner (1997, 1999) who identified associations between repetitive behaviour, impulsivity, hyperactivity and poor inhibition in ADHD and autism respectively, identification of these behaviours in children with severe intellectual disabilities might indicate similar neuropsychological

underpinnings. This proposition could also build upon previously existing operant models, so that challenging behaviour which is evoked under stimulus conditions and reinforced to become a learned response is more prevalent in children with the putative risk markers caused by compromised behavioural inhibition as they find it even harder to inhibit this learned response.

In order to explain the superior predictive validity of repetitive and restricted behaviours and interests with regard to self-injury, as compared to overactivity/impulsivity, consideration must be given to Guess and Carr's (1991) model. Within this model, repetitive behaviour evolves into self-injury under the influence of social reinforcement, a supposition supported by Oliver (1993, 1995) who further developed this model into a mutual reinforcement paradigm. In the development of self-injury, operant reinforcement might be more effective in children with repetitive and restricted behaviours and interests as they have compromised behavioural inhibition, making operant reinforcement more powerful as well as the structurally similar repetitive behaviour which through positive reinforcement can be shaped into self-injury. Thus, children with repetitive and restricted behaviours and interests are more at risk than children with just overactivity/impulsivity, whose only difficulty is compromised behavioural inhibition (Petty *et al.,* in preparation). However, the existence of a common mechanism involved in the presence of both the putative risk markers and challenging behaviours would render this explanation obsolete. Thus, further investigation into the development of both types of behaviour is required.

In terms of the clinical implications of this study, longitudinal evidence has now been provided for the behavioural variables repetitive and restricted behaviours and interests and overactivity/impulsivity, so that they might, with increased confidence, be accepted and labelled as risk markers. However, before identification of children at risk based on these risk markers occurs, data regarding the validity of the screen are required. More about the children at high risk of challenging behaviour must also be learned so that effective interventions might be provided to successfully reduce challenging behaviour in this population.

4.6 Summary and implications

In Chapter 4, an 18 month follow up study was conducted, employing the CBSQ to examine the ability of the putative behavioural risk markers to predict the remission, incidence and persistence of challenging behaviour in a sample of children with severe intellectual disabilities. As predicted, the persistence of challenging behaviour was high. Replicating the findings of the screening study described in Chapter 3, the results of this chapter also indicated that the two composite behavioural risk markers, restricted and repetitive behaviours and interests and overactivity/impulsivity, significantly predicted the presence of challenging behaviour 18 months later, although both risk markers were more successful in predicting the persistence than either the remission or incidence of challenging behaviour. Concern expressed by teachers at the screen was also significantly higher for participants who persisted to show self-injury and aggression at the follow up, although health did not significantly predict self-injury as it did at the screen.

Replicating the results of Chapter 3 in a longitudinal study has two important implications. Firstly, this longitudinal data highlights the strong association between the putative risk markers and challenging behaviour over time, so that they may no longer be considered putative. This is clinically significant, indicating that many children identified as being at high risk for challenging behaviour will remain so for at least another 18 months and thus are worthy candidates for early intervention. From a theoretical viewpoint, these findings also provide further evidence for the potential role of compromised behavioural inhibition for challenging behaviour in this population.

To provide evidence for the concurrent and convergent validity of the CBSQ, Chapter 5 describes a study using a group comparison design of children identified as being at high or low risk of challenging behaviour from the data obtained in Chapter 3.

Chapter 5

Screening for Risk for the Development of Challenging Behaviour in Children with Severe Intellectual Disabilities: Validation of the Challenging Behaviour Screening Questionnaire and Comparison of High and Low Risk Groups

5.1 Abstract

Aims: The primary aim of this study was to test the concurrent and convergent validity of the Challenging Behaviour Screening Questionnaire (CBSQ), which was developed to assess the risk for development of challenging behaviour in children with severe intellectual disabilities.

Method: Natural observations, questionnaires and objective measures were used to examine the behavioural risk markers and challenging behaviour in children at high or low risk of challenging behaviour.

Results: Supporting the concurrent validity of the CBSQ, the high risk group demonstrated significantly greater durations of overactive, repetitive and impulsive like behaviour (although no significant differences in restricted behaviour were identified). The convergent validity of the CBSQ also appeared to be good, with the exception of data obtained from the Challenging Behaviour Interview. Comparison of the high and low risk groups in terms of child characteristics not measured by the CBSQ and environmental variables experienced indicated that the high risk group received more adult denials and showed significantly less sustained attention.

Discussion: These findings provide further support for the role of compromised behavioural inhibition for challenging behaviour in individuals with intellectual

disabilities as well as providing evidence for the effective use of the CBSQ in early intervention programmes.

5.2 Introduction

The prevalence of challenging behaviour in people with intellectual disabilities is well established within the literature, with a recent study reporting rates within the range of 5.5 and 16.8% (Lowe *et al*., 2007). Research has also shown that the prevalence of challenging behaviour tends to increase with age and that this trend is demonstrated within individuals showing self-injury (e.g. Saloviita, 2000), and to a lesser extent aggression and destruction (e.g. Borthwick-Duffy, 1994). Once established, challenging behaviour is also persistent (Emerson *et al.,* 2001), which is a significant issue given the many negative outcomes associated with challenging behaviour for people with intellectual disabilities (Emerson, 2001; Hastings & Brown, 2002; Murphy *et al.,* 2005; Tausig, 1985).

Research has focused on the potential causes of challenging behaviour in this population and debate remains in this area due to the failure of any one single theory to explain all occurrences of challenging behaviour and few attempts to integrate empirical evidence for different causes (Oliver & Head, 1990; Oliver, 1995). Various competing theories exist, emerging from both biological and behavioural models which, between them, attempt to explain the occurrence of challenging behaviour in people with intellectual disabilities. These divergent theories have both received empirical support within the literature (e.g. Oliver, 1993) and are both likely to be important in explaining a range of challenging behaviours in the population of individuals with heterogeneous cause of intellectual disabilities.

An array of interventions based on these explanations has been developed with varied success. A systematic review conducted by Brylewski and Duggan (1999) found no evidence for the effective use of antipsychotic medication in the treatment of challenging behaviour in adults with intellectual disabilities, resulting, in part, from the paucity of good quality research in this area (Sturmey, 2002; Deb *et al*., 2008). Conversely, numerous methodologically sound studies investigating the use of behavioural interventions have been conducted and a review of those published during the last 35 years reported that most behavioural treatments are highly effective in the treatment of particular forms of challenging behaviour (Kahng, Iwata & Lewin, 2002). More recent meta-analyses have also supported these findings (e.g. Harvey, Boer, Meyer & Evans, 2009). However, the administration of these interventions is complex and time consuming (Murphy *et al.*, 1993) and based upon the operant model which minimises the importance of individual child characteristics to the development of challenging behaviour.

Illustrating the significance of these individual differences is a body of research investigating risk markers for challenging behaviour in children with intellectual disabilities. During the last decade, various child characteristics, or risk markers, associated with challenging behaviour have been detected which could play an important role in the early identification of challenging behaviour. One of the first studies in this area (Chadwick, Piroth, Walker, Bernard & Taylor, 2000) identified severity of intellectual disability as a risk marker, finding an association between destructive and self-injurious behaviour and lower level of intellectual functioning. Replicating this finding, Baghdadli, Pascal, Grisis & Aussilloux (2003) identified

greater delay in adaptive skill as a predictor of self-injury, along with more severe autism, presence of a perinatal condition and lower chronological age.

The importance of degree of intellectual disability and a diagnosis of autism as risk markers for challenging behaviour was substantiated by the results of a meta-analysis (McClintock, Hall & Oliver, 2003), which also highlighted the significance of communicative ability for challenging behaviour. Focusing on behavioural characteristics acting as risk markers, Oliver *et al*. (in preparation) identified repetitive and ritualistic behaviours as predictors of both the presence and severity of all challenging behaviours, a finding originally identified in children with the rare genetic syndrome Cornelia de Lange Syndrome (Oliver, Sloneem, Hall & Arron, 2009) and supported by a review (Petty & Oliver, 2005) and recent study (Arron, Oliver, Berg, Moss & Burbidge, in review). Replicating this methodology, Petty *et al*. (in preparation) reported a significant association between repetitive, overactive and impulsive behaviour and challenging behaviour. Substantiating the importance of impulsivity and overactivity, independent associations between self-injurious and aggressive behaviour and attention deficit hyperactivity disorder have also been identified (Cooper, Smiley, Allan *et al*., 2009; Cooper, Smiley, Jackson *et al*., 2009).

The evidence that early behavioural markers are associated with and might precede challenging behaviour in the literature (e.g. Lord, 1995; Berkson & Tupa, 2000) is significant as not only does it contradict an exclusively operant model which would predict similar rates of challenging behaviour across children, but it illustrates the potential utility of early intervention. In light of the previously discussed limited effectiveness and difficulties in administering the already established interventions,

prevention rather than treatment of challenging behaviour is beginning to receive increased attention. Having already been established in various diverse (e.g. education, NESS, 2005) and allied (e.g. in autism; Eikeseth, Smith, Jahr & Eldevik, 2007) fields, early intervention seems a promising strategy for individuals with intellectual disabilities as it could potentially prevent the development of challenging behaviour before it becomes ingrained in the behavioural repertoire (Richman, 2008; Richman & Lindauer, 2005; Symons, Sperry, Dropik & Bodfish, 2005).

Whilst early intervention based on the premise of risk is a new venture in the field of intellectual disabilities, preliminary steps in this direction have already been taken. Recognising the need to measure risk markers in a quick and accessible way if early intervention were to be successful, the Challenging Behaviour Screening Questionnaire (CBSQ) was developed (section 3.3.1), including items related to all of the risk markers previously established in the literature (ie. diagnosis of autism, severity of intellectual disability, presence of health problems and stereotyped, restricted, impulsive and overactive behaviour, section 3.3.2) in a shorter and more accessible format. By systematically reducing questionnaires previously used in this population to measure the various risk markers, a much more condensed assessment was produced. Having piloted the screen, results indicated that the presence of two composite risk markers, repetitive and restricted behaviours and interests and overactivity/impulsivity, both significantly increased the relative risk of challenging behaviour (section 3.4.3.2). The presence of health problems during the previous month also increased the relative risk of self-injury (section 3.4.3.1).

Binary logistic regressions were also utilised in order to develop theoretical models of challenging behaviour, the results of which demonstrated associations between repetitive and restricted behaviours and interests and overactivity/impulsivity for one or more forms of challenging behaviour (section 3.4.3.3). Interestingly, different forms of challenging behaviour were associated with different risk markers, so that overactivity/impulsivity significantly predicted the presence of destruction and aggression whilst repetitive and restricted behaviours and interests significantly predicted the presence of self-injury (it must be noted however that these results were based on a highly conservative approach and that the models produced would have differed had Bonferonni corrections not been applied). Replicating previous findings using a shorter assessment demonstrated the potential of the CBSQ to identify children at risk of challenging behaviour at one point in time in a quick and efficient way, thus indicating the feasibility of early intervention programmes.

In Chapter 4, the ability of the risk markers included in the CBSQ to predict challenging behaviour over an 18 month period was examined within a follow up study which employed the sample recruited for the original screening study (section 3.3.3.2). Replicating the statistical analysis conducted in the screen study, (section 4.3.5) the results of this study illustrated significant associations between the behavioural risk markers repetitive and restricted behaviours and interests and overactivity/impulsivity with challenging behaviour using relative risk analysis. A series of binary logistic regressions also demonstrated varied associations between challenging behaviour and the risk markers depending on the exact form of challenging behaviour. The results were concordant with the results of the screen study (section 3.4) and both repetitive and restricted behaviours and interests and

overactivity/impulsivity predicted one or more forms of challenging behaviour, repetitive and restricted behaviours and interests predicted self-injury and overactivity/impulsivity predicted aggression. However, in contrast to the original models, both repetitive and restricted behaviours and interests and overactivity/impulsivity predicted destruction as opposed to just overactivity/impulsivity in the original model (as previously discussed, it must be noted however that these results were based on a highly conservative approach and that the models produced would have differed had Bonferonni corrections not been applied).

Thus, the behavioural risk markers appear to be robust predictors of challenging behaviour over time. However, to progress toward early intervention, the screen by which children at high risk of challenging behaviour are identified must also be validated to ensure its accuracy as a measure of the nominated risk markers. This can be achieved by identifying groups of children deemed to be at high or low risk for challenging behaviour using data generated by the CBSQ and comparing these groups on a number of key variables.

Therefore, the primary aim of this study is to test the concurrent and convergent validity of the CBSQ through assessment of the presence of the behavioural risk markers in the low and high risk groups using natural observations and objective measures.

It is hypothesised that in comparison to the low risk group the high risk group will demonstrate significantly greater:

a) Duration of each behavioural risk marker (repetitive and restricted behaviours e.g. hoarding and overactive and impulsive behaviour) during natural observations demonstrating the concurrent validity of the screen.
b) Scores on questionnaires related to the risk markers (the Activity Questionnaire, Burbidge, 2005; the Repetitive Behaviour Questionnaire, Moss, Oliver, Arron, Burbidge & Berg, 2009; the Social Communication Questionnaire – Current Version**,** Rutter, Bailey, Berument, Lord & Pickles, 2003) and objective measures of average movement using an actiwatch, illustrating convergent validity.

The duration of challenging behaviour demonstrated by each group during natural observations will also be measured. It is hypothesised that the high risk group will exhibit more challenging behaviour in association with the presence of the behavioural risk markers.

The design of the validation study also facilitates a comparison of the high and low risk groups in terms of environmental and child characteristics that are independent of the content of the screen. Thus, a secondary aim is to conduct a comparison of the proto-imperative and affect behaviours demonstrated, environmental variables experienced by (including adult interactions and broader situational variables) and the relationship between these constructs in the high and low risk group.

5.3 Method

5.3.1 Measures

The Activity Questionnaire (TAQ; Burbidge, 2005)

This eighteen item questionnaire was used as measure of overactivity and impulsivity and is composed of three subscales (overactivity, impulsivity and impulsive speech). The questionnaire includes items such as "Does the person want things immediately" and "Does the person act as if driven by a motor" scored on a 5 point Likert scale ("0- never/almost never" to "4- always or most of the time") in order to assess the frequency of observable behavioural correlates of hyperactivity. Through summation of subscale items, two subscale scores were calculated; impulsivity (ranging from 0 to 24) and overactivity (ranging from 0 to 36). The reported inter-rater and test-retest reliability co-efficients for the overactivity and impulsivity scale are high at .70 and .74 for overactivity and .87 and .88 for impulsivity.

Repetitive Behaviour Questionnaire (RBQ; Moss, Oliver, Arron, Burbidge & Berg, 2009)

The RBQ is an informant report questionnaire used as a measure of stereotyped and other repetitive behaviour. The questionnaire consists of 19 items forming 5 subscales: stereotyped behaviour, compulsive behaviour, restricted preferences, repetitive language and insistence on sameness. Informants are required to rate the frequency of each repetitive behaviour during the last month on a scale of 0 to 4 (never, once a month, once a week, once a day and more than once a day). Total scores range from 0 to 76. Example items include: object stereotypy (repetitive,

seemingly purposeless movement of objects in an unusual way) and cleaning (excessive cleaning, washing or polishing of objects or parts of the body). The RBQ has been reported to show high test-retest and inter-rater reliability as well as strong concurrent validity (Moss & Oliver, 2008).

Social Communication Questionnaire – Current Version (SCQ; Rutter, Bailey, Berument, Lord & Pickles, 2003)

The SCQ was used as a measure of autism spectrum disorder (ASD). Consisting 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 *et al*.). Good concurrent validity has been found with various other measures of ASD (Bishop & Norbury, 2002; Howlin & Karpf, 2004).

The Vineland Adaptive Behavior Scale Teacher Questionnaire (VABS II; Sparrow, Cicchetti & Balla, 2006)

Participants were matched using the VABS, providing a detailed measure of ability. Comprising of four subscales; communication, daily living, socialisation and motor skills (children aged under 7 years only) and 223 items, the VABS II Teacher Questionnaire assesses adaptive behaviour for students aged between 3 and 21 years in the school environment. Summation of the four subscales provides an adaptive behaviour composite score in the range of 0 to 223. The authors report good internal

consistency (83% of the sub domains' reliability at .85 or higher and only 6% below .8), high test-retest reliability of the adaptive behaviour composite (.91) and acceptable inter-rater reliability (ranging from mid .40s to high .60s).

Actiwatches (Cambridge Neuroscience, UK)

Actiwatches are actigraphy-based data loggers that record a digitally integrated measure of gross motor activity using a sensitive uniaxial accelerometer. The wrist worn device is traditionally used to measure sleep quality, although actiwatches have also been widely used in the measurement of general activity levels (e.g. Inoue *et al.,* 1998) by producing a digital integration of the amount and duration of all movement over .05g. An average activity score, measured in Hz, was calculated for each participant. The actiwatch used was formatted to start collecting data at the beginning of the child's school day and to continue to do so every fifteen seconds (or epochs) until the data were downloaded from the watch. The time the watch was put on and taken off the child was recorded by the researcher or teacher.

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 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. The second part of the interview assesses the severity of each form 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 (range = .5 to .8 and .7 to .91 respectively) and part 2 (range = .02 to .77 and .66 to .85 respectively).

5.3.2 Recruitment and participants

Participants were recruited from eleven schools for children with intellectual disabilities in the West Midlands. A composite risk marker for challenging behaviour was calculated through addition of the participants' scores on the repetitive, restricted, overactive and impulsive behaviour items of the CBSQ (for psychometrics see section 3.3.2). Potential composite scores ranged from 0 to 24. All of the participants (629) from a previous study (section 3.3.3.2) were ranked according to the composite score with the exception of any children who were deaf, blind, immobile or rated as having "full sentence speech" on the Wessex speech item. Immobile, blind and deaf children were excluded as they were considered more likely to receive low scores on the impulsivity and overactivity items and thus would be labelled 'low risk' as a result of these physical difficulties. Excluding these children would therefore allow accurate comparability between groups, particularly with regard to the actiwatch data. High verbal ability was included as an exclusion criterion as it was deemed indicative of a relatively higher level of ability and thus potential reactivity to observation by the researcher.

