

VOLUME I

RESEARCH COMPONENT

LITERATURE REVIEW AND EMPIRICAL PAPER

**THE CORRELATES OF CHALLENGING BEHAVIOUR IN
INTELLECTUAL DISABILITIES: CHILD CHARACTERISTICS,
PARENTAL COGNITIONS AND WELLBEING.**

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DEDICATION

For Lee

OVERVIEW

This thesis consists of two volumes that are submitted by Jane Waite as part of the Clinical Psychology Doctorate, University of Birmingham. The first volume includes two papers prepared for publication in *Research in Developmental Disabilities* and the *Journal of Applied Research in Intellectual Disabilities* respectively. The first paper is a literature review integrating research on well-being in parents of individuals with intellectual disabilities and research on correlates of challenging behaviour. The second paper is an empirical study investigating parental perceptions of self-injury, aggression and destruction of property, and associations with child characteristics. In addition, a public domain briefing document is included in Volume I that summarises these papers in an accessible format.

Volume II of this thesis consists of four clinical practice reports and an abstract for an oral presentation presented in place of a written report. The first report describes the assessment of a fifty year old woman with depression and a mild intellectual disability. Formulations are presented from a cognitive-behavioural and systemic perspective. The second report describes a service evaluation that was conducted to gather information about service users' experience of an intellectual disability service. This report presents recommendations for how the service can be developed to support clients with profound intellectual disabilities. The third report describes the assessment, formulation and intervention of generalised anxiety disorder in a thirty-five year old woman. A functional analysis of a twelve year old girl with repetitive screaming is presented in report four. Finally, the abstract for report five describes a service evaluation of health professionals' knowledge of older family carers' needs, and the influence of this knowledge on their practice.

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I would like to acknowledge my supervisor, Chris Oliver, for his support and encouragement throughout this project. Chris has continued to help me to broaden my research interests and develop my ability to critique research. Furthermore, his enthusiasm for the project has been a huge source of motivation during the preparation of this thesis.

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Thank you to Jo Moss, Kate Arron, Cheryl Burbidge and Katy Berg who set up the original cross syndrome database that allowed selection of participants who had been engaging in challenging behaviour over a number of years. In addition, a large number of individuals have contributed to the collection of data over the last 10 years, in particular, Jo Moss, Jen Cullen, Gemma Griffith, Sarah Gorniak, Caroline Richards, Lisa Nelson, Lucy Wilde, Kate Eden and Chris Stinton. The analysis in the empirical paper includes data collected during a 2011 follow-up study and I would like to acknowledge Hayley Mace, Lisa Cochran, Gursharan Barth, Leah Bull, Mary Heald, Natalie Jackson, Jessica Penhallow and Lucy James for their contributions to the collection of data for Fragile-X, Cornelia de Lange, Prader-Willi, Angelman, Lowe, Cri du Chat, Smith-Magenis syndromes. In addition, I would like to thank Sarah Cassidy for her input with Limesurvey.

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Parental stress and cognitions: the role of behavioural dysregulation, self-injury, aggression and destruction of property

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Formatted for Research in Developmental Disabilities

Abstract

Hastings proposed a bi-directional model of parental stress and challenging behaviour (e.g. self-injury, aggression and property destruction) in individuals with intellectual disability. This model is supported by evidence linking parental well-being to child behavioural difficulties, however, research studies that have explored this link have not routinely differentiated between behaviours such as self-injury, aggression and property destruction and behaviours associated with underlying behavioural dysregulation such as impulsivity, over-activity and repetitive behaviour. In this review, research exploring behavioural dysregulation, self-injury, aggression and property destruction in individuals with intellectual disabilities is presented along with broader research on behavioural dysregulation. It is argued that behavioural dysregulation is a correlate of self-injury and aggression and might be linked to underlying executive function impairments that are noted in people with intellectual disabilities. It is proposed that behavioural dysregulation could influence parental perceptions of self-injury, aggression and property destruction and lead to increased parental stress which subsequently affects parental responses to these behaviours. An adapted model of parental stress and challenging behaviour is proposed that incorporates behavioural dysregulation factors.

Highlights:

- Challenging behaviour composite scores might mask associations with parental variables
- Behavioural dysregulation is associated with self-injury, aggression and property destruction
- Behavioural dysregulation can be incorporated into a model of parental stress

Keywords: Challenging behaviour, impulsivity, over-activity, hyper-activity, parental perceptions, stress, well-being.

1.1. Introduction

Aggression, self-injurious behaviour and property destruction shown by children with intellectual disabilities (ID) have been linked to poorer well-being in parents (Hastings, 2002). Hastings (2002) proposed a model of how behaviours such as these might form a bi-directional relationship with parents' emotional reactions, cognitions and well-being. In this review, this relationship will be examined to explore whether an extension of the model to differentiate between behaviours such as self-injury, aggression, property destruction and a more general behavioural dysregulation, underpinned by executive function impairments, is warranted. In the review Hastings' (2002) bi-directional model will be presented followed by a critical examination of the use of umbrella term 'behaviour difficulties' in numerous studies. The challenging behaviour and executive functioning literature will then be examined to argue that specificity is required to differentiate self-injury, aggression and property destruction from behavioural dysregulation variables, such as over-activity, impulsivity and repetitive behaviour, because these variables might show different relationships with parental well-being. Finally, an extended bi-directional model of challenging behaviour and parental stress will be presented. A systematic review would not meet the over-arching aim of this review which requires an integrative approach to draw together a broad range of literature.

1.2. A Bi-directional Model of Challenging Behaviour and Parental Stress

Approximately 10-15% of children and adults with ID engage in behaviours perceived as challenging (Emerson et al., 2001). Emerson (2001) found that aggression, property destruction and self-injurious behaviour were amongst the most frequent challenging behaviours reported by parents. The prevalence of these behaviours is particularly high in genetic syndromes with associated ID (45-92% for self injury and 40-73.8% for aggression). Estimates vary according to syndrome group but are consistently elevated in comparison to individuals with ID of heterogeneous aetiology (Arron, Oliver, Moss, Berg & Burbidge, 2011). Evidence has accumulated that suggests behaviours such as self-injury, aggression and property destruction might be related to poorer parental well-being (Hastings, 2002; Hodapp, Fidler & Smith, 1998; Eisenhower, Baker & Blacher, 2005, Beck, Hastings, Daley & Stevenson, 2004). Hastings (2002) proposed a bi-directional model which integrates research on the maintenance of self-injury, aggression and property destruction with research on parental stress in relation to these behaviours. This is shown in figure 1.1.

In this model, parent behaviour is proposed to influence the maintenance of behaviours such as self-injury, aggression and property destruction through the process of operant reinforcement. This aspect of the model is supported by functional analytic studies that link operant reinforcement by

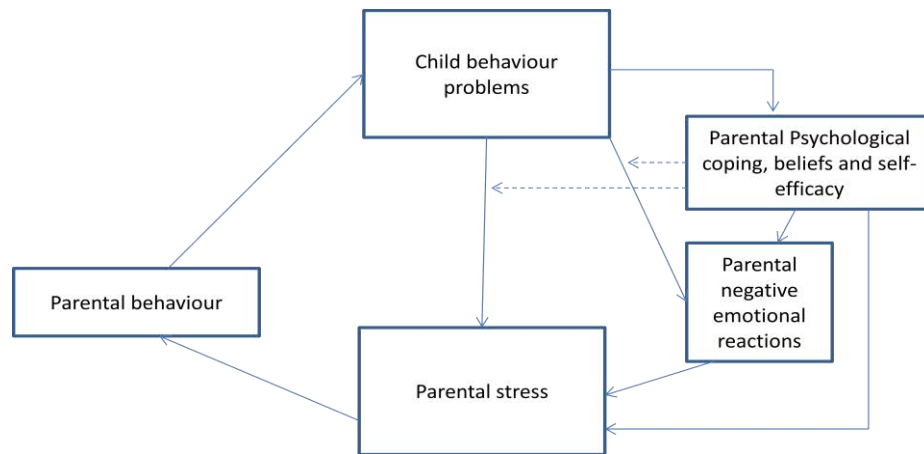


Figure 1.1. Hastings (2002) bi-directional model of parental stress and challenging behaviour