The parents/guardians of the thirty five highest ranking children on the composite scores were contacted to participate in the study. From this sample, the highest ranking fifteen children for whom parental consent was gained formed the high risk group. The low risk group comprised of the fifteen lowest ranking children for whom

parental consent was provided and who best matched the high risk participants. Participants were initially matched for ability using a disability percentile score based on either three items from the Wessex Behaviour Scale (Kushlick, Blunden & Cox, 1973) for participants aged 6 years and over or twenty from the Denver Developmental Screening Test II (DDST II; Frankenburg, Dodds, Archer, Shapiro, & Bresnick, 1992) for participants less than 6 years of age. Two different measures of ability were used due to the large age range of participants (5 to 14 years). Disability percentile scores for each measure were calculated to allow comparison of scores across measures. Therefore, matching was based on age and level of intellectual disability percentile score, so that a low risk child was considered to match a high risk child if they were aged within two chronological years of the high risk child and had the closest percentile disability score. The final sample comprised 30 children, 15 in each group, aged between 5 and 14 years (see Table 5.1 for demographic information).

Table 5.1: Demographic information for the high and low risk group (median and inter-quartile ranges)

Demographic information	High risk	Low risk
Mean age	9.28 (2.58)	10 (2.39)
Mean percentile severity of intellectual disability	61.51 (20.13)	66.43 (19.95)
Male	12 (40%)	9 (30%)
Diagnosis of 'probable ASD'	10 (33%)	7 (23%)
Other diagnoses	1 DiGeorge Syndrome, 3 Cerebral Palsy	3 Down Syndrome, 1 Cerebral Palsy, Trisomy X, 1 translocation of chromosome 4 onto 6 (partial trisomy 4q)

% - Proportion of total sample, 'probable ASD' based on teacher report

Statistical analysis showed that the 25 children for whom consent was not obtained did not differ from the final participants in terms of age (t (28) = .17, $p > .05$), gender (χ^2 (1, $N = 55$) = .96, $p > .05$), level of intellectual disability (χ^2 (1, $N = 55$) = 1.73, $p > .05$) or presence of any form of challenging behaviour (χ^2 (1, $N = 55$) = .25, $p > .05$).

The median composite risk scores (summation of repetitive and restricted behaviours and interests and overactivity/impulsivity) for the high and low risk participants were 21 (range = 19-24, IQR = 4) and 0 (range = 0-1, IQR = 0) respectively, with the high risk group scoring significantly higher (U = 0, $p < .001$). A chi squared test also showed that the groups did not significantly differ on gender (χ^2 (1, $N = 30$) = 1.43, $p > .05$). The difference between the mean age of the high (9.277, SD = 2.576) and low risk (10, SD = 2.39) groups also failed to reach significance (t (28) = -.81, $p > .05$). A series of t-tests demonstrated that there were also no significant differences between the groups on level of intellectual disability as measured by the VABS total raw score (communication, daily living and social skills subscales combined) communication, or daily living skills subscale (Appendix F). However, the low risk group (mean = 61.2, SD = 8.16) did score significantly higher than the high risk group (mean = 51.53, SD = 10.47) on the socialisation subscale (t = (1) -2.82, p <.01). These results indicate that the high and low risk groups did not significantly differ with regard to gender, age or level of intellectual disability. However, this is not to say that they are significantly similar as the tests used are not specifically for this purpose and thus are less sensitive to similarity than difference.

5.3.3 Procedure

Once parental consent was received, the children's' teachers were contacted in order to arrange a convenient time to observe the children. Naturalistic observations were conducted with all participants at their school during one whole school day. The majority (22; 73%) of the natural observations were video recorded and subsequently coded in real time using Obswin software (Martin, Oliver & Hall, 2000). Four children from each group were coded in-situ using EEE PC laptops due to difficulties gaining consent to video these children and/or several other children in their class. Between 4.5 and 6 hours of naturalistic observations were conducted for each child, during all break and meal times, free play and lessons. Observers remained as unobtrusive as possible throughout the observations. Each participant was also fitted with an actiwatch at the beginning of the natural observations which was later removed at the end of the school day (or earlier if the child showed obvious distress). For example, if a child communicated active dissent to wearing the actiwatch, or continuously attempted to remove the actiwatch after the initial hour of first wearing it, the actiwatch was removed.

Immediately following each visit, participants' teachers were sent a questionnaire pack, including the AQ, RBQ, SCQ and VABS and asked to complete this within one month of receiving it. Once this had been completed and returned, teachers then completed the CBI with a researcher trained in its administration over the telephone.

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

5.3.4 Data collection

Child behaviours coded included; challenging, repetitive, restricted and communicative behaviour, attention and movement. Verbal and physical contact from adults and broader situational variables were also coded. An operational definition for each behaviour can be seen in Table 5.2.

Table 5.2: Operational definitions of behaviours coded during natural observations

Behavioural class	Behaviour	Operational definition
Movement	**Slow gross motor activity**	Any slow (defined as at walking speed) gross motor movement involving movement at shoulder or hip, whole arm or whole leg
	Rapid gross motor activity	Any fast (defined as faster than walking speed) gross motor movement involving movement at shoulder or hip, whole arm or whole leg e.g. run, jump, bounce, arm wave
	Fidgeting	Purposeless restless non repetitive or rhythmical motor movements
Repetitive	**Contact**	Repetitive movements that involve contact with the body e.g. chin tapping, hand wringing, body posturing
	Non contact	Repetitive movements that do not involve contact with the body e.g. hand flapping, body rocking and spinning
	Restricted	Excessive tidying/cleaning, rituals, organising/arranging objects, completing tasks, hoarding
Challenging	**Self-injury**	Non-accidental contact behaviours that may result in tissue damage. Includes hand biting, head slapping and head banging.
	Aggression	Physical aggression directed towards another person e.g. hitting, kicking, grabbing and scratching
	Destruction	e.g. Tearing items, tipping furniture, pulling items off walls, throwing/swiping items away
Proto-imperatives	**Positive affect**	Positive vocalisation eg. laughing or positive facial expression eg. Smiling
	Negative affect	Negative vocalisation eg. crying or negative facial expression eg. Frowning
	Protest	Resistance of physical prompts, e.g. pushing item or person's hand away, going limp, trying to move away
	Approach	Attempt to engage adult e.g. walk towards, tug at clothing, attend to adult, speak to adult
Attention	**Sustained attention**	Attending to the same object or task
Situation (natural observations)	**Undivided attention**	Direct attention from adult to child with no other child present
	Shared attention	Group setting with adult engaging in activity with two or more children
	Low attention	No structured activity, free to play without instruction or expectation from staff
Adult	**Vocalisation/touch**	A vocalisation or touch which is not an imperative
	Demand	Any verbal or physical prompt or instruction that is not a denial or reprimand
	Denial	Any verbal request to stop or surrender materials or physical prevention of a response or removal of an item
	Challenging behaviour response	Adult stops or restrains challenging behaviour or tells child to stop the behaviour

Inter-observer reliability using 5 second time intervals was calculated for approximately 15% of all video recorded sessions. Mean Kappa values for the naturalistic observations were .72 (range 0.45 – 0.97) illustrating good agreement (Fleiss, 1981).

Data were downloaded from the actiwatches after each visit and edited so that only the actiwatch data between the times the teacher or researcher had reported the actiwatch to be put on and taken off were collected. If there were four consecutive scores of 0 (corresponding to four 15 second epochs) around this point, the time closest to the last or first 0 was used as a cut off point for the beginning or end of data collection respectively (a well established procedure in the literature e.g. Angulo-Barroso, Burghardt, Lloyd & Ulrich, 2008). Twenty six of the participants (two from each group showed active dissent) wore actiwatches for the duration of the natural observations (average time = 5 hours seven minutes, SD = 58.14 minutes).

5.3.5 Data analysis

Due to the skewed nature of the data, non parametric tests were used for the majority of the analyses. Bonferonni corrections were applied where appropriate in order to control for the large amount of statistical analysis conducted and the corrected Alpha values are indicated below each table.

5.4 Results

5.4.1 The validity of the CBSQ and the presence of the behavioural risk markers in the low and high risk groups

To determine the validity of the CBSQ, the percentage duration of all behaviours relating to the behavioural risk markers was calculated for each group (see Table 5.3).

Table 5.3: Percentage median, inter-quartile range and Mann Whitney U analysis for the duration of the risk markers demonstrated across natural observations

Risk marker	Behaviour	High risk	Low risk	U	*p* (1 tailed)
Overactivity	**Slow**	30.33 (21.69)	24.21 (15.52)	74	.058
	Rapid	9.65 (6.05)	3.44 (3.85)	37	**.001**
	Fidget	3.77 (5.16)	2.52 (4)	84	.125
	Total	51.48 (17.12)	33.2 (10)	28	**< .001**
Repetitive behaviour	**Contact**	10.09 (14.62)	.02 (.79)	15	**< .001**
	Non contact	2.99 (20.05)	.28 (1.02)	44.5	**.002**
	Total	15.37 (25.89)	.85 (2.41)	17	**< .001**
Restricted behaviour	**e.g. hoarding, rituals**	.01 (.68)	.0 (.25)	91	.193

Bonferroni corrections – movement = .0125, stereotypy = .025, bold = significant

Table 5.3 shows that, in accordance with the hypotheses made, the high risk children showed significantly greater duration of *total* and *rapid movement*, reflecting greater overactivity as compared to the low risk group. Similarly, high risk children also showed significantly greater levels of both *contact* and *non contact repetitive behaviours*. However, in contrast to the hypotheses made, there was no significant difference between groups for duration of *restricted* behaviour.

The complex nature of impulsivity as a construct created difficulties in developing a single code to accurately measure it. Thus, in order to compare levels of impulsivity between groups, the reaction of the participants to denials using lag sequential analysis was compared. This analysis compared the conditional probability of a correlate of impulsivity, the mean of negative affect and protest given a denial, with the unconditional probability of the composite. It was expected that the high risk group would have a higher conditional probability of this correlate given a denial than the low risk group. Figure 5.1 shows that, as would be expected, whilst the difference between the groups for both the conditional (U = 72, $p < .05$, 1 tailed) and unconditional (U = 55, $p < .008$, 1 tailed) probabilities was significant, the difference between the unconditional and conditional probability within groups was only significant for the high risk participants (Z = - 2.727, $p < .008$, as opposed to Z = - 1.274, $p > .05$ for the low risk).

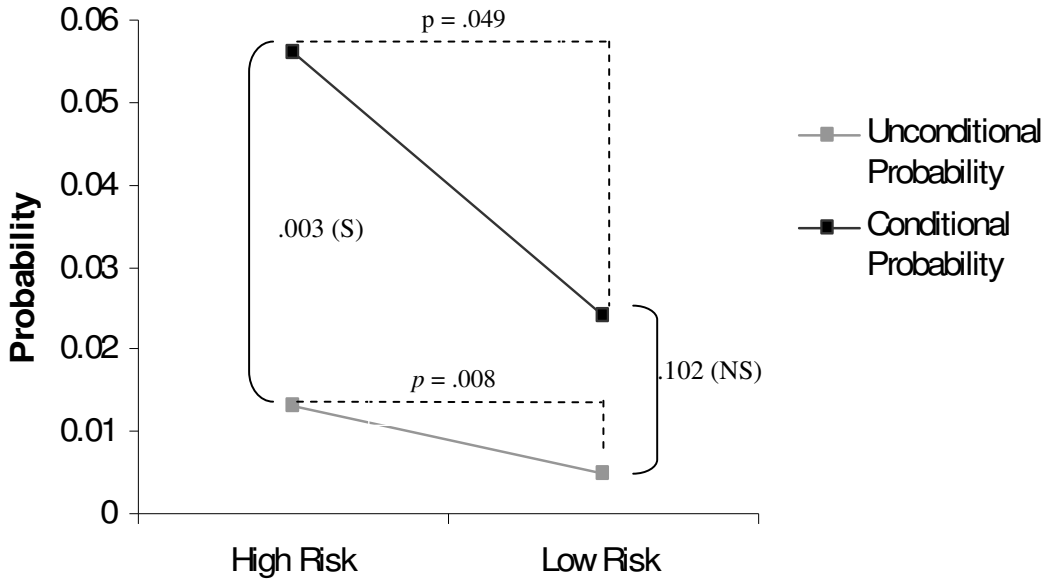


Figure 5.1: The mean unconditional and conditional probability of the impulsivity composite given denial for both high and low risk groups

The results of this analysis show that only the high risk group demonstrated significantly greater negative affect and protests in the presence of a denial, therefore indicating that the high risk group showed a higher level of a correlate of impulsive behaviour, as predicted. Whilst this measure is not a direct assessment of impulsivity, it is a likely correlate and thus indicates the increased rate of impulsive like behaviour in the high risk group. Combining these results with those of Table 5.3, which demonstrated that the high risk group showed significantly longer movement and repetitive movement, it appears that the screen is an accurate measure of the behavioural risk markers repetitive, overactive and impulsive behaviour.

5.4.2 The convergent validity of the CBSQ with other measures of the behavioural risk markers

In order to examine the convergent validity of the screen, the data gained from particular subscales of the teacher completed questionnaires were analysed and the group medians compared, as demonstrated in Table 5.4.

Table 5.4: Median, inter-quartile range and Mann Whitney U analysis for TAQ and RBQ scores for the high and low risk groups

Questionnaires	Subscale	High risk	Low risk	U	*p* (1 tailed)
TAQ	Impulsivity	15 (10)	1 (2)	66.5	.028
	Overactivity	19 (8)	3 (11)	33	**.001**
RBQ	Stereotyped behaviour	9 (7)	.0 (5)	49	**.003**
	Compulsive behaviour	7 (13)	.0 (3)	61.5	**.017**
	Insistence sameness	.0 (5)	.0 (5)	95	.486

Bonferonni corrections – TAQ - .025, RBQ - .017, bold = significant.

As shown in Table 5.4, the high risk group scored significantly higher on the *overactivity* subscale of the TAQ whilst group differences on the *impulsivity* subscale approached significance. Similarly, the high risk group scored significantly higher on the *stereotyped behaviour* and *compulsive behaviour* subscales of the RBQ, detecting some significant differences between the groups in terms of restricted behaviour which could not be identified in the natural observations.

Differences between the remaining RBQ subscales (*restricted preference and repetitive use of language*) and the three SCQ subscales could not be calculated due to the large number of non verbal participants in each group (10 high risk and 3 low risk). However, total SCQ scores for both the verbal and non verbal participants were calculated and compared to the clinical cut off scores suggested by Rutter *et al*. (2003). Chi square analysis showed that significantly more of the high risk (fourteen) than low risk (six) participants scored 15 or above (χ^2 (1, N = 30) = 9.6, $p < .01$) on the SCQ indicating significantly more participants in the high risk group have a cut off score indicative of an autistic spectrum disorder. A score of 22 or above, the cut off point for a diagnosis of autism, was also more frequent in the high (seven) than low risk group (four), although this difference was not significantly different (χ^2 (1, N = 30) = 1.29, $p > .05$). Additionally, whilst differences on the SCQ subscales could not be analysed due to the number of participants with no speech, mean scores for each subscale were calculated, with the high risk group scoring higher on each one (Appendix G).

Average activity as recorded by the actiwatches (Hz) for each group was also analysed and compared using a t-test. Analysis of these data showed that compared to

the low risk (mean = 180.02, SD = 64.49), the high risk group (mean = 262.52, SD = 75.52) had significantly greatly average activity levels (Hz) (t (1) = 3, $p < .05$). A spearman's rho correlation also indicated that the overactivity subscale of the TAQ was significantly associated with the participants' average activity level (Hz) as measured by the actiwatch (r (26) = .56, $p < .001$, 1 tailed) and the duration of rapid movement coded during the natural observations (r (26) = .55, $p < .001$).

In combination, these analyses provided good support for the convergent validity of the screen. As discussed, the high risk group scored significantly higher on three of the five questionnaire subscales analysed than the low risk group suggesting that they demonstrate the risk markers to a greater extent. The actiwatch data also provide objective support for the increased duration of movement as indicated by coding the natural observations.

5.4.3 Comparing the prevalence of challenging behaviour demonstrated by the high and low risk groups

In order to examine the prevalence of challenging behaviour, the percentage duration of challenging behaviour demonstrated by the high and low risk groups during the natural observations was compared using Mann Whitney U analysis, as illustrated in Table 5.5.

Table 5.5: Median, inter-quartile range and Mann Whitney U analysis for percentage duration of challenging behaviour for the high and low risk groups

Predictor variable	Behaviour	High risk	Low risk	U	*p* (1 tailed)
Challenging behaviour	**Aggression**	.19 (.42)	.01 (.18)	60.5	**.015**
	Destruction	.58 (1.67)	.01 (.09)	35.5	**.001**
	Self-injury	.29 (4.36)	.0 (.0)	43	**.002**
	Total	2.34 (12.91)	.04 (.37)	22.5	**< .001**

Bonferroni correction = .017, bold = significant

The results of this analysis demonstrated that the high risk group showed significantly more challenging behaviour, at each individual form and at a total level.

Data from the CBI was also examined in order to compare the prevalence of challenging behaviour between the groups. These results showed that ten (66.7%) of the high risk participants showed at least one form of challenging behaviour, compared to only five of the low risk participants (33.3%). However, a chi squared test indicated that there was no significant difference between groups in terms of presence of challenging behaviour (χ^2 (1, N = 30), $p > .05$). Of the ten high risk participants showing challenging behaviour, six (40%) showed only one form whilst the remaining four showed two forms of challenging behaviour (26.7%). In contrast, the majority of the low risk participants showed no challenging behaviour (10, 66.7%). Of the remaining five, one participant showed one form (6.7%), two showed two forms (13.3%) and two showed three forms (13.3%). No significant difference between the number of forms demonstrated by the high (mean = .93, SD = .8) and low risk (mean = .73, SD = 1.16) groups was found (t (1) = .549, $p > .05$). Additionally, there was no significant difference in terms of severity of challenging behaviour (t

(28) = .17, $p > .05$) between the high (mean = 15.33, SD = 13.17) and low risk (mean = 14.2, SD = 22.72) groups. However, a significant positive association between presence of challenging behaviour as measured by the CBI and as coded from the natural observations (Kappa = .33, $p = .014$) was identified. Therefore, whilst evidence for the predictive validity of the CBSQ was only provided by the observational data, associations between these data and the CBI did exist.

5.4.4 Comparison of the proto-imperatives demonstrated, environmental variables experienced by and the relationship between these constructs in the high and low risk groups

Having established the validity of the CBSQ, differences between the high and low risk groups in terms of child characteristics not included in the CBSQ and environment experienced were investigated to provide a more detailed examination of the high and low risk participants.

5.4.4.1 Environmental differences between the high and low risk groups

In order to examine the potentially different environment experienced by each group on both a macro and micro environmental level, the duration of various associated variables experienced by the high and low risk groups was calculated (see Table 5.6).