others to self-injury, aggression and property destruction (Carr & Durand, 1985; Grey & Hastings, 2005; Iwata, 1994). Operant reinforcement is effective at shaping behaviours that serve a function for an individual; either non-social (automatic sensory reinforcement) or social (escape from demands/access to attention), and a specific behaviour (e.g. skin picking) can serve different functions across individuals, or can differ in function depending on context (Emerson & Bromley, 1995; Matson, Bamburg, Cherry & Paclawskyj, 1999; Matson & Boisjoli, 2007). There is evidence that particular behaviours are more likely to serve particular functions, for example, self-injury is more often associated with non-social sensory reinforcement, and aggression with social reinforcement (Didden, Korzilius & Curfs, 2007; Emerson & Bromley, 1995; Rojahn, Zaja, Turygin, Moore, van Ingen, 2012).

Hastings (2002) promoted a broader perspective when considering how self-injury, aggression and destruction might be maintained. In agreement with Oliver (1995) the model proposes that parental responses to these behaviours may be influenced by stress and their emotional reactions to them. Thus, to escape from the aversive experience of self-injury, aggression or property destruction, parents adopt strategies which inadvertently reinforce these behaviours, and this increases the likelihood of parents responding in the same way in the future as the aversive experience is temporarily removed. Additionally, parental cognitive variables are hypothesised to moderate or mediate relationships between parental emotional responses and behaviours such as self-injury, aggression or property destruction (Hastings, 2002). The model is focused in that it does not include genetic, biological or neurological factors, although Hastings (2002) acknowledges that these factors are also likely to impact on the stated associations.

Hastings (2002) devised this model as a framework for further research rather than a robust synthesis of current evidence, as research for particular components of the model is limited. As discussed, an underlying premise of this model is an operant account of self-injury, aggression and

destruction of property. However, in the following section studies that are often cited as evidence for a link between parental well-being and challenging behaviours (self-injury, aggression and property destruction) are examined and it is argued that measurement of 'behavioural difficulties' in these studies is often confounded by inclusion of variables for which an operant interpretation is not established.

1.3. The challenge of 'Challenging Behaviour': What is wrong with an umbrella term?

The literature on parental stress/well-being and behaviour problems in ID is too vast to review systematically here. However, to illustrate methodological issues evident in the literature that influence interpretation, a search was conducted for review articles of parental well-being and ID since 2000¹ (see Table 1.1.). The majority of reviews returned by this search were not systematic and focused on specific questions, for example, examining models of coping behaviour, rather than taking an inclusive approach to reviewing factors associated with parental well-being. While the use of individual unsystematic reviews is likely to increase bias, examining a broad range of reviews provides a snap-shot of the current position of the wider literature.

The review articles were examined for cited studies of associations between 'behaviour difficulties' and parental stress or well-being. The quality of the measures used in these cited studies was then examined by recording: a) whether an ID specific measure of behaviour was used, b) whether there was specificity in the analysis between parental well-being and child behaviour (i.e. the type of behaviour was specified rather than using a 'composite score' c) whether behaviours associated with 'behavioural dysregulation' such as over-activity, hyper-activity, repetitive behaviour and impulsivity were measured independently of self-injury, aggression and property destruction.

It is clear that in the majority of these studies 'behavioural problems' are quantified largely using composite behaviour scores i.e. composite scores from the Adaptive Behavior Scales: Resident and Community-II, or broad subscales such as 'externalising' and 'internalising' behaviours (e.g. Baker, Blacher, Crnic & Edelbrock, 2002; Eisenhower, Baker & Blacher, 2005; Pruchno, Patrick & Burant, 1996). Externalising difficulties refer to behaviours such as aggression and hyperactivity, whereas examples of internalising problems are depression and anxiety (Achenbach & Rescorla, 2001).

¹ Search strategy: Search conducted in Jan 2012 in psyc-info, medline. A hand search in google scholar was conducted. Search period from Jan 2000 to obtain most up to date reviews. Limits set to: peer-reviewed articles, review articles and articles in published in English. Level one search terms: stress, well-being, depression, anxiety, adjustment, coping, distress; Level 2: mother*, father*, maternal, paternal, family, parent, carers, care-givers; Level 3: intellectual disabili*, developmental disabili*, mental retardation, learning disabili*. 29 articles returned. Articles discarded if irrelevant to subject area or no relevant challenging behaviour/parental well-being papers cited. One systematic review article was excluded because it listed all papers at the beginning of the article, merged 'behavioural difficulties' and ASD characteristics (communication and social impairments, and repetitive behaviour) into one category, and then presented percentages of papers showing a link between this category and parental well-being. This made it difficult to identify specific papers and hand searching all papers was not possible within the remit of this review.

Table 1.1.

A snapshot of the parental well-being and 'behaviour problems' literature. Papers cited within reviews as providing evidence of a link between 'behavioural difficulties' and parental well-being were examined to ascertain the specificity of the measure of behavioural difficulties.

Review Article (from most recent)	Purpose of Review	Type of Review	Cited 'behavioural difficulties' and Parental Well-Being papers	Degree of specificity cited papers
Serrata (2012)	Psychosocial aspects of parenting a child with ASD	Selective	Davis and Carter (2008)	-+-
Karst, Van Hecke and Vaughan (2012)	Proposed model for parenting well-being in ASD	Selective review to support model of intervention evaluation	Carter, Martinez-Pedraza and Gray (2009) Gray (1994) Hastings and Brown (2002) Hastings (2003) Herring et al. (2006) Lecavalier, Leone and Wiltz (2006) Lee, Harrington, Louie and Newschaffer (2008) Little and Clark (2006) Meltzer (2011) Sharpley, Bitsika and Efremidis. (1997) Tomanik, Harris and Hawkins (2004)	-+- --- +-- +-- +-- +++ --- --- +-- --- +++
Esbensen (2011)	ID, behaviour problems and co-morbid mental health problems - impact on parents	Selective	Baker, Blacher, Crnic and Edelbrock (2002) Baker, McIntyre, Blacher, Crnic Edelbrock and Low (2003) Eisenhower, Baker and Blacher (2005) Esbensen, Seltzer and Greenberg (2006) Floyd and Gallager (1997) Floyd and Phillippe (1993) Miltiades and Pruchno (2001) McIntyre, Blacher and Baker (2002) Orsmond, Seltzer, Krauss and Hong (2003) Pruchno, Patrick, and Burant, (1997) Seltzer, Greenberg and Krauss, (1995)	--- ^a --- --- +-- --- --- +-- +-- +-- ++- +-- +--
Hill and Rose (2010)	Parenting stress models and application to parents of adults and ID	Systematic search for models of parenting stress	Hassall, Rose and MacDonald (2005) Minnes, Woodford and Passey (2007) Pruchno, Patrick and Burant (1996) Quine and Pahl (1991)	--- --- +-- +--
Miodrag and Hodapp (2010)	Chronic stressors of parents and health review	Selective	No additional papers cited	
Mancil, Boyd, Bedesem (2009)	Coping strategies in parents with ASD	Selective synthesis	Higgins, Bailey and Pearce (2005) Hutton and Caron (2005) Pakenham, Sohrnoff and Samios (2004)	+-- +-- ---

+++ criteria defined as: + ID specific measure of 'behaviour difficulties' used + behaviour reported beyond composite scores + any variable associated with behavioural dysregulation included as individual behaviour variable.

^a Included an emotional regulation composite (hyperactivity, inattentiveness and emotional reactivity).

Note. Cited journal articles only appears in the table once even if cited by multiple reviews.