Table 5.6: Median, inter-quartile range and Mann Whitney U analysis for the percentage duration of environmental variables for the high and low risk groups

Class	Variable	High risk	Low risk	U	*P*
Macro environment	**Undivided attention**	21.19 (18.59)	10.86 (12.42)	53	**.013**
	Shared attention	25.2 (28.21)	32.84 (23.46)	89	.345
	Low attention	47.4 (20.95)	54.86 (13.69)	82	.217
Micro environment	**Vocalisation/touch**	12.03 (9.61)	6.44 (6.4)	51.5	**.01**
	Challenging behaviour response	.08 (.14)	.0 (.08)	65	.05
	Demand	7 (4.92)	6.48 (3.83)	89	.345
	Denial	.89 (1.67)	.23 (.57)	41	**.002**

Bonferroni corrections - macro = .017, micro = .0125

Analysis of the situational variables demonstrated that the high risk group received almost twice as much undivided attention from teachers and that this difference was significantly greater, although there were no significant differences between the amount of shared and low attention the participants received. The amount of adult interaction the participants received within the environment was also significantly greater for the high risk group, who received significantly more adult vocalisations and touch and denials. Increased adult vocalisation and touch would be expected considering the higher amount of undivided attention received by the high risk participants; however, significantly higher rates of denials suggest that the high risk group required greater levels of adult behaviour management. The small duration and non significant difference in the duration of challenging behaviour responses received by the two groups also suggests that the higher rate of undivided attention was not merely a result of greater response to challenging behaviour.

5.4.4.2 Child characteristic differences between the high and low risk groups

Differences between the high and low risk group in terms of the duration of child affect, proto-imperatives and attention span were also investigated (see Table 5.7).

Table 5.7: Median, inter-quartile range and Mann Whitney U analysis for the percentage duration of child characteristic variables for the high and low risk groups

Class	Variable	High risk	Low risk	U	*P*
Affect	**Negative**	.35 (.83)	.04 (.4)	65	.05
	Positive	2.67 (5.61)	3.77 (3.04)	90	.367
	Neutral	.99 (1.74)	1.07 (1.25)	102	.683
Proto-imperatives	**Protest**	.36 (1.06)	.01 (.07)	38.5	**.001**
	Approach	3.33 (5.91)	2.59 (4.84)	82	.217
Attention span	**Sustained attention**	16.18 (18.12)	25.74 (12.44)	52	**.005**

Bonferroni corrections - affect = .017, proto-imperatives = .017, attention span = .05

Table 5.7 shows that the high risk children protested significantly more and showed higher levels of negative affect, although this only approached significance. These results suggest that this group required higher levels of adult attention and appeared to react more adversely to this than the low risk group. The low risk group also showed significantly more sustained attention, suggesting that they have a greater attention span.

5.5 Discussion

The Challenging Behaviour Screening Questionnaire (CBSQ) was created to provide an assessment of previously examined risk markers in an accessible format. The

primary aim of this chapter was to test the validity of the CBSQ in terms of concurrent and convergent validity. Additionally, differences in child characteristics and environment experienced by the high and low risk groups were investigated in order to provide a more detailed description of the groups.

Investigating these differences within the school environment afforded the study high ecological validity. Operationally defining the variables to be coded also meant that the coding of these data was highly reliable. By carefully matching participants so that there were no significant differences between the groups in terms of age, level of intellectual disability or gender, increased the likelihood that the results gained were a consequence of the large difference in composite risk between the groups and not confounding variables (although the tests used to establish matching were more sensitive to difference than similarity and so the groups were only assessed to be not significantly different, not significantly similar). Interestingly though, the high risk group did score significantly lower on the socialisation subscale of the VABS, indicating that these children demonstrate more autistic like traits, an established correlate of challenging behaviour (e.g. McClintock, Hall & Oliver, 2003). Supporting this supposition a recent study also found that participants with autism scored significantly lower on the socialisation subscale of the VABS than children with an intellectual disability but without autism (Perry, Flanagan, Dunn Geier & Freeman, 2009).

Supporting the concurrent validity of the CBSQ, the high risk group demonstrated significantly greater durations of overactive, (total and rapid movement in particular), repetitive (both contact and non contact stereotypies) and impulsive like behaviour

(negative reactions to denials). Whilst there were no significant differences between the restricted behaviour demonstrated by the groups, this was not entirely unexpected given the difficulties in observing this type of behaviour in a relatively short time window. The significantly longer challenging behaviour demonstrated by the high risk group, at a total and specific form level, also supported the labelling of these participants as being at high or low risk.

The convergent validity of the CBSQ also appeared to be good, with the high risk group scoring significantly higher on the TAQ overactivity subscale and the RBQ stereotyped and compulsive behaviour subscales. This is particularly important given the sparse number of items from these questionnaires included in the CBSQ. The high risk group also demonstrated significantly more challenging behaviour during the natural observations, although it must be noted that there were no significant differences in the presence or severity of challenging behaviour demonstrated by the high or low risk groups according to the CBI. However, there was a significant positive association between presence of challenging behaviour as measured by the CBI and natural observation coding. This indicates that there are similarities in the results gained from these two measures of challenging behaviour, but that perhaps the CBI is less sensitive. Whilst the natural observations were made over the course of only one day and the CBI based on a far longer time period, it is plausible that the researcher coding the natural observations noticed challenging behaviour which a teacher might not. For example, according to the results of the CBI, participant H4 was reported not to demonstrate any form of challenging behaviour. However, the participant clearly had calloused skin covering both hands, apparently from biting. Whilst this evidence is purely anecdotal, the rich nature of data provided from natural

observations means that behaviours might be identified which teachers responsible for the care of up to ten children might fail to notice.

Interestingly, the high risk group experienced a greater duration of undivided attention and this appeared to comprise of more adult vocalisation and touch and denials. In view of the fact that no significant differences were found between the groups in terms of duration of demands, it appears that the higher duration of adult attention the high risk children received is likely to be due to the behaviour management required by this group, rather than teachers demanding more of the high risk children. This inference is supported by the results of a study by Carr, Taylor & Robinson (1991) which illustrated that teaching staff provided less demands to children who were likely to react adversely to them so that teachers behave to avoid the antecedent, demonstrating that the behavioural characteristics shown by children directly affect teachers' behaviour. These findings are also in line with Oliver's (1993, 1995) mutual reinforcement paradigm so that a child reacts adversely to a demand by screaming and becoming aggressive. The adult finds this behaviour aversive and thus responds by removing the denial which in turn results in a cessation of the child's behaviour. Because the adult finds this rewarding, they are more likely to stop providing demands in the future. However, due to the need to ensure the child's safety, they are unable to remove denials.

Whilst comparing the child characteristics exhibited by the high risk children, it came to light that the high risk children also attended to tasks and objects for a significantly shorter duration. Bearing in mind the greater levels of impulsive like behaviour demonstrated by the high risk group, it might be suggested that the high risk children

exhibited behaviour consistent with a diagnosis of Attention Deficit Hyperactivity Disorder (ADHD). Similarly, the stereotyped behaviour demonstrated by the high risk children, along with the higher RBQ compulsion and SCQ cut off scores, could be indicative of a diagnosis of autism spectrum disorder. Whilst it is beyond the scope of this study to suggest links between diagnoses and behaviour on the basis of risk markers, it is possible that many of the behaviours illustrated by the high risk group are downstream from diagnoses of ADHD and ASD.

This is consistent with previous research which has already identified the presence of these diagnoses in children with a severe intellectual disability. Prevalence rates between 10 and 18% for ADHD (e.g. Jou, Handen & Harden, 2004; Epstein, Cullinan & Gadow, 1986) and up to 40% for ASD (eg. Bouras, Holt, Day & Dosen, 1999; Kraijer, 1997) have been reported in people with intellectual disabilities, illustrating the potential for the overactive, impulsive, repetitive and restricted behaviour demonstrated by the high risk participants to be related to these diagnoses. Whilst further investigation of this supposition is required to understand whether diagnosing children with risk markers for challenging behaviour with ASD and ADHD is appropriate, identifying associations between these diagnoses and the behaviour of children at high risk for challenging behaviour is important. Labelling high risk children with these disorders is unlikely to be helpful, but understanding similarities in the neurological underpinnings of such disorders and behavioural risk markers and examining the potential role of compromised behavioural inhibition (as described in section 3.5 and section 4.5), is of great use, particularly if early intervention strategies are to be successful. This issue is also particularly pertinent given the established association between these disorders and challenging behaviour (Ando & Yoshimura,

1979; McClintock, Hall & Oliver, 2003; Fischer, Barkley, Fletcher & Smallish, 1993).

The relatively small sample from which the results of this study were obtained and the many significant differences identified indicate the robust nature of these findings. Detecting these differences within this sample size was also imperative if the CBSQ is to be accurately applied within clinical populations. However, including just 30 participants does imply difficulties in generalising the findings of this study to the general population of children with intellectual disabilities. Failure to detect a significant difference in the restricted behaviour demonstrated by the two groups could also be a result of the sample size.

Despite the significant differences between the high and low risk groups in terms of the percentage duration of the risk marker variables, there was also large heterogeneity within the groups. Thus, whilst the CBSQ appears to have robust validity, false positives and negatives might be detected through its use and further research is required to investigate why this might be. This validity can also only be assumed for the less able and mobile children with good vision and hearing as participants without these traits were excluded from the study. This was deemed necessary in order to avoid the risk of greater reactivity to observation by the researcher and inaccurate group comparisons which could threaten the validity of the study.

Every effort was made to operationally define the variables to be coded as tightly as possible for the risk marker constructs, although this was particularly difficult for

impulsivity. Thus, the impulsivity composite formed (combined negative affect and protest associated with denials) is only a close correlate of impulsivity. However, in addition to the higher duration of attention demonstrated by the high risk group, the supposition of ADHD like behaviours in this group is supported and in turn, the impulsivity composite is given weight as a measure of impulsivity in this population.

The data provide good support for the validity of the CBSQ on multiple levels. This is clinically significant as it indicates the potential of the CBSQ as the basis of future early intervention research. The significant differences found between the groups in terms of adult attention received and reactions to demands and denials also indicates that the high risk group are not merely challenging in terms of the various forms of challenging behaviour, but demonstrate generally difficult behaviour which requires greater resources. When the number of children demonstrating these risk markers is considered, this highlights the difficulties faced by teachers of these children and the education system as a whole, which must not be underestimated.

5.6 Summary and implications

By comparing the presence of the behavioural risk markers in children identified as being at high or low risk using natural observations, questionnaires and objective measures, evidence was provided for the concurrent and convergent validity of the CBSQ. High risk children also demonstrated more challenging behaviour during the natural observations, supporting the findings reported in Chapters 3 and 4 and the significance of the behavioural risk markers for this population. Comparison of the high and low risk groups in terms of child characteristics demonstrated and environmental variables experienced also indicated significant differences, so that the

high risk group appeared to require significantly more adult attention and demonstrated more characteristics in line with diagnoses of ADHD and autism.

Thus, on a theoretical level, these findings provide further support for the potential role of compromised behavioural inhibition for challenging behaviour in individuals with intellectual disabilities. Clinically, these findings are also significant as by indicating the validity of the CBSQ, they also provide support for the use of the CBSQ within early intervention programmes. Also on a practical level, these findings suggest the huge strain put upon those working with children at high risk of challenging behaviour, not merely in terms of increased resources required, but also with regard to the assumed stress associated with challenging and generally difficult behaviour.

The final empirical chapter of this thesis provides a further, more detailed examination of children at high risk. Using experimental functional and descriptive analyses, Chapter 6 investigates the potential function of the challenging behaviour demonstrated by this group, as well as the communicative repertoire available to them in order to assess the possible success of established interventions such as functional communication training for this population.

Chapter 6

Children with Severe Intellectual Disabilities at High or Low Risk for Challenging Behaviour: Differences in Challenging, Proto-Imperative and Affect Behaviours

6.1 Abstract

Aims: The primary aim of this study was to investigate the challenging, proto-imperative and affect behaviours demonstrated by children who show characteristics that predict the presence of challenging behaviour.

Method: Experimental functional and descriptive analyses were used in order to determine a potential function and communicative nature of the challenging behaviour demonstrated by these participants.

Results: The results of this study indicated that all of the high risk participants showing challenging behaviour fifteen times or more during experimental functional or descriptive analyses, demonstrated more behaviour during a particular experimental condition or when a setting event or adult behaviour occurred, indicating potentially functional behaviour. In both the experimental functional and descriptive analyses, only one participant failed to show a significant association between a proto-imperative and challenging behaviour, indicating a close association between communicative and challenging behaviour for the majority of participants. High risk participants also demonstrated significantly more negative affect during the low attention and protest during the high attention and demand experimental functional analysis condition, indicating that they found these situations more aversive. **Discussion**: These results suggest that despite presenting behavioural characteristics

associated with challenging behaviour, the environment continues to have a significant effect on the behaviour demonstrated by high risk children. This high risk group also appear to have a repertoire of communicative behaviours which they can employ appropriately, so that high risk individuals are potentially suitable for early intervention programmes using functional communication training.

6.2 Introduction

The high prevalence of challenging behaviour, in combination with increased rates of this behaviour with age (until early to mid-adulthood) and marked persistence once established in the repertoire of people with intellectual disabilities, allude to the potential for effective early intervention for this population (Borthwick-Duffy, 1994; Emerson *et al*., 2001b; Eyman & Call, 1977; Harris, 1993; Lowe *et al*., 2007; Qureshi & Alborz, 1992; Schroeder, Schroeder, Smith & Dalldorf, 1978). For such a programme to be efficient, the identification of children with intellectual disabilities who are most likely to develop challenging behaviour in the future would be beneficial. A wealth of research investigating child characteristics associated with challenging behaviour in children with intellectual disabilities has now identified several child and behavioural characteristics which predict the presence of challenging behaviour contemporaneously (e.g. Chadwick, Piroth, Walker, Bernard & Taylor, 2000; Baghdadli, Pascal, Grisis & Aussilloux, 2003; McClintock, Hall & Oliver, 2003, Oliver, Sloneem, Hall & Arron, 2009; Oliver *et al.*, *in preparation,* Petty *el al., in preparation*) (section 3.2).

Behavioural characteristics (i.e. repetitive, restricted, impulsive and overactive behaviour) are particularly significant for children with intellectual disabilities because as well as being associated with challenging behaviour, they typically predate

its presence at a population level. Several studies investigating the prevalence of challenging behaviour in individuals with intellectual disabilities have illustrated acceleration in the proportions showing the behaviour and the severity of behaviour between the teenage years and mid to late 20's. For example, whilst a number of participants in a total population study conducted by Oliver, Murphy and Corbett (1987) showed self-injurious behaviour aged as young as ten years, the highest rates of this behaviour were in participants in adolescence and their 20's. These findings have been supported by other studies which have also investigated the relationship between age and the prevalence of self-injury, aggression and destruction (Kebbon & Windahl, 1986; Rojahn *et al.* 1993).

Research investigating the development of behavioural characteristics that predict challenging behaviour such as repetitive, restricted, overactive and impulsive behaviour, has shown an earlier age of development. By the age of 2 to 3 years, many children with intellectual disabilities are already demonstrating stereotyped behaviour (Lord, 1995; Berkson & Tupa, 2000) and interests (Young, Brewer & Pattison, 2003). It is also now widely recognised that autism can be reliably diagnosed in a substantial proportion of children before the age of three (Gillberg *et al.*, 1990; Stone *et al.*, 1999). Given the inclusion of "restricted repetitive and stereotyped patterns of behaviour, interests and activities" in the Diagnostic and Statistical Manual (DSM; APA, 1994) criteria for autism, it could be inferred that both repetitive and restricted behaviours, such as hoarding, develop prior to the age at which the proportion of people showing challenging behaviour rises dramatically. A study examining the age of onset of ADHD has also shown that over 80% of participants with ADHD who met symptom criteria had an age of onset of impairment less than 7 years (Applegate *et*

al., 1997). Many of these participants demonstrated symptoms of hyperactivity and impulsivity as young as 1 year of age, indicating that, like repetitive behaviours and interests, these behaviours develop earlier than the age at which the proportion of people showing challenging behaviour rises dramatically.

This evidence for the age difference in the emergence of the predictors of challenging behaviour and the challenging behaviour itself suggests that these characteristics might act as early risk markers. If these were identified in young children, they could indicate a level of risk for developing challenging behaviour and, more importantly, severe challenging behaviour, before the behaviour is evident or persistent. Identification of risk markers would make an early intervention strategy more efficient by associating resources with risk. Despite the lack of longitudinal evidence demonstrating that behavioural risk markers precede challenging behaviour, the child characteristics of repetitive, restricted, overactive and impulsive behaviour might usefully be considered to be risk markers as they are associated with the presence of, and appear to develop prior to, at least at a population level, the development of severe challenging behaviour.

The characteristics that predict the presence of challenging behaviour at a population level are inconsistent with an exclusively operant theory approach which emphasises selection and maintenance of behaviour by environmental consequences alone, regardless of child characteristics. This assertion is broadly supported by research utilising experimental and descriptive functional analyses, and an extensive and robust applied behaviour analytic intervention literature (Hall & Oliver, 1992; Iwata *et al.,* 1994, Scotti, Evans, Meyer & Walker, 1991). Theoretical models of the development

of self-injury, such as Guess and Carr's (1991) stage model impute operant processes as driving the emergence of more severe behaviour. There is also some evidence for this part of the model in longitudinal studies of early self-injury (Oliver, Hall & Murphy, 2005).

The social function of self-injurious behaviour, as well as the strong temporal association between communicative and challenging behaviours identified in a recent study (Petty, Allen & Oliver, 2009) provides further evidence for the importance of operant processes. Petty *et al.,* also report close temporal proximity between repetitive and self-injurious behaviour, indicating the emergence of self-injurious from repetitive behaviours, as initially proposed by Guess and Carr (1991). It is likely therefore that operant processes are influential in the development of self-injurious behaviour for some children.

To date the parallel literatures on child characteristics and operant processes have not been integrated. Integration would be beneficial because the child characteristics that might be considered as risk markers might indicate which children should be prioritised for preventative or early intervention, whilst operant approaches might indicate the nature of the intervention to be implemented.

As discussed previously (section 3.2), numerous studies have identified child characteristics associated with challenging behaviour. Supporting this, a short and accessible screening tool, the Challenging Behaviour Screening Questionnaire (CBSQ), which identifies the presence of these characteristics, has recently been developed (section 3.3.1). When piloted, the use of the CBSQ replicated previous

findings using more comprehensive measures, by identifying repetitive, restricted, overactive and impulsive behaviours as child characteristics associated with challenging behaviour in a cohort (section 3.4) and longitudinal study (section 4.4). Results of a related study provided evidence for the concurrent and convergent validity of the CBSQ (section 5.4.1), the use of which would enable the identification of children with characteristics that predict the presence of challenging behaviour. Now that children at risk of challenging behaviour can be successfully identified, the focus of research can shift to better understanding the influences on this behaviour and, in particular, the applicability of operant theory to these children's challenging behaviour.

There is broad acceptance of experimental functional analysis and descriptive analysis as methods that can evaluate the influence of environmental variables on behaviour and, by implication, operant learning. Additionally, evidence for challenging behaviour as functional can be gleaned from examining other child behaviours demonstrated at the same time as the challenging behaviour at greater than chance levels. For example, if a child reliably demonstrated challenging behaviour in the same time window as a proto-imperative communicative behaviour (e.g. dissent), this would imply that the challenging behaviour has a communicative function for the child.

Illustrating this, Petty *et al.* (2009) identified a strong temporal relationship between pragmatic communicative behaviours and challenging behaviours, whereby proto-imperative behaviours reliably preceded self-injury. These results have several important implications. Firstly, they provide a potential explanation for the transition

from repetitive to socially-maintained behaviour, vital to Guess and Carr's (1991) developmental model of self-injurious behaviour. Establishing the presence of proto-imperative communicative behaviours and their association with challenging behaviour is also useful for intervention purposes as it indicates that the child has in their repertoire a behaviour which can be shaped and is likely to be functionally equivalent and also identifies the point in a behavioural chain at which functional communication intervention should be implemented in order to provide an effective early intervention strategy. Identifying similar results in children with risk markers associated with challenging behaviour would indicate that such early intervention strategies would also be beneficial to those at high risk of challenging behaviour and thus those most in need of early intervention.

The primary aim of this study is to investigate the challenging behaviour demonstrated by children who show characteristics that predict the presence of challenging behaviour using experimental functional and descriptive analyses in order to determine a potential function of this behaviour. The presence of proto-imperative communicative behaviours in this group will also be examined in order to gain an understanding of the communicative repertoire of these participants. Finally, the association between these proto-imperative communicative and affect behaviours, specific environmental conditions and settings and challenging behaviours will be investigated to ascertain whether the challenging behaviour demonstrated has a communicative function.

To achieve these aims, the design incorporates comparisons of children with characteristics that are associated with challenging behaviour (the high risk group) and children who do not evidence these characteristics (the low risk group).

6.3 Method

6.3.1 Measures

6.3.1.1 Experimental functional analysis

All participants took part in an experimental functional analysis of their behaviour using experimental analogue conditions similar to those developed by Carr and Durand (1985) to assess the influence of social reinforcement and task demand on challenging behaviour. The conditions included;

A. **High attention** - The researcher maintained a high level of verbal and physical attention and remained within close proximity to the child. No demands were issued by the researchers and no response was made to any form of challenging behaviour. This control condition provides a basis for comparison for the low attention and demand conditions. This condition was analogous to the 'Easy 100' condition implemented by Carr and Durand (1985).

B. **Low attention** – The researcher maintained the same level of attention as condition A, but interacted with another adult as opposed to the child. All child behaviours were ignored apart from any form of challenging behaviour. At this point the researcher said "Don't do that" and then redirected attention back

towards the other adult. Greater demonstration of challenging behaviour in this condition as compared to the high attention and demand condition indicates behaviour occasioned by attention deprivation and maintained by contingent attention. This condition was analogous to the 'Easy 30' condition implemented by Carr and Durand (1985).

C. Demand – The researcher prompted the participant through a task chosen by their class teacher which was one less preferred by the child and thus likely to produce non-compliance and a need for adult encouragement. Every child was given a puzzle type task appropriate to their level of ability. The researcher used a hierarchy of prompts starting with verbal, then verbal and model (showing the child what to do) and finally verbal and physical (holding the child's hand whilst doing the task). Upon task completion, the child received verbal and physical (patting child's arm) praise. The researcher responded to any challenging behaviour by removing the task and stepping away from the child. If after ten seconds the child ceased to demonstrate this behaviour, the researcher reinstated physical proximity and the task. However, if the behaviour was maintained during the 10 second time out period, the researcher waited until the child had ceased demonstrating any challenging behaviour for 5 seconds before reinstating physical proximity and the task. A higher level of challenging behaviour in this condition as compared to the high and low attention condition indicates behaviour occasioned by an aversive task and maintained by contingent removal of that task.

Conditions were alternated in an ABAC ABAC ABAC ABAC design and each lasted 5 minutes with a brief interval between conditions. All experimental functional analysis was conducted in a quiet room in the child's school and was videotaped.

For two of the participants (H2 and L5), the experimental functional analysis had to be terminated before completion due to obvious distress. For participant H2, only two full repeats (ABAC ABAC) were completed, whilst all repeats were conducted with participant L5 but the 2nd, 3rd and 4th repeats were reduced to 1 minute per analogue condition.

6.3.1.1.1 Experimental functional analysis integrity

To ensure that the experimental functional analysis was administered to participants according to the methodology, the percentage duration of adult vocalisation/touch, challenging behaviour response and demand was compared between the three analogue conditions.

Providing evidence for the integrity of the experimental functional analysis methodology, Friedman analysis revealed significant differences between the three analogue conditions for adult vocalisation or touch (χ^2 (1, N = 30) = 60, p < .001) challenging behaviour response (χ^2 (1, N = 30) = 37.83, p < .001) and demand (χ^2 (1, N = 15) = 54.69, p < .001). Wilcoxon analysis also showed that these significant differences were in line with the methodology, so that there was significantly greater duration of adult vocalisation or touch during the high attention (median = 99.01, IQR = .1) than the low attention (median = 1.61, IQR = .85) (Z = -4.78, p < .001) and demand conditions (median = 5.49, 12.84) (Z = -4.78, p < .001), challenging

behaviour response during the low attention (median = .5, IQR = 3.03) than the high attention (median = 0, IQR = 0) (Z = -4.11, $p < .001$) and demand conditions (median = 0, IQR = 0) (Z = -3.88, $p < .001$) and demand during the demand (median = 88.14, IQR = 6.79) than the high (median = .02, IQR = .24) (Z = -4.78, $p < .001$) and low attention (median = 0, IQR = 0) (Z = -4.78, $p < .001$) conditions.

6.3.1.2 Naturalistic observations

Naturalistic observations were conducted with all participants at their school during one whole school day. The majority (n = 22; 73%) of the natural observations were video recorded and subsequently coded in real time using Obswin software (Martin, Oliver & Hall, 2000). Four children from each group were coded in-situ using EEE PC laptops due to difficulties gaining consent to video these children and/or several other children in their class. Between 4.5 and 6 hours of naturalistic observations were conducted for each child, during all break and meal times, free play and lessons. Observers remained as unobtrusive as possible throughout the observations.

Adult variables during the natural observations were coded on both a macro and micro analysis level. Macro analysis involved the general level of attention adults provided for the participants; undivided, shared and low attention. Micro analysis involved more fine grained adult behaviours; demand, denial and adult vocalisation/touch (see section 6.3.3 for a full list of coded behaviours and their operational definitions). These situational variables are far less controlled than the experimental functional analysis conditions and thus, although apparently similar to the high and low attention experimental functional conditions, they are not directly comparable. Illustrating this, challenging behaviour demonstrated during the low attention situation, as with the

experimental functional analysis condition, is taken to indicate behaviour occasioned by attention deprivation and maintained by contingent attention. However, challenging behaviour demonstrated during the high attention situation might indicate social and/or demand escape and cannot be used as a control condition due to the lack of control over the use of demands or denials and exact levels of adult vocalisation and touch during this condition. Challenging behaviour demonstrated in response to adult vocalisation or touch, denial or demand indicated that these variables acted as establishing operations for this challenging behaviour.

6.3.2 Participants and recruitment

Participants were recruited as the result of a previous screening questionnaire study which identified the children as being at high or low risk of challenging behaviour using a composite risk marker index (see Table 6.1 for basic demographic information, see section 5.3.2, for further information).

Table 6.1: Demographic information for the high and low risk groups (medians and inter-quartile ranges)

Demographic information	High risk	Low risk
Median risk score	21 (4)	0 (0)
Mean age	9.28 (2.58)	10 (2.39)
Mean percentile severity of intellectual disability	61.51 (20.13)	66.43 (19.95)
Male	12 (40%)	9 (30%)
Diagnosis of "probable ASD"	10 (33%)	7 (23%)
Other diagnoses	1 DiGeorge Syndrome, 3 Cerebral Palsy	3 Down Syndrome, 1 Cerebral Palsy, 1 Trisomy X, 1 translocation of chromosome 4 onto 6 (partial trisomy 4q)

The highest and lowest risk children for whom parental consent was obtained formed the high and low risk groups of this study.

6.3.3 Behaviours coded across analyses

Child behaviours coded included challenging, proto-imperative and affect behaviours. Verbal and physical contact from adults was also coded. Additionally, situational variables were coded for the natural observations (for operational definitions of each behaviour see Table 6.2).

Table 6.2: Operational definitions of behaviours coded across analyses

Behavioural class	Behaviour	Operational Definition
Challenging	**Self-injury**	Non-accidental contact behaviours that may result in tissue damage. Includes hand biting, head slapping and head banging.
	Aggression	Physical aggression directed towards another person e.g. hitting, kicking, grabbing and scratching
	Destruction	e.g. Tearing items, tipping furniture, pulling items off walls, throwing/swiping items away
Proto-imperatives	**Positive affect**	Positive vocalisation e.g. laughing or positive facial expression eg. smiling
	Negative affect	Negative vocalisation e.g. crying or negative facial expression eg. frowning
	Protest	Resistance of physical prompts e.g., pushing item or person's hand away, going limp, trying to move away
	Approach	Attempt to engage adult e.g. walk towards, tug at clothing, attend to adult, speak to adult
Adult	**Vocalisation/touch**	A vocalisation or touch which is not an imperative
	Demand	Any verbal or physical prompt or instruction that is not a denial or reprimand
	Denial	Any verbal request to stop or surrender materials or physical prevention of a response or removal of an item
	Challenging behaviour response	Adult stops or restrains challenging behaviour or tells child to stop the behaviour
Setting (natural observations)	**Undivided attention**	Direct attention from adult to child with no other child present
	Shared attention	Group setting with adult engaging in activity with two or more children
	Low attention	No structured activity, free to play without instruction or expectation from staff

Inter-observer reliability using 5 second time intervals was calculated for approximately 15% of all video recorded sessions. Mean Kappa values for the analogue sessions were .77 (range = 0.55 to 0.95) illustrating good agreement (Fleiss, 1981).

6.3.4 Procedure

Once parental consent had been received, the teacher of each participant was contacted in order to arrange a mutually convenient day to observe the child for a whole school day (section 5.3.3). Having completed the natural observation, a second date was arranged to complete the experimental functional analysis. This visit was arranged as close to the natural observation as possible. Completing the experimental functional analysis usually required most of the school day, including regular breaks.

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

6.3.5 Data analysis

Due to the skewed nature of the data, non parametric tests were used for the majority of the analyses. Bonferonni corrections were also applied where appropriate in order to avoid type 1 errors and Alpha values are indicated in parentheses under tables or within analyses. In order to determine the function of challenging behaviour demonstrated by the high risk group participants during the experimental functional analysis, multi-element graphs were constructed and Cliff's *d* statistic (Cliff, 1993) calculated for each form of challenging behaviour demonstrated by a participant fifteen times or more. By comparing the extent to which one sample distribution tends

to lie above another, ie. challenging behaviour in one analogue condition as compared to another, the *d* statistic provides a measure of effect size. A *d* value of 1.0 indicates that there is no overlap between the two sampling distributions. For the purpose of this study, a *d* statistic of +/-.33 or more was used as an arbitrary indicator of meaningful difference.

To analyse associations between various forms of challenging behaviour, proto-imperatives and affect, lag sequential analysis was conducted. This analysis produces a Yule's Q value which displays the likelihood (whether it be increased or decreased) of challenging behaviour given an environmental condition (similar to the experimental functional analysis) or an adult variable at lag 0. An arbitrary score of +/-.33 was used to indicate statistical significance as this equated to an odds ratio of 2, indicating that a behaviour is twice as likely to occur in the presence of another behaviour or environmental condition (Bakeman & Quera, 1995; Hall & Oliver, 1997). Occurrences of behaviour in 1-second intervals were combined into 5-second bins using a partial interval rationale, to lag the co-occurence of challenging and teacher's behaviour in the same 5-second bin.

Finally, in order to compare the proto-imperative and affect behaviours demonstrated by the high and low risk group, Mann Whitney U analysis was used to investigate significant between group differences and Friedman and Wilcoxon signed rank tests to detect differences in the behaviours demonstrated between analogue conditions within each group.

6.4. Results

6.4.1 Demographic Information

The median composite scores for the high and low risk children were 21 (range = 19 to 24, IQR = 4) and 0 (range = 0 to 1, IQR = 0) respectively, with the high risk group scoring significantly higher on the composite score (U = 0, $p < .001$). A chi squared test also showed that the groups did not significantly differ on gender (χ^2 (1, $N = 30$) = 1.43, $p > .05$). The difference between the mean age of the high (9.267, SD = 2.58) and low risk (10, SD = 2.39) groups was also not significant (t = -.81, (1), $p > .05$).

A series of t-tests demonstrated that there were also no significant differences between the groups on level of intellectual disability as measured by the VABS total raw score (communication, daily living and social skills subscales combined) communication, or daily living skills subscale (Appendix F). However, the low risk group (mean = 61.2, SD = 8.16) did score significantly higher than the high risk group (mean = 51.53, SD = 10.47) on the socialisation subscale (t = (1) -2.82, $p <.01$). Therefore, these results indicate that the high and low risk groups did not significantly differ with regard to gender, age or level of intellectual disability. However, this is not to say that they are significantly similar as the tests used are not specifically for this purpose and thus are less sensitive to similarity than difference.

6.4.2 The relationship between challenging behaviour and environmental variables in children at high risk for developing severe challenging behaviour

6.4.2.1 Duration of challenging behaviour

In order to test whether the high risk participants showed greater levels of challenging behaviours during experimental functional analysis, the median percentage duration of three forms of challenging behaviour for each group were compared across all analogue conditions. As predicted, the high risk group (median = 4.63, IQR = 8.85) demonstrated significantly longer total challenging behaviour (U = 23, $p < .001$) than the low risk group (median = .12, IQR = 1.47). In particular, there were also significant differences between the two groups in terms of aggression (U = 19.5, $p < .001$) and destruction (U = 41, $p = .001$), but not self-injury (high risk median = .06, IQR = 4.4; low risk median = 0, IQR = .51; U = 83, $p > .05$) with the high risk group (aggression: median = 1.24, IQR = 1.49; destruction: median = .65, IQR = 1.83) demonstrating these two forms significantly longer than the low risk group (aggression: median = .06, IQR = .19; destruction: median = .0, IQR = .12).

6.4.2.2 The function of the challenging behaviour shown by the high risk group participants during experimental functional analysis

The function of challenging behaviour when demonstrated by each high risk participant fifteen times or more was illustrated using multi-element graphs (Appendix H) and investigated using Cliff's *d*. These results are reported in the left hand side of Table 6.3 and indicate that twelve of the high risk participants demonstrated one or more forms of challenging behaviour fifteen times or more and at least one form of challenging behaviour was functional for eleven of these children

(behaviour was labelled as functional if it was demonstrated at significantly higher duration in one condition than another i.e. equal to or above .33, the criteria for Cliff's *d* statistic).

Table 6.3: The association between experimental functional analysis conditions and challenging behaviour using Cliff's *d* statistic (left of the bold line) and challenging behaviour, adult attention and adult behaviours during natural observations using lag sequential and Yule's Q analysis (right of the bold line).

		Experimental functional analysis Cliff's *d* statistic		Natural observations Yule's Q					
Participant	**Behaviour**	**Low attention/ High attention**	**Demand/ High attention**	**Undivided attention**	**Shared attention**	**Low attention**	**Adult vocalisation touch**	**Demand**	**Denial**
H1	Aggression	nc	nc	+	0	n/a	n/a	+	n/a
	Destruction	nc	nc	n/a	0	+	-	-	+
H2	Aggression	+	**+**	+	0	-	-	0	+
	Self-injury	nc	nc	-	-	+	0	-	-
H3	Aggression	0	-	**+**	0	0	0	0	**+**
	Destruction	nc	nc	0	n/a	**+**	0	n/a	0
H4	Destruction	0	+	nc	nc	nc	nc	nc	nc
	Self-injury	-	-	**-**	0	**+**	**-**	**-**	0
H5	Aggression	+	0	nc	nc	nc	nc	nc	nc
	Destruction	0	0	nc	nc	nc	nc	nc	nc
	Self-injury	0	-	**-**	0	0	0	**-**	**-**
H6	Aggression	-	-	nc	nc	nc	nc	nc	nc
	Destruction	nc	nc	**-**	**+**	0	**-**	0	0
H7	Aggression	0	0	**+**	**-**	0	**+**	n/a	n/a
	Destruction	0	0	**-**	0	0	0	**-**	0
	Self-injury	**-**	-	**-**	**+**	0	**-**	n/a	n/a
H8	Aggression	**-**	-	**+**	0	**-**	**+**	**+**	**+**
H9	Destruction	nc	nc	n/a	**-**	**+**	n/a	n/a	n/a
	Self-injury	nc	nc	**-**	0	**+**	n/a	n/a	n/a
H10	Destruction	nc	nc	**+**	**+**	**-**	**-**	0	**+**
H11	Aggression	+	+	nc	nc	nc	nc	nc	nc
H12	Aggression	-	-	nc	nc	nc	nc	nc	nc
	Destruction	nc	nc	**+**	**-**	**-**	0	**-**	**+**
	Self-injury	0	0	**-**	**+**	0	0	**-**	**-**
H13	Self-injury	0	0	**-**	n/a	**+**	**-**	**-**	n/a
H14	Destruction	0	+	**-**	0	**+**	0	**-**	**-**
	Self-injury	nc	nc	0	**+**	0	0	n/a	n/a
H15	Destruction	0	-	nc	nc	nc	nc	nc	nc

Cliff's *d* statistic: + = .33 or above, - = -.33 or below, 0 = less than + or -.33; Yules Q: + = .3 or above, - = -.3 or below, 0 = less than + or -.3,
n/a = no conditional probability, nc = challenging behaviour not reaching criteria

The results in Table 6.3 demonstrated that three participants showed significantly more aggression in the low than the high attention condition indicating that for these participants, this behaviour was attention maintained. Conversely, three participants showed significantly more aggression and two participants showed significantly more self-injury in the high than low attention condition, which is not consistent with traditional experimental functional analysis which utilises the high attention condition as a control condition. Destruction however appeared to be demonstrated randomly across the analogue conditions, with no participants showing significantly more destruction in the high or low attention condition. A further two participants showed no difference in the demonstration of a form of challenging behaviour between the high or low attention analogue conditions, indicating no attention maintained function of these behaviours.