Table 1.1. continued

A snapshot of the parental well-being and 'behaviour problems' literature. Papers cited within reviews as providing evidence of a link between 'behavioural difficulties' and parental well-being were examined to ascertain the specificity of the measure of behavioural difficulties.

Review Article	Purpose of Review	Type of Review	Cited 'behavioural difficulties' and Parental Well-Being papers	Degree of specificity
Hastings (2002)	Bi-directional model of parental stress and child behaviour	Selective review prior to proposing model.	Baxter, Cummins and Yiolitis (2000) Blacher, Shapiro, Lopez, Diaz and Fusco (1997) Donenberg and Baker (1993) Dumas, Wolf, Fisman and Culligan Floyd and Gallagher (1997) Hodapp, Dykens and Masino (1997) Hodapp, Fidler and Smith (1998) Keogh, Garnier, Bemheimere and Gattimore (2000) Konstantareas and Homatidis (1989) Orr, Cameron, Dobson and Day (1993) Quine and Pahl (1985) Sloper, Knussen, Turner and Cunningham (1991) Stores, Stores, Fellows and Buckley (1998)	--- --- --- --- --- -++ -++ --- -++ +-- +-- +++ ---
Hassall and Rose (2005)	Parental cognitions and adaption to caring for a child with ID	Selective	Friedrick, Wiltturner and Cohen (1985)	+--
Blacher, Neece and Paczkowski (2005)	Family functioning and ID	Selective	Abbeduto et al. (2004) Beck, Daley, Hastings and Stevenson (2004) White and Hastings (2004)	+-- --- +--
Hatton and Emerson (2003)	ID: Stress and impact. One year review.	No search strategy described	No additional papers cited	
Boyd (2002)	Predictors of stress and coping in mothers of ASD	Selective	Factor, Perry and Freeman (1990)	---
Blacher and Hatton (2001)	One year review of family stress and burden	Selective	Feldman, Hancock, Rielly, Minnes and Cairns (2000)	---
Chan and Sigafoos (2000)	Correlates of respite and impact on parental stress	Selective	Hoare, Harris, Jackson and Kerley (1998) Treneman, Corkery, Dowdney and Hammond (1997)	+-- +--

+++ criteria define as: + ID specific measure of 'behaviour difficulties' used + behaviour reported beyond composite/total scores + any variable associated with behavioural dysregulation included as individual behaviour variable.

Note. Cited journal articles only appears in the table once even if cited by multiple reviews.

Note. A small number of articles were excluded because, when examined, it was found that the studies measured ASD symptoms (communication, socialisation and repetitive behaviour) but measuring 'behavioural difficulties' was not an aim of the study.

Therefore, specific behaviours that are linked to operant reinforcement, such as aggression, self injury and property destruction, are grouped together with additional child characteristics such as impulsivity, hyperactivity, repetitive behaviour. It may be that one of these factors is more influential on parental well-being than the other, or that they interact; however, without a fine-grained approach to studying self-injury, aggression and property destruction these conclusions cannot be drawn (Lecavalier, Leone & Wiltz, 2006). In addition, as Lecavalier et al. (2006) have noted, a high proportion of studies discern composite scores from questionnaire measures that are not ID specific such as the Child Behavior Checklist or the Vineland Adaptive Behavior Scales: Maladaptive Index (VABS) (e.g. Baker, McIntyre, Blacher & Crnic, 2002; Minnes, Woodford & Passey, 2007). This is particularly problematic as these measures were not developed with a focus on self-injury, property destruction and aggression as they are understood in the ID literature.

Even when ID specific measures are used at subscale level, the measure might not capture the specific challenging behaviours most frequently noted as problematic. For example, a measure such as the Aberrant Behavior Checklist (e.g. used by Estes, Munson, Dawson, Koehler, Zhou & Abbott, 2009) includes subscales: (1) irritability, agitation and crying, (2) lethargy, social withdrawal (3) stereotypic behaviours, (4) hyperactivity/non-compliance and (5) inappropriate speech. Therefore, it does not explore behaviours associated with operant reinforcement: self-injury, aggression, property destruction, specifically. The Developmental Behavior Checklist overcomes many of these issues, however, self-injury is still grouped with repetitive behaviour (e.g. Hastings, 2003).