Comparing rates of challenging behaviour in the high attention and demand condition, four participants showed significantly higher rates of challenging behaviour in the latter (two aggression, two destruction), indicating behaviour which has previously been reinforced by demand escape. Conversely, eight participants showed significantly higher amounts of challenging behaviour in the high attention than the demand condition (4 aggression, 3 self-injury and 1 destruction), which is not consistent with traditional experimental functional analysis which utilises the high attention condition as a control condition. Only one participant showed challenging behaviour whereby none of the forms were more prevalent in the high attention or demand condition.

These results show that for the majority of participants (eleven out of twelve), at least one of the forms of challenging behaviour demonstrated was exhibited significantly

longer during one analogue condition than another (six aggression, three destruction and three self-injury). This suggests a function for that behaviour during the experimental functional analysis; whether it be attention maintained behaviour or behaviour which is reinforced by demand escape. Seven children also demonstrated behaviour which was multi-functional, as illustrated by participant H11, who showed significantly more aggression during the low attention and demand than the high attention condition, indicating behaviour maintained by both attention and demand escape.

6.4.2.3 The function of the challenging behaviour shown by the high risk group participants during natural observations

In order to investigate the function of challenging behaviour demonstrated by the high risk participants during the natural observations, lag sequential analysis was conducted using data from the thirteen high risk participants who demonstrated fifteen or more incidents of a form of challenging behaviour (see Table 6.3).

Results of the lag analysis demonstrated in Table 6.3 showed that of these thirteen participants, each showed at least one functional form of challenging behaviour. For eight participants challenging behaviour was more prevalent during low attention, indicating that the behaviour functioned in order to gain social attention. Seven participants also demonstrated more challenging behaviour during undivided attention, indicating challenging behaviour maintained by social and/or demand escape. Six participants also demonstrated more challenging behaviour in response to a denial, whilst only two participants showed this association with demands or adult

vocalisation or touch, indicating that denials were a common establishing operation for challenging behaviour in high risk participants.

Given the high frequency of denials as an antecedent to challenging behaviour in the high risk group during the natural observations, the duration of denials experienced by the high risk as compared to the low risk group during the experimental functional analysis warranted investigation. Mann Whitney U analysis showed that within the high attention and demand conditions, the high risk group (high attention: median = 1.94, IQR = 9.93; demand: median = .99, IQR = 2.26) were no more likely to receive denials (high attention: U = 80, p >.05; demand: U = 105.5, p >.05) than the low risk group (high attention: median = 1.87, IQR = 4.73; demand: median = 1.07, IQR = 2.09). However, during the low attention condition, the high risk (median = 3.78, IQR = 6.8) received significantly more denials (U = 54.5, p < .05) than the low risk group (median = .17, IQR = .82), indicating that during the low attention condition, the high risk group were demonstrating more behaviour which the researcher was unable to ignore than the low risk group.

6.4.3 Comparison of proto-imperatives and affect between the high and low risk groups

6.4.3.1 Proto-imperatives during experimental functional analysis

In order to compare the use of proto-imperatives and affect (a proxy indicator of aversiveness) by the high and low risk groups during specific environmental conditions, the percentage duration of approach, protest, negative and positive affect behaviours during the demand and high and low attention conditions was calculated

between groups and across experimental functional analysis conditions within groups (as demonstrated in Figure 6.1).

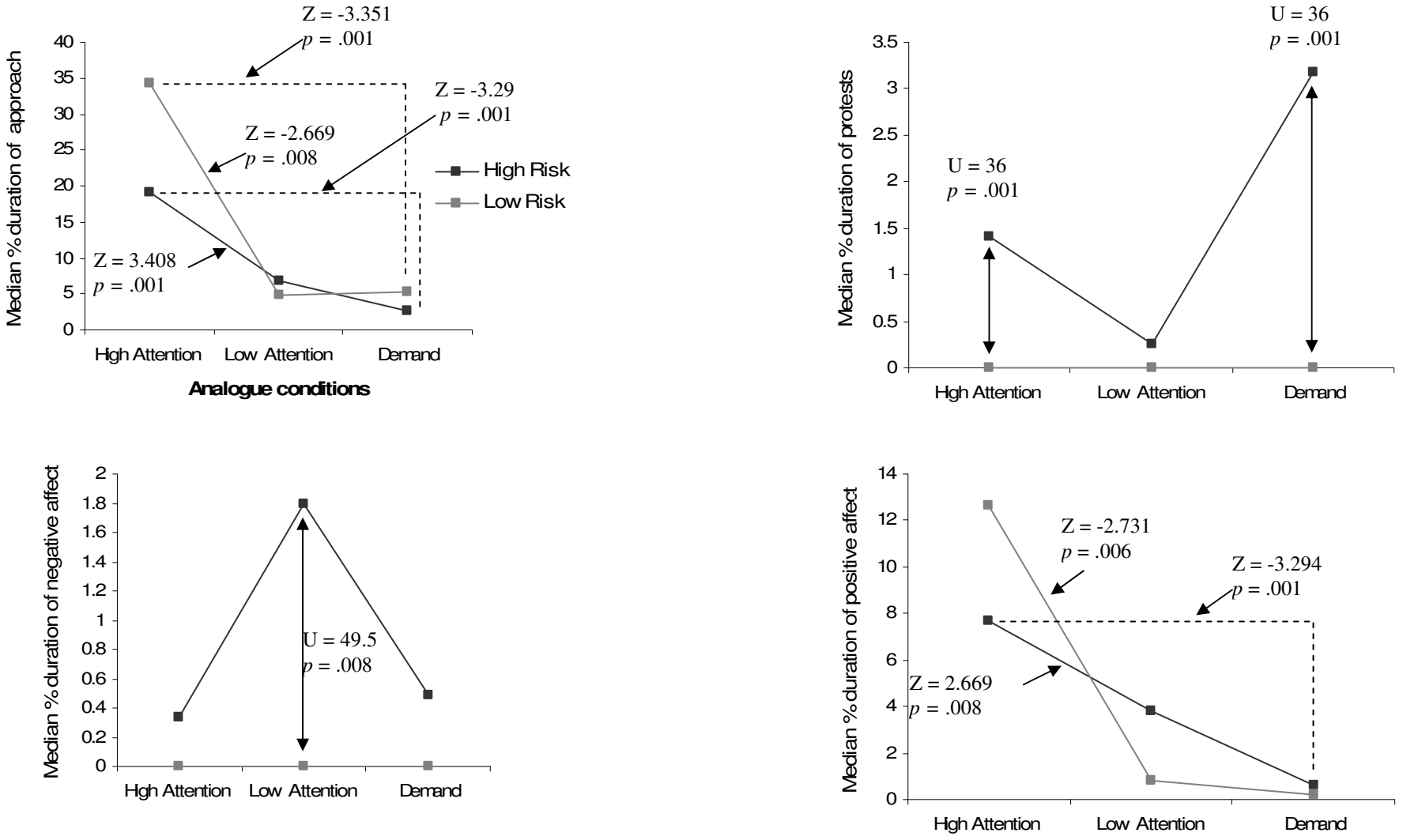


Figure 6.1: Median percentage duration of approach, protest, negative and positive affect during separate analogue conditions

Figure 6.1 shows that the high risk group exhibited significantly longer negative affect during the low attention condition than the low risk group. However, there were no significant differences in terms of duration of negative affect in the high attention and demand condition between the high and low risk groups. Conversely, the high risk group showed significantly longer protests during the high attention and demand conditions than the low risk group. The high risk group also showed more protests than the low risk group during the low attention condition although this difference was not statistically significant. There were no significant differences between the approach behaviour and positive affect shown by the high and low risk groups in any analogue conditions (Appendix I).

These results suggest that the two groups do not differ in terms of positive interactive behaviours during the analogue conditions. However, the high risk group appeared to react more adversely to low levels of adult attention and protested more in the high attention and demand conditions, suggesting that they were more adverse to low levels of adult attention than the low risk group, but they also found adult interaction aversive if it placed a demand upon them.

In order to determine whether there were significant differences in the proto-imperatives and affect behaviours shown within each group across analogue conditions, Friedman's analysis was conducted. For both the high and low risk groups, this analysis rendered significant results for approach (high risk = χ^2 (1, N = 15) = 12.4, p = .002; low risk = χ^2 (1, N = 15) = 19.73, p < .001) and positive affect behaviours (high risk = χ^2 (1, N = 15) = 8.93, p = .01; low risk = χ^2 (1, N = 15) = 9.15, p = .01), but not for protest (high risk = χ^2 (1, N = 15) = 2.58, p = .28; low risk = χ^2 (1,

$N = 15) = 3.94$, $p = .14$) or negative affect (high risk = χ^2 (1, $N = 15) = 2.74$, $p = .25$; low risk = χ^2 (1, $N = 15) = 4.17$, $p = .12$).

Post hoc analyses were then conducted to ascertain, for the high and low risk groups, which conditions differed significantly in terms of duration of approach and positive affect. Comparing these behaviours demonstrated by the high risk participants across analogue conditions, it appears that approach and positive affect behaviours were demonstrated significantly more during the high attention than the low attention and demand conditions (as demonstrated in Figure 6.1). However, there was no significant difference (Z = -1.02, $p = .307$) in the approach behaviour demonstrated by the high risk group between the low attention and demand conditions. This trend was also demonstrated for positive affect, whereby the high risk group showed no significant difference (Z = -.45, $p = .65$) in the duration of protests across the low attention or demand conditions.

Interestingly, the low risk group also showed significantly longer approach and positive affect during the high than the low attention and demand conditions. The difference between the approach demonstrated in the demand and the low attention was also non significant (Z = -.28, $p = .78$), although in the low risk group, there was also no significant difference (Z = -1.92, $p = .06$) in positive affect between the high attention and demand condition. As with the high risk group, there was also no significant difference (Z = -.09, $p = .93$) in the positive affect demonstrated between the low attention and demand conditions. This pattern of results indicates that both the high and low risk participants were using proto-imperatives at a similar and, arguably, appropriate time.

6.4.3.2 The relationship between challenging behaviour and proto-imperatives during natural observations and experimental functional analysis

Sequential lag analysis was used to identify associations between two forms of proto-imperative (protest and approach) and affect and each form of challenging behaviour demonstrated fifteen times or more during the natural observation conditions, as demonstrated in Table 6.4.

Table 6.4: Association between protest, approach, negative affect and challenging behaviour during natural observations (left of the bold line) and experimental functional analysis conditions (right of the bold line) using lag sequential analysis

		Natural observations			Experimental functional Analysis		
Participant	**Behaviour**	**Negative affect**	**Protest**	**Approach**	**Negative affect**	**Protest**	**Approach**
H1	Aggression	n/a	n/a	n/a	nc	nc	nc
	Destruction	n/a	n/a	+	nc	nc	nc
H2	Aggression	+	+	0	+	+	-
	Self-injury	n/a	0	0	nc	nc	nc
H3	Aggression	+	+	+	+	+	-
	Destruction	0	0	0	nc	nc	nc
H4	Destruction	nc	nc	Nc	n/a	n/a	n/a
	Self-injury	n/a	-	0	n/a	n/a	+
H5	Aggression	nc	nc	Nc	+	+	0
	Destruction	nc	nc	Nc	-	n/a	n/a
	Self-injury	+	-	-	+	0	-
H6	Aggression	nc	nc	Nc	n/a	n/a	+
	Destruction	n/a	-	-	nc	nc	nc
H7	Aggression	n/a	0	+	n/a	+	0
	Destruction	-	-	-	0	0	n/a
	Self-injury	+	-	-	+	0	-
H8	Aggression	+	n/a	0	n/a	n/a	+
H9	Destruction	n/a	n/a	n/a	nc	nc	nc
	Self-injury	n/a	n/a	n/a	nc	nc	nc
H10	Destruction	n/a	n/a	-	nc	nc	nc
H11	Aggression	nc	nc	Nc	+	n/a	0
H12	Aggression	nc	nc	Nc	0	n/a	-
	Destruction	-	n/a	-	nc	nc	nc
	Self-injury	-	-	+	+	n/a	-
H13	Self-injury	n/a	+	n/a	-	n/a	-
H14	Destruction	n/a	n/a	-	n/a	0	n/a
	Self-injury	n/a	n/a	-	nc	nc	nc
H15	Destruction	nc	nc	Nc	-	-	-

Yules Q: + = .3 or above, - = -.3 or below, 0 = less than + or -.3, n/a = no conditional probability, nc = challenging behaviour not reaching criteria

Table 6.4 demonstrates that of the thirteen high risk participants demonstrating challenging behaviour at this level, only one participant failed to show a significant relationship between a form of challenging behaviour and a proto-imperative behaviour. Challenging behaviour was associated with negative affect in five participants and protest and approach in three and four participants respectively.

Sequential lag analyses were also used in order to examine associations between proto-imperatives and challenging behaviour during the experimental functional analysis conditions for the high risk participants. As in the natural observations, Table 6.4 demonstrates that only one participant failed to show an association between a proto-imperative and a challenging behaviour. Negative affect also appeared to be associated with challenging behaviour for more participants (seven) than protest (four) and approach (three) (see Table 6.4), although again, because of the small sample size, statistical analysis could not be conducted.

6.5 Discussion

Having established in a companion paper that children at high risk for challenging behaviour do indeed show significantly longer durations of challenging behaviour than children at low risk (section 5.4.1), the primary aim of this paper was to examine the challenging behaviour in controlled and natural environments. The influence of the environment on challenging behaviour at an experimental and naturalistic level was examined in order to determine a potential function of this behaviour. The demonstration of proto-imperative and affect behaviours by each group was also investigated to provide an insight into the communicative repertoire available to both the high and low risk participants. Finally, the association of proto-imperative and

affect behaviours in the same time window as challenging behaviour and specific environmental conditions and settings in the high risk group was examined to ascertain whether the challenging behaviour demonstrated was associated with communicative behaviour.

Examining the presentation of challenging behaviour within experimental and naturalistic settings allowed an in depth assessment of the function of this behaviour in both a controlled and ecologically valid environment. The careful matching of participants and the strict adherence to the experimental functional analysis methodology controlled for many of the extraneous variables which might have affected the challenging behaviour demonstrated by the participants.

Consistent with the results of the companion paper (section 5.4.1), the high risk participants demonstrated significantly longer durations of challenging behaviour (aggression and destruction in particular) during the experimental functional analysis than the low risk participants. For the majority (eleven out of twelve) of high risk participants demonstrating challenging behaviour above a level that enabled analysis (fifteen times or more), at least one of the forms of challenging behaviour demonstrated occurred more frequently during one experimental condition than another, indicating that this behaviour was attention maintained behaviour or reinforced by demand escape, or both. Indeed, around half of the children (seven) showed forms of challenging behaviour which were multifunctional. Despite the use of the high attention condition as a control condition, eight participants also demonstrated significantly more challenging behaviour in this condition than in the

low attention or demand conditions, illustrating a potential social escape function of this behaviour.

At least one form of challenging behaviour demonstrated above a level that enabled analysis by all of the high risk participants during the natural observations was also functional, occurring more during low or undivided attention. Similar to the low attention analogue condition, high rates of behaviour during low attention in the natural observations would suggest behaviour which is attention maintained. However, unlike the high attention experimental functional analysis condition whereby the participant experiences no demands or denials, undivided attention in the natural environment was less controlled and so this behaviour might have been maintained by demand escape for the participants. Indeed, just under half (six out of thirteen) of the high risk participants demonstrated more challenging behaviour in response to a denial in comparison to two in response to a demand or adult vocalisation or touch respectively, indicating that denials were a particularly aversive form of adult interaction for the high risk participants and might have been the cause of much challenging behaviour during the undivided attention condition.

Previous research has already indicated the aversive nature of denials, demonstrating an association between denials and challenging behaviour (e.g. Edelson, Taubman & Lovaas, 1983); however, the characteristics of the high risk participants might cause them to perceive denials as being particularly aversive. Illustrating this, the results of a case study conducted by Murphy, Macdonald, Hall and Oliver (2000) indicated that aggressive behaviour followed prevention of the individual's ritual completion, so that aggression functioned in order to prevent interruption of a ritual. Therefore,

individuals at high risk of challenging behaviour who demonstrate more stereotyped and ritualistic behaviours may find denials particularly aversive if they interrupt these behaviours. Individuals at high risk of challenging behaviour also demonstrate more behavioural correlates of impulsivity, indicating compromised behavioural inhibition (section 3.5). These individuals are thus more likely to find it difficult to inhibit a response when required to do so and thus might perceive denials to be particularly demanding and thus aversive.

Direct comparisons between the challenging behaviour shown in the experimental functional analysis and natural observations are difficult due to the small n and the sometimes different behaviours shown by participants in each setting. Nonetheless, a general overview of these data reveals that all of the high risk participants demonstrated more behaviour during a particular experimental condition or when a setting event or adult behaviour was present. These relationships indicate a significant influence of the environment on challenging behaviour at both an experimental and natural observation level and shows that the behaviour demonstrated by this group is not atypical or random, but a typical reaction to adversely experienced environments. This is significant clinically for the high risk participants as it indicates that despite the presence of behavioural markers, which research has shown increases the likelihood of developing challenging behaviour, the environment continues to have a significant effect on their behaviour and function for their behaviour can be determined.

Examining the demonstration of proto-imperative and affect behaviours across analogue conditions illustrated both similarities and differences between the high and

low risk participants. Both the high and low risk group showed a higher duration of positive affect and approach behaviours than negative affect and protest behaviours, demonstrating more of these behaviours during the high attention, less during low attention and similar or lesser duration during the demand condition. However, the high risk group showed markedly different patterns of negative affect and protest during the separate analogue conditions to the low risk group. These participants showed generally greater duration of negative affect or protest behaviours (although these were not always significant) and significantly more negative affect during the low attention condition and protest during the high attention and demand conditions.

This behavioural trend suggests that the high risk participants found these situations more aversive. The high risk group therefore appear to already have a repertoire of communicative behaviours (supported by the fact that all but one of the high risk participants showed an association between negative affect, protest or approach and challenging behaviour) which they utilise aptly. These children would thus be good candidates for early intervention programmes including functional communication training, a proposition supported by the temporal association identified between proto-imperative and challenging behaviour during the natural observations. The presentation of behavioural risk markers in these high risk children might also enable them to be identified early.

As well as indicating the possibility of success of early intervention programmes for this high risk group, these results also have implications for the broader educational context. Examination of adult variables during individual analogue conditions indicated that the high risk group experienced more adult denials during the low

attention condition, suggesting that without adult attention, these children demonstrated behaviour which the researcher was unable to ignore due to the potential risk to the child. Given the number of children potentially showing behaviours which might put them at high risk of challenging behaviour, this might indicate the high demand these children put on teaching staff. More research in other natural settings is needed to see how widespread these demanding behaviours are and to assess whether they would also impact on the stress experienced by teachers and parents.

Identifying patterns in function within the high risk group would have been more accurate had the sample been greater in size. However, identifying function and temporal associations between proto-imperatives, affect and challenging behaviour in almost every high risk participant indicates that this group are not demonstrating randomly occurring challenging behaviour, but behaviour which would be suitable for functional communication training. Thus, future research could focus on piloting the efficacy of early intervention in this high risk group, to provide proof of principle for the widespread extension of these programmes.

6.6 Summary and implications

Chapter 6 examined the challenging and communicative behaviours demonstrated by high risk participants using both experimental functional and descriptive analyses in order to determine a potential function of this challenging behaviour and the communicative repertoire available to the high risk group. The results of this study indicated that all of the high risk participants showing challenging behaviour above a level which enabled analysis demonstrated more behaviour during a particular

experimental condition, setting event or adult variable, indicating that this behaviour was attention maintained or reinforced by demand escape, or both. In both the experimental functional and descriptive analyses, the majority of participants also showed a significant association between proto-imperative and challenging behaviour, indicating a communicative function of challenging behaviour. High risk participants also demonstrated significantly more negative affect during the low attention and protest during the high attention and demand experimental functional analysis conditions, indicating that they found these situations more aversive.

These results suggest that despite presenting behavioural markers which research has shown increases the likelihood of developing challenging behaviour, the environment continues to have a significant effect on the behaviour demonstrated by high risk children. Since all but one of the high risk participants showed an association between negative affect, protest or approach and challenging behaviour, the high risk group also appear to have a repertoire of communicative behaviours which they can employ appropriately. The combination of these factors suggests that children at high risk would make good candidates for early intervention programmes including functional communication training, whilst the presentation of behavioural risk markers might enable these high risk children to be identified early.

Chapter 7 describes the main findings of this and the preceding three empirical chapters and highlights the main theoretical and clinical implications of these.

Chapter 7

General Discussion

7.1 Background and aims

Challenging behaviour has a detrimental effect on the lives of a significant minority of individuals with intellectual disabilities and is a recurrent theme in research in the intellectual disabilities field. However, despite advances in understanding of the causes of and effective interventions for challenging behaviour, the proportion of individuals with intellectual disabilities developing and continuing to demonstrate this behaviour in later life remain high (e.g. Borthwick-Duffy, 1994; Chadwick, Kusel, Cuddy & Taylor, 2004; Emerson & Bromley, 1995; Emerson *et al.,* 2001a; Emerson *et al.,* 2001b; Lowe *et al.,* 2007; Murphy *et al.,* 1993; Qureshi & Alborz, 1992; Schroeder, Schroeder, Smith & Daldorf, 1978). Bearing in mind the impact of this behaviour on people and the high cost of challenging behaviour for services (e.g. Hallam & Trieman, 2001; Knapp, Comas-Herrera, Astin, Beecham & Pendaries, 2005; Mansell, 1992) it is perhaps unsurprising that researchers are beginning to advocate early intervention as a potentially effective strategy (Richman, 2008; Richman & Lindauer, 2005; Symons, Sperry, Dropik & Bodfish, 2005).

For early intervention programmes to be successful however, models of the development of challenging behaviour suggest that individuals with intellectual disabilities need to receive intervention before challenging behaviour becomes too ingrained in their behavioural repertoire. Thus, children likely to develop challenging behaviour in the future must be identified. Whilst numerous studies have focused on

the child characteristics, or risk markers, associated with challenging behaviour at one point in time (e.g. Chadwick, Piroth, Walker, Bernard & Taylor, 2000; Baghdadli, Pascal, Grisis & Aussilloux, 2003; McClintock, Hall & Oliver, 2003; Holden & Gitlesen, 2006; Oliver *et al.,* in preparation; Petty *et al.,* in preparation) none have addressed the issue of prediction over time or have used a methodology suitable for use in large scale early intervention strategies.

Thus, the aims of this thesis were to examine:

1) The child characteristics acting as putative risk markers for challenging behaviour in children with a severe intellectual disability through the development of a brief screen (Chapter 3).
2) The persistence of the putative risk markers identified as well as their ability to significantly predict the future presence of challenging behaviour in a follow up study (Chapter 4).
3) The concurrent and convergent validity of the brief screen developed (Chapter 5).
4) The challenging, proto-imperative and affect behaviours demonstrated by children who show characteristics that predict the presence of challenging behaviour, in order to determine a potential function of this behaviour and the communicative repertoire available to these individuals (Chapter 6).

7.2 Overview of findings

7.2.1 Age related changes in the prevalence of aggression and self-injury

In Chapter 2, the results of a systematic literature review indicated associations between both aggression and self-injury and age, supporting the results of previous research. Both forms of challenging behaviour appeared to increase in prevalence until early to mid-adulthood, although this association appeared to be more robust for self-injury than aggression. The results from a small number of studies also indicated that the prevalence of self-injury significantly decreased in older adulthood, illustrating a curvilinear relationship between this form of challenging behaviour and age. This might also be true for aggression, although this relationship is less clear. The results from this systematic review were based on relative risk analyses generated from the results of the studies identified with prevalence of aggression and/or self-injury by age band data, allowing an examination of trends in the results across many studies utilising varying samples and age bands. The methodologies as well as the psychometrics of all measures employed by each study were also assessed in order to ensure conclusions drawn were based on a sound understanding of the quality of the results produced by each study. Thus, in summary, the analysis conducted on the findings of studies considered to be the most methodologically robust indicated an association between the prevalence of self-injury, and to a lesser extent aggression, and age.

7.2.2 Putative risk markers of challenging behaviour

In Chapter 3 the results of a cohort study using a large, representative sample of young children and the specifically designed and reliable Challenging Behaviour

Screening Questionnaire (CBSQ) supported the findings of previous research by demonstrating the significantly increased relative risk of challenging behaviour (aggression, destruction, self-injury and one or more forms) given 'probable ASD', repetitive and restricted behaviours and interests and overactivity/impulsivity. A series of binary logistic regressions also indicated that different putative behavioural risk markers act as significant predictors for specific forms of challenging behaviour, so that overactivity/impulsivity significantly predicted the presence of aggression and destruction whilst both overactivity/impulsivity and repetitive and restricted behaviours and interests significantly predicted the presence of self-injury. However, the results of these analyses were affected by the highly conservative use of Bonferroni correction, so that these conclusions must be considered tentatively.

Both 'probable ASD' and the putative behavioural risk markers also significantly increased the relative risk of one or more forms of severe challenging behaviour, although aggression was the only specific form to be significantly associated with these putative risk markers. With only one or more forms of severe challenging behaviour and severe aggression significantly predicted by the putative behavioural risk markers in the binary logistic regression analyses, it appeared that these child characteristics have a stronger association with the presence than the severity of challenging behaviour. The significantly increased relative risk of self-injury and severe self-injury, given the presence of one or more health problems, also indicated the role of health, and by implication pain, for this form of challenging behaviour.

Thus, in summary, four putative risk markers for challenging behaviour in children with severe intellectual disability were identified, the presence of which significantly

increased the relative risk of challenging behaviour. These risk markers appear to have a stronger association with the presence than severity of challenging behaviour though and significantly predict the presence of different forms of challenging behaviour (although these results must be considered in light of the highly conservative approach utilised).

7.2.3 The ability of the behavioural risk markers to significantly predict the future presence of challenging behaviour

Replicating the screen methodology, a follow up study employing over 66% of the original screen sample provided further support for the behavioural risk markers. As described in Chapter 4, the results of this study demonstrated significantly increased relative risks of persistent challenging behaviour given the presence of repetitive and restricted behaviours and interests and overactivity/impulsivity 18 months previously (with the exception of overactivity/impulsivity which failed to significantly increase the relative risk of self-injury). Both behavioural risk markers were less successful in predicting the remission or incidence of challenging behaviour. Concern regarding a participant's self-injury expressed by teachers at the screen was also significantly higher for participants who persisted to show self-injury at the follow up, although health did not significantly predict self-injury as it did at the screen.

Thus the results of Chapter 4 indicated that behavioural risk markers identified in Chapter 3 continue to predict the persistence of challenging behaviour. Concern at screen was also significantly associated with self-injury although health was not.

7.2.4 The concurrent and convergent validity of the CBSQ

In order to examine the concurrent and convergent validity of the CBSQ, natural observations (affording high ecological validity), questionnaires and objective measures were employed in a study presented in Chapter 5. Participants identified as being at high or low risk of challenging behaviour based on the presence of the behavioural risk markers were recruited from the sample described in Chapter 3 and closely matched for age and degree of intellectual disability. The results of this study supported the concurrent validity of the CBSQ as high risk participants demonstrated significantly greater duration of overactive, repetitive and impulsive like behaviour at observation. With the exception of the results obtained from the Challenging Behaviour Interview, the convergent validity of the CBSQ also appeared to be good. The significantly longer duration of challenging behaviour demonstrated by the high risk group, at a total and specific form level, indicated that children were correctly identified as being at high or low risk of challenging behaviour.

7.2.5 The function and communicative nature of challenging behaviour demonstrated by high risk participants

Having established the ability of the CBSQ to identify children at risk of challenging behaviour at one point in time and those still demonstrating the behaviour 18 months later, as well as the validity of these findings, the function and communicative nature of the challenging behaviour demonstrated by the high risk participants was examined in order to assess the potential suitability of these children for the kinds of behavioural interventions likely to be central to early intervention programmes. Using both descriptive and functional analyses, the study outlined in Chapter 6 allowed an in depth assessment of challenging and other relevant behaviours in both a controlled

and ecologically valid environment. The results of this study showed the functional quality of this behaviour in the high risk children, as all of the high risk participants showing challenging behaviour fifteen times or more during experimental functional or descriptive analyses demonstrated more behaviour during a particular experimental condition or when a setting event or adult behaviour occurred. Additionally, in both the experimental functional and descriptive analyses, only one participant failed to show a significant association between a proto-imperative and challenging behaviour, indicating a close association between communicative and challenging behaviour for the majority of participants. The functional and potentially communicative nature of the challenging behaviour demonstrated by the high risk participants indicated that they are likely to benefit from functional communication training as part of a wider early intervention strategy.

7.3 Limitations

7.3.1 Sample heterogeneity

As discussed in Chapters 3 and 4, perhaps the most significant limitation of these two studies was the administratively defined sample employed. Having recruited participants from schools for children with a severe intellectual disability, it was assumed that the majority of participants had a severe intellectual disability, although it was also acknowledged that variability in the degree of participants' intellectual disability was inevitable and that, subsequently, some children included in these studies might have a more or less severe intellectual disability. This potentially large sample heterogeneity was justified however by the need to recruit a large population in order to examine the putative risk markers and test their suitability for indicators of

high risk in an early intervention context. Such sample heterogeneity would also occur in clinical practice, since the recruitment of a large administratively defined population would also be required for the identification of children at high risk in this context.

Further heterogeneity of the duration of risk markers demonstrated by participants, particularly within the high but also within the low risk group for some risk markers, was also a limitation of the sample employed in Chapters 5 and 6. With some large interquartile range differences in the duration of risk markers demonstrated within both groups, it was clear that broad heterogeneity within each group was present. Given the similar composite risk scores computed from the original screen scores within each group, these differences were not predicted. The source of this within group variability might have been a change in participant's characteristics in the 18 months between the screen and validation study, so that participants lost or acquired a risk marker in this time period. Poor test-retest reliability of the CBSQ is also a possibility, although this would not be predicted based on the satisfactory inter-rater reliability of the measure. Whatever the cause of this variability, it must be acknowledged that the use of the CBSQ in this context might result in some false classification of children as being at low or high risk of challenging behaviour. Given the serious consequences of challenging behaviour in this population, this is a limitation requiring further investigation.

7.3.2 Use of Bonferonni correction

The application of Bonferroni corrections to the binary logistic regressions conducted in Chapters 3 and 4 (sections 3.4.3.3 and 4.4.12) represents a highly conservative

approach in an attempt to avoid Type 1 errors. Whilst this is advantageous, in utilising this approach, risk markers were, on occasion, deemed not to significantly predict challenging behaviour (RRBI and destruction in section 3.4.3.3 and RRBI and aggression in section 4.4.12), even though the likelihood of challenging behaviour was similar given other risk markers deemed to significantly contribute to the model. Thus, whilst the use of Bonferroni corrections is legitimate, had they not been applied, the models would have been different and there would have been less evidence for the notion of different risk markers predicting different challenging behaviours (section 7.2.2)

7.3.3 Limitations associated with functional and descriptive analyses

The conclusions regarding the functional and communicative nature of challenging behaviour demonstrated by high risk participants, based on the experimental functional and descriptive analysis conducted in Chapter 6, must also be considered carefully due to limitations associated with these methodologies. Whilst the results gained from experimental functional analysis allude to the potential function of challenging behaviour, several participants failed to show any difference in the rate of behaviours demonstrated across conditions. Whether this indicates behaviour which is multifunctional or has no function whatsoever could not be deduced from these findings. Many participants also showed significantly higher rates of challenging behaviour during high attention, the putative control condition. Given the high rate of behaviour during this condition, it is possible that the behaviour demonstrated functioned as social escape for these participants. Whilst this is an apparently adequate explanation, it does indicate that participants were experiencing the

experimental functional analysis conditions in a different way to that intended in the methodology.

Additionally, although the descriptive analysis allowed an ecologically valid measure of behavioural function, the level of control afforded during this analysis was far less. Thus, although the close temporal association between the setting events, adult behaviours and child behaviours indicates a function and communicative quality for the challenging behaviour demonstrated, other explanations cannot be ruled out due to the lack of control inherent in this methodology.

7.3.4 Inter-rater reliability of the CBSQ

Finally, the inter-rater reliability of the CBSQ, whilst good within one environment, was for some items quite low when rated across environments. However, as discussed in Chapter 3, the low inter-rater reliability of overactivity and impulsivity across teachers and parents is commonly reported within the literature, with parents tending to rate overactivity and impulsivity higher than teachers, (Amador-Campos, Forns-Santacana, Guardia-Olmos & Pero-Cebollero, 2006; Charach, Chen, Hogg-Johnson & Schachar, 2009; Papageorgiou, Kalyva, Dafoulis & Vostanis, 2008) and was thus not unexpected. It is also perhaps fair to assume that other constructs such as repetitive behaviour might also have had a low inter-rater reliability across environments due to genuine behavioural differences across environments.

7.3.5 Presence of genetic syndromes

Considering the well documented association between an increased prevalence of challenging behaviour and particular genetic syndromes (e.g. Clarke & Boer, 1998;

Kenworthy & Charnas, 1995; Nyhan, 1994), it might be argued that each of the high risk participants was merely high risk due to higher rates of genetic syndromes in this group. However, teachers were asked to indicate the cause of participants' intellectual disability so that this confound might be ruled out. Additionally, research has indicated that risk markers such as restricted behaviours continue to increase the risk of challenging behaviour within genetic syndromes, such as Cornelia de Lange and Prader-Willi syndromes (Clarke *et al.,* 2002; Hyman, Oliver & Hall, 2002).

7.4 Theoretical implications

7.4.1 The developmental progressions of different forms of challenging behaviour

The results of the systematic review conducted in Chapter 2, indicated that both aggression and self-injury were associated with age, although the association between age and challenging behaviour appeared to be stronger for self-injury than aggression. As a result of these findings, the focus of research conducted in this area might now shift to the theoretical underpinnings of the discrepancy between forms of challenging behaviour. More methodologically robust studies aimed specifically at investigating the prevalence of various forms of challenging behaviour with age should also be conducted, as conclusions based on the prevalence of specific forms of challenging behaviour with age might change given the results of more methodologically robust studies.

To date, much attention has been paid to the development of challenging behaviour generally, although comparatively less to the ontogeny of specific forms. Guess and

Carr's (1991) stage model however does provide a detailed account for the development of self-injury, indicating its emergence from repetitive behaviours as a unique course and whilst the results of this literature review cannot support this model in any concrete fashion, they do suggest potentially different developmental progressions of different forms of challenging behaviour, as proposed in the model. This supposition is also supported by previous research which has also indicated that self-injury is related to health problems and pain, (e.g. Carr & Owen-DeSchryver, 2007; de Lissovoy, 1962; Hart, Bax & Jenkins, 1984; Luzzani, Macchini, Valade, Milani & Selicorni, 2003) although these characteristics have not been associated with aggression.

More generally, these results also have wider implications for this field of research as a whole, indicating that assumptions, such as those based on the increased prevalence of challenging behaviour with age, should be rigorously tested so that more accurate conclusions from existing research can be drawn.

7.4.2 Interactions between repetitive behaviour, operant reinforcement and compromised behavioural inhibition

In Chapters 3 and 4, the presence of repetitive and restricted behaviours and interests significantly increased the relative risk of self-injury at one point in time as well as its persistence 18 months later, indicating a strong relationship between these two behaviours. This relationship is central to Guess and Carr's (1991) stage model of self-injury, whereby repetitive behaviour evolves into self-injury under the influence of social reinforcement, a supposition supported by Oliver (1993, 1995) who further

developed this model into a mutual reinforcement paradigm. The results described in Chapter 3 and 4 could thus be interpreted as supporting Guess and Carr's proposal.

The superior predictive validity of repetitive and restricted behaviours and interests with regard to self-injury, as compared to overactivity/impulsivity, identified in Chapter 4 was explained with reference to Guess and Carr's (1991) model, in conjunction with compromised behavioural inhibition. Thus, in the development of this form of challenging behaviour, self-injury which has arisen as the result of operant reinforcement is more prevalent in children with compromised inhibition as they find it harder to inhibit this learned response. It was also suggested that children with repetitive behaviour already demonstrate a behaviour which is structurally similar to self-injury and thus through positive reinforcement is easily shaped to become self-injurious, as suggested by Guess and Carr. Thus, children with repetitive and restricted behaviours and interests are more at risk for self-injury than children with overactivity/impulsivity alone, whose only difficulty is compromised behavioural inhibition (Petty *et al.,* in preparation).