If studies have included individual topographies of 'behaviour difficulties' in the analysis the inclusion of these variables appears to be driven by the structure of the measures rather than theoretical considerations about self-injury, aggression and property destruction and their correlates. In addition to a variable such as 'conduct difficulties', studies may include one or two isolated child characteristic variables such as hyperactivity or stereotyped behaviour (e.g. Konstantareas & Homatidis, 1989; Hodapp, Dykens & Masino, 1997; Lecavalier et al., 2006). However, as will be discussed in the next section, factors such as over-activity, impulsiveness and repetitive behaviour are consistently related to behaviours such as self-injury, aggression and property destruction, and may be indicative of underlying behavioural dysregulation in people with ID (Oliver, Sloneem, Hall & Arron, 2009). Additionally, there is limited evidence that behaviours indicative of behaviour dysregulation are mediated by social operant processes in the same way as self-injury, aggression and property destruction. Therefore, studies that only measure behaviour dysregulation partially might fail to adequately measure additional factors that contribute to parental well-being, either independently from self-injury, destruction and aggression, or as mediating/moderating variables.

In conclusion, there is little specificity when measuring self-injury, aggression and property destruction in the parental stress literature and composite scores of behaviour difficulties are often used. These scores combine behaviours typically associated with operant reinforcement, self-injury, aggression and property destruction with a range of other behaviours such as hyper-activity, repetitive behaviours and attention deficits or characteristics associated with autism spectrum disorder (ASD). The contribution of these individual factors is frequently ignored. An important question is whether this matters when building models of parental stress and challenging behaviour. The correlates of self-injury, aggression and property destruction will now be discussed to argue that behavioural dysregulation might contribute to the development of these behaviours and that dysregulation may be related to compromised executive function. It will be argued that incorporating behavioural dysregulation into a model of parental stress and challenging behaviour may lead to different hypotheses being generated from this model.

1.4. Child Factors and Challenging Behaviour²

Child factors that have been related to aggression, self-injury and property destruction include impaired communication, autism spectrum disorder or some genetic disorders, repetitive behaviours and restricted interests, limited mobility, visual impairments, a more severe degree of disability, and traits associated with attention deficit hyperactivity disorder (ADHD) (Cooper et al., 2009; Danquah et al., 2009; Matson, Boisjoli & Mahan, 2009; McClintock, Hall & Oliver, 2003; Oliver, Petty, Ruddick & Bacarese-Hamilton, 2012; Rojahn, Matson, Naglieri and Mayville, 2004).

Factors such as ADHD warrant further attention due to accumulating evidence across research disciplines highlighting an association with aggression. ADHD is a developmental disorder defined by behavioural dysregulation (deficits in attention, hyperactivity and impulse control) (DSM-IV; American Psychiatric Association, 1994). Children with ADHD are more likely to engage in aggressive behaviour than TD comparison groups (Connor, Chartier, Preen, & Kaplan, 2010; Hay, Hudson & Liang, 2010). Neece, Baker, Crnic and Blacher (2012) have recently demonstrated that adolescents with ID are at increased risk of ADHD (3.38:1) and that ADHD is a valid diagnosis for ID populations. Young (2002) argued that many people with ID would meet the diagnostic criteria for ADHD but that due to diagnostic over-shadowing ADHD is often missed. While Young makes a

² While this review was conceptual, a search was conducted in Ovid Psyc-Info and Medline to ensure literature in the following two sections presented an unbiased review. The search strategy was as follows: Level 1: ADHD, impulsivity, impulsiveness, hyperactive, hyper-activity. Level 2: Challenging behavio*, aggression, aggressive behaviour, destruction, destructive behaviour, self injury, self injurious behaviour. Level 3: intellectual disabilit*, mental retardation, learning disabilit*. Search was limited to peer-reviewed articles. Search was conducted January 2013. Search from 1987. 190 and 284 articles returned for Psyc-Info and Medline respectively. Articles were discarded if they did not directly explore association between behavioural dysregulation and degree of challenging behaviours (i.e. aggression, self-injury and property destruction).