However, as also discussed in Chapter 4, the lack of an association between overactivity/impulsivity at screen and the incidence of aggression and destruction at follow up was explained by the significantly higher number of participants acquiring, as opposed to losing, the overactivity/impulsivity risk marker. It was therefore suggested that the incidence of aggression and destruction was caused by a mechanism which also brought about the increase in overactive and impulsive behaviour demonstrated by participants. This supposition was supported by the results of a recent study, which identified temporal associations between repetitive and self-

injurious behaviour (Petty, Oliver & Allen, 2009). Such temporal associations between the putative behavioural risk markers and challenging behaviour allude to the potential presence and function of some sort of common mechanism, responsible for the presence of both types of behaviour. If this were proven to be the case, it would indicate that challenging behaviours such as self-injury might not emerge from prior putative risk markers like repetitive and restricted behaviours, as indicated by Guess and Carr (1991), but that these behaviours might co-exist. Therefore, the results of Chapter 3 and 4 potentially provide some indirect evidence for Guess and Carr's model, although further research is required in order to examine whether the relationship between repetitive and self-injurious behaviour is causative, as proposed by Guess and Carr (1991), or merely one of association whereby two behaviours happen to co-occur.

7.4.3 Health and pain

Also reported to be significantly associated with self-injury at one point in time, although not the persistence of this behaviour 18 months later, was the presence of one or more health problems, and by implication pain. This association was explained with reference to three mechanisms. The first two mechanisms proposed both implied that self-injury functioned as a method of pain removal, either directly or via the gate control theory of pain (Melzack & Wall, 1965). The third suggested that the presence of pain might cause individuals to perceive a task as more aversive, leading to challenging behaviour, as indicated in previous research (Carr, Smith, Giacin, Whelan & Pancari, 2003). Thus, health appears to be an important risk marker for self-injury and is a particularly significant issue for individuals with intellectual disabilities due to the proportion demonstrating health problems (Berg, Arron, Burbidge, Moss &

Oliver, 2007). Whilst health did not significantly increase the relative risk of the persistence of self-injury 18 months later, increases in the prevalence of health problems over this 18 month period were demonstrated, indicating that individuals with intellectual disabilities might experience more health problems with age. This indication warrants further investigation and implies that health problems in individuals with intellectual disabilities should be closely monitored.

7.4.4 Biological models of challenging behaviour: compromised behavioural inhibition and monoamine oxidase A (MAOA)

As already discussed in this chapter, the repetitive and restricted behaviours and interests and the overactivity and impulsivity demonstrated by participants at high risk of challenging behaviour potentially indicated compromised behavioural inhibition, based on the work of Barkley (1997a, 1997b) and Turner (1997, 1999) in ADHD and autism respectively. In conjunction with operant theory (e.g. Oliver, 1995; Oliver, Hall & Murphy, 2005), compromised behavioural inhibition provides a cognitive extension to models of challenging behaviour, so that challenging behaviour is evoked under stimulus conditions and reinforced so that it becomes a learned response. Children with compromised behavioural inhibition however find it even harder to inhibit this learned response and thus might show a greater prevalence of challenging behaviour. Further research into the executive functioning abilities of children with intellectual disabilities and challenging behaviour and inhibition specifically are required though in order to provide empirical support for this model.

Another biological explanation of challenging behaviour described in Chapter 3 involved the promoter gene monoamine oxidase A (MAOA). Already implicated in

the expression of aggression in typically developing individuals (Caspi *et al.,* 2002; Brunner, Nelen, Breakefield, Ropers & Oost, 1993), the authors of a recent study also reported a significantly higher prevalence of the short allele MAOA polymorphism in individuals with intellectual disabilities and challenging behaviour than individuals with or without intellectual disabilities and no challenging behaviour (May *et al.,* 2009). Whilst research regarding MAOA is still in its infancy, evidence is accumulating in support of biological vulnerabilities to challenging behaviour.

MAOA might be associated with aggression via two distinct mechanisms, by influencing particular neurotransmitter function or modifying levels of impulsivity. As previously discussed (section 1.2.1.1.5) MAOA is involved in the biodegradation of serotonin and other monoaminergic neurotransmitters associated with an aggressive phenotype in both animal and human models. It is also possible that MAOA is associated with aggression via increased levels of impulsivity, an already established behavioural risk marker (section 7.2.2) related to compromised behavioural inhibition. MAOA might have a diverse range of presently undetected effects and one of these could involve parallel structural or functional changes, influencing the levels of particular neurotransmitters as well as the brain regions responsible for the demonstration of behavioural inhibition and impulsive behaviour.

7.4.5 An integrated model of challenging behaviour

Whilst both biological and behavioural theories have been proposed with regard to the cause of challenging behaviour in children at high risk of challenging behaviour, it is clear that a complete model of challenging behaviour requires reference to both, so that risk markers with a biological underpinning, such as impulsivity, function to

cause challenging behaviour in interaction with operant reinforcement. As already discussed, compromised behavioural inhibition becomes particularly problematic for children at risk of challenging behaviour when challenging behaviour has become a learned response. Children with repetitive and restricted behaviour might also become self-injurious due to operant reinforcement. Indeed, many of the child characteristics which increase the risk of challenging behaviour do so in the context of the environment. As described in Chapter 7, participants with restricted behaviour in the form of rituals might also demonstrate challenging behaviour in response to termination of these rituals (Murphy, Macdonald, Hall & Oliver, 2000; Reese, Richman, Belmont & Morse, 2005). Thus environmental factors, such as adult denials in conjunction with the restricted behaviour risk marker might increase the prevalence of challenging behaviour demonstrated.

As described in Chapter 6, high risk participants also demonstrated a significantly lower attention span and significantly higher activity level than low risk participants. The combination of these factors would indicate that these children show behaviours similar to those demonstrated by children with ADHD. Such behaviour is contrary to that expected or required in many situations, such as the classroom, and, as a result, children displaying this behaviour are perhaps more likely to receive behaviour management in the form of demands and denials which they find aversive (as demonstrated in Chapter 6) and thus demonstrate challenging behaviour (e.g. Edelson, Taubman & Lovaas, 1983).

Thus, considering child characteristics in the context of environmental factors is important for understanding the causes of challenging behaviour. Advances in the

understanding of this interaction will also enable effective interventions to be developed as part of early intervention strategies.

7.5 Clinical implications

The fundamental aim of this thesis was to examine whether the identification of children with severe intellectual disabilities at risk of developing challenging behaviour using a short and accessible measure was possible. In answer to this, the results described in Chapter 3 and 4 demonstrated that two behavioural risk markers were able to significantly predict the presence of challenging behaviour at one point in time, as well as its persistence 18 months later, so that children at risk of challenging behaviour could be identified through presentation of these behavioural characteristics. The results presented in Chapters 5 and 6 subsequently demonstrated that these findings were valid and that children identified as being at high risk for challenging behaviour were also apparently good candidates for functional communication training.

Building on the work of established advocates of early intervention services (Richman, 2008; Richman & Lindauer, 2005; Symons *et al.,* 2005) these findings have substantial clinical implications, providing the first robust, empirical evidence for the potential effectiveness of early intervention programmes. A reliable and valid measure has also been developed and purposefully designed to be accessible to those working with children with severe intellectual disabilities, so that the identification of children with intellectual disabilities at risk of challenging behaviour could begin as soon as the relevant services are established. Motivation to implement early intervention strategies is also provided by the results of Chapter 6 which demonstrated

the amount of resources currently required to work with and manage the behaviour of high risk children and thus the need to target interventions directly at these individuals.

If early intervention with this population is to succeed, serious thought must be given as to how services currently using reactive strategies can best become proactive. In order to identify young children at high risk of challenging behaviour, education and health services must work collaboratively so that children in child development services and school nurseries demonstrating risk markers can be quickly referred to health services before challenging behaviour develops and becomes ingrained in the child's behavioural repertoire. Such collaborations are likely to initially be both resource intensive and expensive, but could drastically reduce future economic and personal costs.

7.6 Future research

Whilst the results presented provide promise for the effective implementation of early intervention services in children with severe intellectual disability, further research is required to enhance our understanding of the general developmental trajectory of challenging behaviour in this population as a whole, as well as in high risk participants in particular. It was concluded in Chapter 2 that aggression and self-injury appear to increase in prevalence until early to mid-adulthood, at which point they decline. However, this association was more robust for self-injury than aggression. It was also reported that the number of studies providing the data for this analysis was small and that methodological limitations inherent in many of them made drawing conclusions from these data difficult, thus, further research is required.

Also requiring further investigation are the underlying sources of risk. Although compromised behavioural inhibition has been implicated in the presence of repetitive, restricted, overactive and impulsive behaviour in children at high risk of challenging behaviour, the cause of this compromised executive dysfunction has not been examined. The potential source of this limitation might well be genetic and more specifically, the presence of genetic syndromes in participants at high risk. However, other causes are possible and thus should be investigated in order to aid our understanding of the fundamental cause of risk in these high risk individuals. It is therefore recommended that further studies aimed specifically at examining the prevalence of challenging behaviour with age and sources of risk for specific risk markers should be conducted to better understand the causes of different forms of challenging behaviour and so adequately provide for the future needs of children with intellectual disabilities.

The development of the risk markers and challenging behaviour demonstrated by the high risk participants should also be examined at a later date, so that the ability of the putative risk markers to predict the presence of challenging behaviour over a more substantial time period can be assessed. Pilot studies recruiting these or other children with intellectual disabilities identified as being at high risk of challenging behaviour should also be conducted in order to provide more direct proof of principle for early intervention strategies with this population.

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University of Birmingham and Birmingham Children's Hospital NHS Trust

Children's Behaviour Questionnaire

School Questionnaire – Children Under 6 Years of Age

Instructions

Please read all questions carefully before answering.

For all questions, please tick the appropriate box or circle the most appropriate response.

For example, for this question, if the child has never had eye problems, circle 0 under never.

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

	Never	Mild	Moderate	Severe
Eye problems (eg. infections)	**(0)**	**1**	**2**	**3**

Thank you very much for taking the time to complete this questionnaire.

If you have any questions or would like any further information please do not hesitate to contact:

Chris Oliver on phone
Louise Davies on phone

UNIVERSITY OF BIRMINGHAM

Birmingham Children's Hospital NHS
NHS Trust

Background Information

Child's name: ..... **Child's date of birth:**.....

Child's gender: **Male** **Female**

Child's ethnic origin: (please tick one box)

White-British	Bangladeshi .....
White-Other	Black-African .....
Chinese	Black African Caribbean .....
Indian	Pakistani .....
Vietnamese	Yemeni .....
Other ethnic group	Mixed parentage .....
(please specify)	(please specify) .....

Child's home address: .....

……**Postcode:** ……..……

Has any professional (eg. doctor, clinical geneticist, paediatrician) said that the child:

Is autistic .....Yes/No	Has an autistic spectrum disorder..... Yes/No
Has autistic like traits.....Yes/No	Has features of autism ..... Yes/No
Has cerebral palsy or muscular dystrophy .....Yes/No	
Has a genetic syndrome: .....Yes/No	

If you answered yes to genetic syndrome, please state which: ……..…….

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

.....

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

0-5 5-10 10-15 20+ N/A

General Health and Development

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

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

Please tick the appropriate response regarding the child's characteristics

Vision	Blind or almost…..	Poor…..	Normal…..
Hearing	Deaf or almost…...	Poor…..	Normal…..

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

	Never	Mild	Moderate	Severe
Eye problems (eg. infections)	0	1	2	3
Ear problems (eg. infections)	0	1	2	3
Dental problems (eg. cavities/gum problems)	0	1	2	3
Digestive problems (eg. reflux/stomach problems)	0	1	2	3
Skin problems (eg. eczema/dry skin)	0	1	2	3
Any other health or painful condition	0	1	2	3

(please specify) .....

Behaviour

	How frequently does this problem occur from 1 (never) to 5 (very often)					**How difficult is it to manage this problem from 1 (not difficult) to 5 (seriously difficult to manage)**				
Does the child show:										
Repetitive movements	1	2	3	4	5	1	2	3	4	5
Obsessions and rituals	1	2	3	4	5	1	2	3	4	5

1. <u>In the last month</u>, has the child displayed physical <u>aggression</u> eg. punching, pushing, kicking, pulling hair, grabbing other's clothing? Yes No

<u>If you answered yes to Q1, please answer Questions 1a-1d. If you answered no, please go to Question 2.</u>

1a. <u>In the last month</u>, how long did the <u>longest</u> episode or burst of <u>aggression</u> last?

Less than a minute	Less than 5 minutes	Less than 15 minutes	Less than an hour	More than an hour

1b. <u>In the last month</u>, as a result of this behaviour, has physical contact or prevention or restraint by others been necessary eg. blocking, taking objects from an individual, temporary restraint of an arm?

Never	At least once a month	At least once a week	At least once a day	At least once an hour

1c. <u>In the last month</u>, think about how often this behaviour occurred. If there was no change and you watched the child now, then would you definitely see the behaviour?

By this time next month	By this time next week	By this time tomorrow	In the next hour	In the next 15 minutes

1d. How concerned are you about this <u>aggressive</u> behaviour?

0	1	2	3	4
Not at all concerned				Extremely concerned

2. <u>In the last month</u>, has the child displayed <u>disruption and destruction of property or the environment</u> eg tearing or chewing own clothing, tearing newspapers, breaking windows or furniture, slamming doors, spoiling a meal? Yes No

<u>If you answered yes to Q2, please answer Questions 2a-2d. If you answered no, please go to Question 3.</u>

2a. In the last month, how long did the longest episode or burst of destructive behaviour last?

Less than a minute	Less than 5 minutes	Less than 15 minutes	Less than an hour	More than an hour

2b. In the last month, as a result of this behaviour, has physical contact or prevention or restraint by others been necessary eg. blocking, taking objects from an individual, temporary restraint of an arm?

Never	At least once a month	At least once a week	At least once a day	At least once an hour

2c. In the last month, think about how often this behaviour occurred. If there was no change and you watched the child now, then would you definitely see the behaviour?

By this time next month	By this time next week	By this time tomorrow	In the next hour	In the next 15 minutes

2d. How concerned are you about this destructive behaviour?

0	1	2	3	4
Not at all concerned				Extremely concerned

3. In the last month, has the child displayed self injurious behaviour eg head banging, head-punching or slapping, removing hair, self-scratching body hitting, eye poking or pressing? Yes No

If you answered yes to Q3, please answer Questions 3a-3d. If you answered no, please go to the final Question in this pack

3a. In the last month, how long did the longest episode or burst of self injury last?

Less than a minute	Less than 5 minutes	Less than 15 minutes	Less than an hour	More than an hour

3b. In the last month, as a result of this behaviour, has physical contact or prevention or restraint by others been necessary eg. blocking, taking objects from an individual, temporary restraint of an arm?

Never	At least once a month	At least once a week	At least once a day	At least once an hour

3c. In the last month, think about how often this behaviour occurred. If there was no change and you watched the child now, then would you definitely see the behaviour?

By this time next month	By this time next week	By this time tomorrow	In the next hour	In the next 15 minutes

3d. How concerned are you about this self injurious behaviour?

0	1	2	3	4
Not at all concerned				Extremely concerned

Does the child…….	**Never/ almost never**	**Some of the time**	**Half of the time**	**A lot of the time**	**Always/ almost all the time**
Find it difficult to wait?.....	0	1	2	3	4
Act as if driven by a motor?.....	0	1	2	3	4
Want things immediately?.....	0	1	2	3	4
Find it difficult holding still?	0	1	2	3	4

University of Birmingham
and Birmingham Children's Hospital NHS Trust

Children's Behaviour Questionnaire

School Questionnaire – Children Aged 6 Years and Older

Instructions

Please read all questions carefully before answering.

For all questions, please tick the appropriate box or circle the most appropriate response.

For example, for this question, if the child has never had eye problems, circle 0 under never.

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

	Never	Mild	Moderate	Severe
Eye problems (eg. infections)	(0)	1	2	3

Thank you very much for taking the time to complete this questionnaire.

If you have any questions or would like any further information please do not hesitate to contact:

[redacted]

UNIVERSITY OF BIRMINGHAM

Birmingham Children's Hospital NHS
NHS Trust

Background Information

Child's name: ..... **Child's date of birth:**.....

Child's gender: **Male** **Female**

Child's ethnic origin: (please tick one box)

White-British	Bangladeshi .....
White-Other	Black-African .....
Chinese	Black African Caribbean .....
Indian	Pakistani .....
Vietnamese	Yemeni .....
Other ethnic group	Mixed parentage .....
(please specify)	(please specify) .....

Child's home address: .....

……**Postcode:** ……..……

Has any professional (eg. doctor, clinical geneticist, paediatrician) said that the child:

Is autistic .....Yes/No	Has an autistic spectrum disorder..... Yes/No
Has autistic like traits.....Yes/No	Has features of autism ..... Yes/No
Has cerebral palsy or muscular dystrophy .....Yes/No	
Has a genetic syndrome: .....Yes/No	

If you answered yes to genetic syndrome, please state which: ……..…….

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

.....

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

0-5 5-10 10-15 20+ N/A

General Health and Development

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

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

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

	Never	Mild	Moderate	Severe
Eye problems (eg. infections)	0	1	2	3
Ear problems (eg. infections)	0	1	2	3
Dental problems (eg. cavities/gum problems)	0	1	2	3
Digestive problems (eg. reflux/stomach problems)	0	1	2	3
Skin problems (eg. eczema/dry skin)	0	1	2	3
Any other health or painful condition	0	1	2	3

(please specify) .....

Behaviour

	How frequently does this problem occur from 1 (never) to 5 (very often)					**How difficult is it to manage this problem from 1 (not difficult) to 5 (seriously difficult to manage)**				
Does the child show:										
Repetitive movements	1	2	3	4	5	1	2	3	4	5
Obsessions and rituals	1	2	3	4	5	1	2	3	4	5

1. <u>In the last month</u>, has the child displayed physical <u>aggression</u> eg. punching, pushing, kicking, pulling hair, grabbing other's clothing? Yes No

<u>If you answered yes to Q1, please answer Questions 1a-1d. If you answered no, please go to Question 2.</u>

1a. <u>In the last month</u>, how long did the <u>longest</u> episode or burst of <u>aggression</u> last?