valid point that behavioural dysregulation might be overlooked in ID, it should be noted that ADHD as a diagnostic category may not be the most informative way of thinking about behavioural dysregulation in ID because it assumes an integrated and unified disorder. However, as will be described in this review, characteristics of behavioural dysregulation might present differentially across genetic syndromes and a more informative approach to understanding behaviour dysregulation might be to explore shared cognitive and neurobiological pathways that lead to the manifestation of behaviours such as over-activity and impulsivity in a range of disorders.

Over the last ten years further evidence has accumulated that impulse control, ADHD characteristics, or general behaviour dysregulation, are linked to self-injury, property destruction and aggression in individuals with ID. Impulse control has recently been recognised as a correlate of challenging behaviour in a number of review articles (Furniss & Biswas, 2012; Gosalakal, 2003; Petty & Oliver, 2005; Oliver & Richards, 2010; Tunnicliffe & Oliver, 2011).

Several studies have linked behavioural dysregulation, such as impulsivity, over-activity and repetitive behaviour, to aggression and self-injury. Fee, Matson, Moore and Benavidez (1999) found comparable levels of aggression between children with ID and those with ID who fell above a cut-off for ADHD. Despite the absence of differences between these groups, over-activity and impulsivity were correlated with aggression in both groups. Rojahn et al., (2004) used the Diagnostic Assessment for the Severely Handicapped-II (DASH-II) and the Behavior Problems Inventory (BPI) to assess people with profound to severe ID (N = 180). Greater impulsivity and conduct difficulties were associated with self-injury and aggression/destruction, and individuals with *severe* self-injury were over twice as likely to have impulse control problems. In contrast to Rojahn et al. (2004), Crocker, Mercier, Allaire and Roy (2007) found that individuals that displayed the most topographies of aggressive behaviour at the highest severity were more likely to have a physical disability, a psychiatric disorder (Axis I) and less engagement in social and vocational pursuits. However, in agreement with Crocker et al. (2007), they were also most likely to score highly on the Barrett Impulsiveness Scale (BIS-11; Patton & Stanford., 1995). Similarly, Cooper et al. (2009) demonstrated that lower ability level, not living with a family carer, having ADHD, not having Down syndrome and visual impairment were associated with self-injurious behaviour. ADHD was the second strongest association after ability level. Finally, Matson, Mahan, Sipes and Koziowski (2010) explored whether associations between these variables were present in young children (17-36 months) with ID of mixed aetiology without a diagnosis of ASD. In accordance with other studies a link was established between higher degrees of inattention/impulsivity, aggressive and self-injurious behaviours and stereotypy. The finding that repetitive behaviour such as stereotypy is linked to aggression has recently been replicated in a cross-sectional questionnaire study of people with severe ID (Oliver et al., 2012). In particular, individuals who engaged in repetitive behaviour were sixteen times more likely to engage

in severe self-injurious behaviour.

A strength of studies that have explored the link between behavioural dysregulation variables and challenging behaviours such as self injury and aggression is that they adopted a fine-grained approach to studying correlates of self-injury and aggression. Therefore, they differentiated child characteristics into traits such as impulsivity, stereotypy and psychiatric conditions, such as ADHD, rather than grouping child behavioural difficulties into one composite score. However, a weakness is the use of correlational designs when considering the development or maintenance of challenging behaviour over time.

Davies (2010) conducted one of the few studies with a longitudinal design. Associations were explored between child characteristics and challenging behaviours such self injury, aggression a property destruction using a sophisticated longitudinal questionnaire study. Challenging behaviour was measured at two time points separated by 18 months. Children who engaged in the most repetitive, restricted, overactive and impulsive behaviour were most likely to be engaging in challenging behaviour at follow-up. Observations showed that the majority of challenging behaviour was functional. This study demonstrates how individual characteristics such as impulsivity may interact with environmental contingencies to maintain behaviours such as self-injury and aggression.