Less than a minute	Less than 5 minutes	Less than 15 minutes	Less than an hour	More than an hour

1b. <u>In the last month</u>, as a result of this behaviour, has physical contact or prevention or restraint by others been necessary eg. blocking, taking objects from an individual, temporary restraint of an arm?

Never	At least once a month	At least once a week	At least once a day	At least once an hour

1c. <u>In the last month</u>, think about how often this behaviour occurred. If there was no change and you watched the child now, then would you definitely see the behaviour?

By this time next month	By this time next week	By this time tomorrow	In the next hour	In the next 15 minutes

1d. How concerned are you about this <u>aggressive</u> behaviour?

0	1	2	3	4
Not at all concerned				Extremely concerned

2. <u>In the last month</u>, has the child displayed <u>disruption and destruction of property or the environment</u> eg tearing or chewing own clothing, tearing newspapers, breaking windows or furniture, slamming doors, spoiling a meal? Yes No

<u>If you answered yes to Q2, please answer Questions 2a-2d. If you answered no, please go to Question 3.</u>

2a. In the last month, how long did the longest episode or burst of destructive behaviour last?

Less than a minute	Less than 5 minutes	Less than 15 minutes	Less than an hour	More than an hour

2b. In the last month, as a result of this behaviour, has physical contact or prevention or restraint by others been necessary eg. blocking, taking objects from an individual, temporary restraint of an arm?

Never	At least once a month	At least once a week	At least once a day	At least once an hour

2c. In the last month, think about how often this behaviour occurred. If there was no change and you watched the child now, then would you definitely see the behaviour?

By this time next month	By this time next week	By this time tomorrow	In the next hour	In the next 15 minutes

2d. How concerned are you about this destructive behaviour?

0	1	2	3	4
Not at all concerned				Extremely concerned

3. In the last month, has the child displayed self injurious behaviour eg head banging, head-punching or slapping, removing hair, self-scratching, body hitting, eye poking or pressing? Yes No

If you answered yes to Q3, please answer Questions 3a-3d. If you answered no, please go to the final Question in this pack

3a. In the last month, how long did the longest episode or burst of self injury last?

Less than a minute	Less than 5 minutes	Less than 15 minutes	Less than an hour	More than an hour

3b. In the last month, as a result of this behaviour, has physical contact or prevention or restraint by others been necessary eg. blocking, taking objects from an individual, temporary restraint of an arm?

Never	At least once a month	At least once a week	At least once a day	At least once an hour

3c. In the last month, think about how often this behaviour occurred. If there was no change and you watched the child now, then would you definitely see the behaviour?

By this time next month	By this time next week	By this time tomorrow	In the next hour	In the next 15 minutes

3d. How concerned are you about this self injurious behaviour?

0	1	2	3	4
Not at all concerned				Extremely concerned

Does the child…….	**Never/ almost never**	**Some of the time**	**Half of the time**	**A lot of the time**	**Always/ almost all the time**
Find it difficult to wait?	0	1	2	3	4
Act as if driven by a motor?.....	0	1	2	3	4
Want things immediately?	0	1	2	3	4
Find it difficult holding still?.....	0	1	2	3	4

Table of medians, inter-quartile range differences and Mann Whitney U analysis for the putative risk markers between groups who were and were not included in the follow up sample

Putative Risk Markers	Included	Not included	U	*p*
Repetitive and Restricted Behaviours and Interests	3 (4)	3 (4)	36051.5	.77
Overactivity/Impulsivity	3 (7)	3 (6)	33569.5	.8
Health	0 (2)	0 (2)	40911.5	.83
Concern Regarding Aggression	2 (2)	2 (2)	4800.5	.88
Concern Regarding Self Injury	2 (2)	2 (2)	1050	.85

Table of Chi Square results for differences in frequency and severity of challenging behaviour between groups who were and were not included in the follow up sample

Challenging Behaviour	χ^2	p
Severe Aggression	.285	.36
Destruction	.1	.19
Severe Destruction	.251	.4
Self Injury	.117	.38
Severe Self Injury	.014	.54

Table of the relative risk of remission, incidence and persistence of challenging behaviour given presence of one or more health problems during the last month at screen

Challenging Behaviour	Groups	Relative Risk (CI)
Aggression	**Remission**	1.03 (.39, 2.69)
	Incidence	.69 (.29, 1.64)
	Persistence	.98 (.55, 1.75)
Destruction	**Remission**	1.3 (.43, 3.91)
	Incidence	.94 (.41, 2.18)
	Persistence	.96 (.38, 2.45)
Self Injury	**Remission**	.99 (.32, 3.04)
	Incidence	71 (.2, 2.46)
	Persistence	2.02 (.79, 5.15)
One or more	**Remission**	.77 (.31, 1.93)
	Incidence	.63 (.28, 1.41)
	Persistence	.1.09 (.7, 1.72)

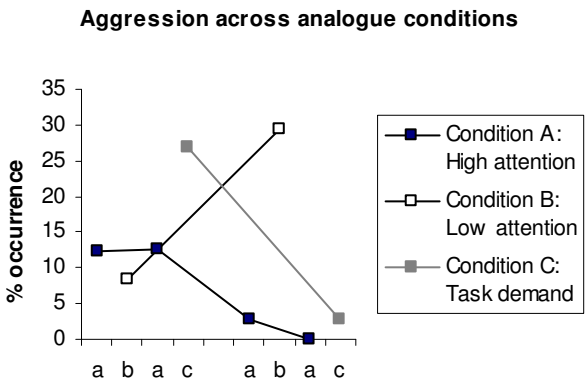
Table of mean and standard deviation Vineland Adaptive Behavior Scale total raw, communication and daily living skills scores for the high and low risk groups and t-test analysis

VABS Domain	High Risk	Low Risk	t	*p*
Total raw score	134.73 (30.02)	149.73 (27.58)	1.425	.165
Communication	41.87 (10.79)	43.07 (10.89)	.303	.764
Daily Living Skills	41.33 (13.96)	45.47 (10.3)	.923	.364

Table of SCQ subscale scores for the high and low risk groups

	Verbal		Non-Verbal	
Subscale	**High Risk**	**Low Risk**	**High Risk**	**Low Risk**
Communication	6.8	5.5	6.6	4.23
Social Interaction	8.98	6.64	4.6	3.5
Repetitive Behaviour	4.6	3.5	2.6	1.77

H2



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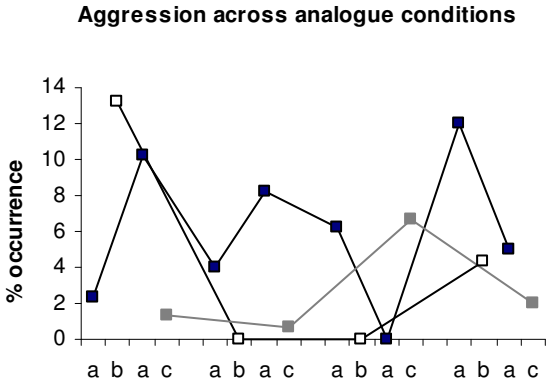

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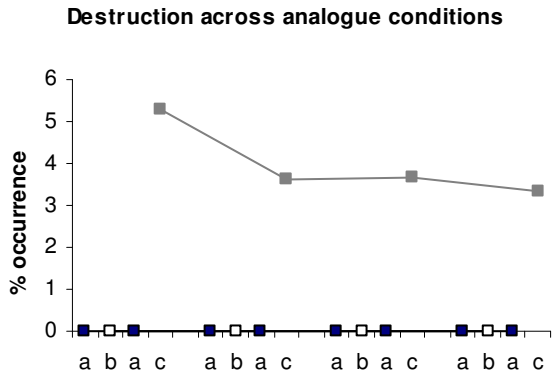

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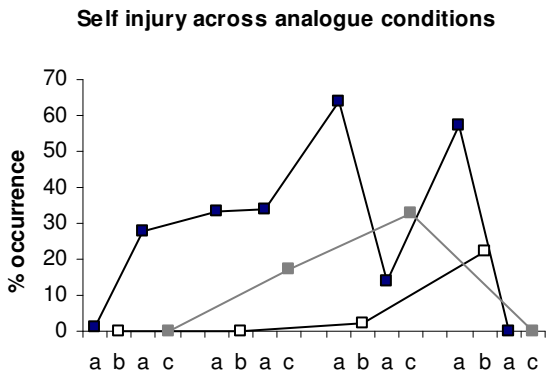

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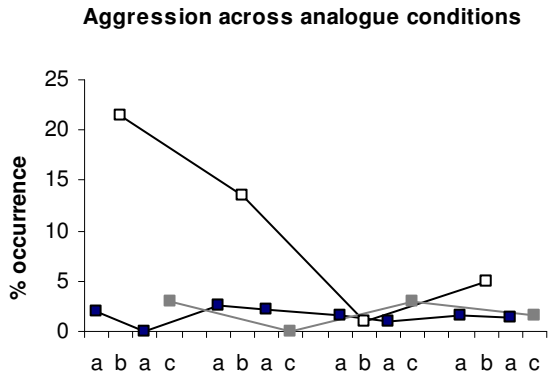

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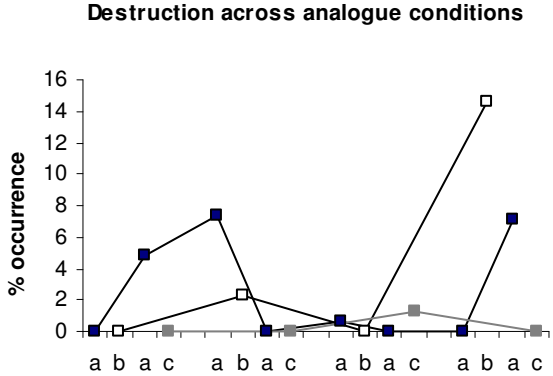

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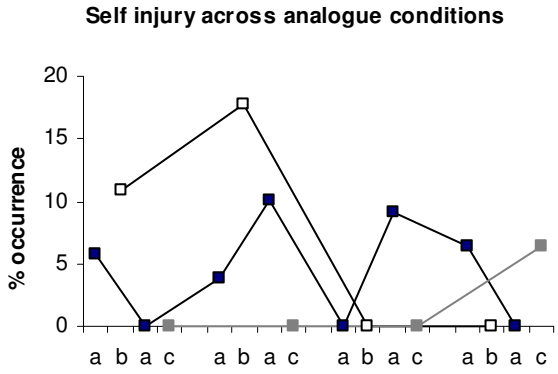

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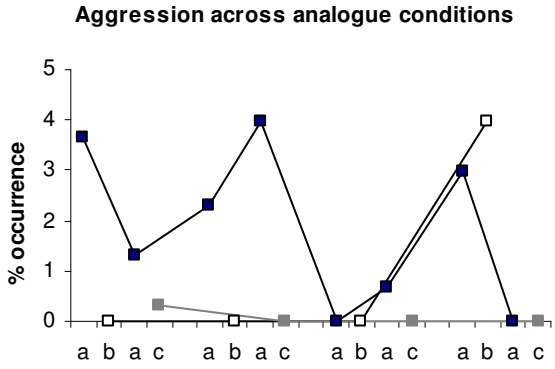

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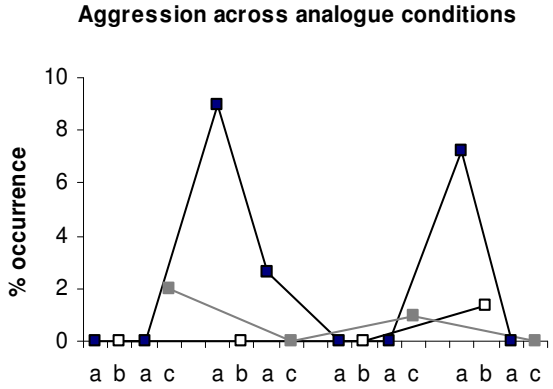

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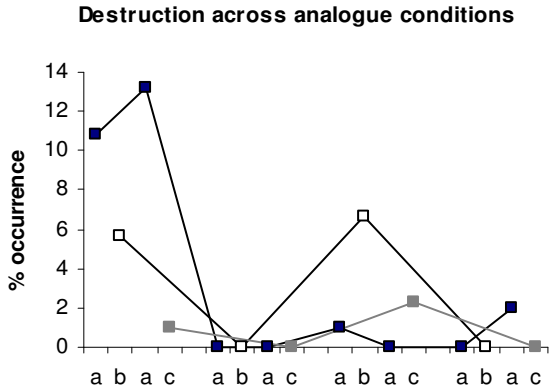

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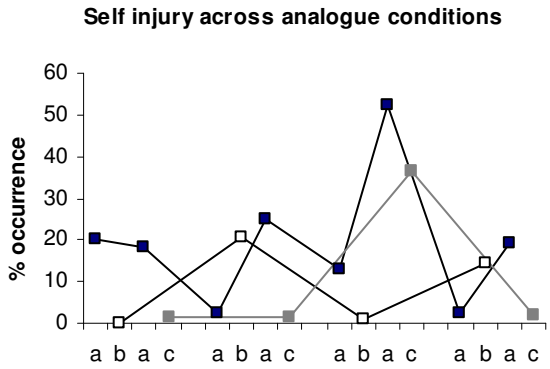

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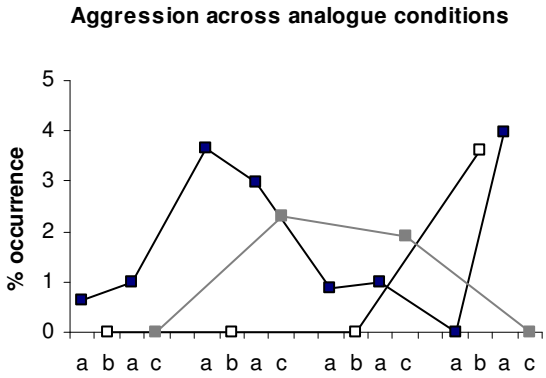

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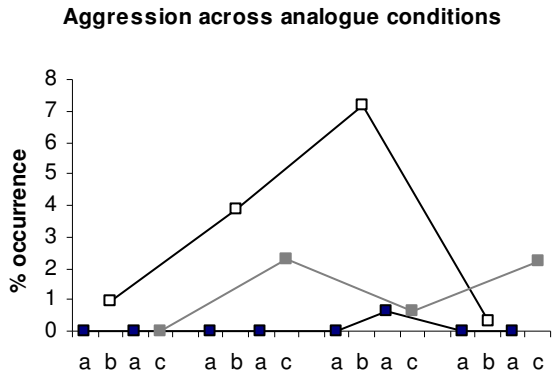

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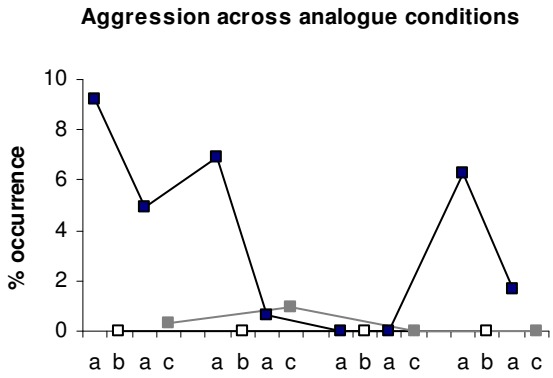

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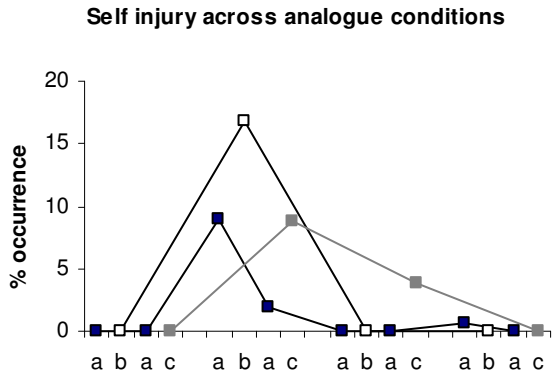

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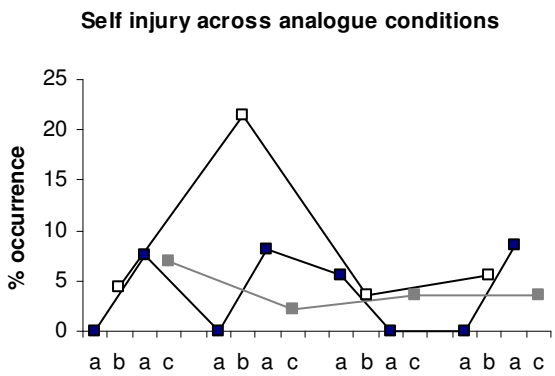

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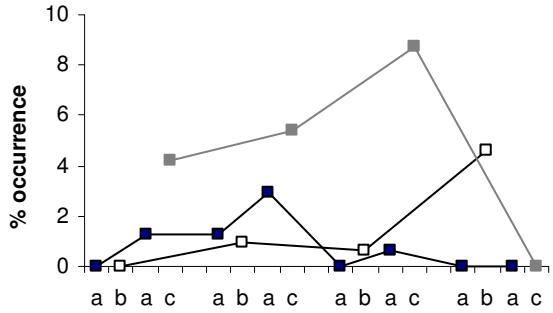

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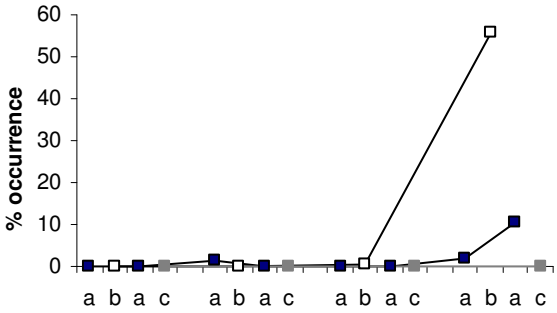

Table of median, inter-quartile range and Mann Whitney U analysis for approach and positive affect behaviours shown by the high and low risk groups across experimental functional analysis conditions

Variables	Condition	HR	LR	U	*p*
Approach	High attention	19.03 (40.83)	34.32 (37.92)	79	.174
	Low attention	6.78 (19.16)	4.88 (16.1)	102	.683
	Demand	2.73 (9.02)	5.17 (11.05)	97.5	.539
Positive Affect	High attention	7.71 (13.76)	12.63 (20.94)	106	.806
	Low attention	3.8 (6.8)	.82 (9.1)	96.5	.512
	Demand	.58 (6.53)	.17 (11.36)	111.5	.967