1.5. Behavioural Dysregulation and Challenging Behaviour in Genetic Syndromes

Self-injury, aggression and property destruction is more prevalent in certain genetic syndromes. It is of interest that impulsivity and hyperactivity are frequently reported in syndrome groups where these behaviours are present. Basile, Villa, Selicorni and Molteni (2007) employed direct cognitive assessment, the Vineland Adaptive Behaviour Scales-II (VABS-II) and a range of behavioural measures to explore the phenotype of CdLS (N =56). 59% of participants showed hyperactivity, 86% attention disorders, 46% compulsive disorders, 36% had self-injurious behaviour and 30% engaged in aggression. Self-injury was related to classic CdLS phenotype, ASD characteristics, compulsive behaviours, communication skills and pain threshold.

Arron et al. (2011) found associations between self-injury, repetitive behaviour, over-activity and impulsivity in Cornelia de Lange Syndrome, Fragile-X syndrome, Prader-Willi Syndrome, and Lowe Syndrome. One weaknesses of the above study is that a matched ability control group was not included. Therefore, it is not possible to estimate accurately how much greater self-injury is in these syndromes than people with ID of unknown etiology. In an earlier study of CdLS, Oliver et al. (2009) compared individuals with CdLS (N = 54) to individuals with ID of mixed aetiology (N = 46). When age, gender, mobility and ability level were controlled there were no differences in the prevalence of self-injury. A binary logistic regression showed that compulsive behaviour, hyperactivity, stereotyped behaviour predicted self-injurious behaviour and all three variables were found to load onto one factor

that was named 'behaviour dysregulation,' which accounted for 55.93% of the variance. Multiple regression, conducted with behavioural deregulation as the dependent variable, revealed that wheelchair use and ASD contributed significantly to the model. Oliver et al. (2009) argued that a number of correlates of self-injury may be present in CdLS that might increase the likelihood of the presence of self-injury, rather than self-injury being part of the phenotype of CdLS specifically. The 'behaviour dysregulation' factor derived in this study fits with the findings of Burbidge et al. (2011) who found over-activity was associated with stereotypy and that impulsivity was related to restricted preferences in individuals with severe intellectual disability.

ASD is an example of another developmental disorder where these associations have been noted. Richards, Oliver, Nelson and Moss (2012) found that in ASD self-injurious behaviour was related to impulsivity, hyperactivity, lower ability and speech levels. This link between impulsivity and self-injury has been replicated by Richman et al. (2012) who explored correlates of self-injurious behaviour in a large sample (N = 617) of individuals with ASD. Structural equation modelling revealed that impulsivity and stereotyped behaviour, as measured by the Aberrant Behavior Checklist, predicted self-injury even after cognitive ability and ASD symptoms were controlled for. The strengths of this study are its sample size, the use of the Autism Diagnostic Behavior Schedule and the use of structural equation modelling. Repetitive behaviour has also been linked to aggression in a large sample of children and adolescents with ASD (N = 1380) even when degree of ASD, level of adaptive ability and communication skills were controlled (Kanne & Mazurek, 2011). This link between repetitive behaviour and aggression is of interest due to the co-occurrence of repetitive behaviour with impulsivity and over-activity, and evidence that these factors might load onto one factor: behavioural dysregulation (Oliver et al., 2009). In addition, aggression has been linked to behavioural dysregulation in Smith-Magenis syndrome (SMS). Sloneem, Oliver, Udwin and Woodcock (2011) demonstrated that individuals with SMS were more likely to engage in more severe aggression if they were rated as having high impulsivity. They found that aggression was more likely to be related to environmental contingencies in this syndrome. This replicates Davies' (2010) findings of a potential inter-play between child characteristics and environmental reinforcement.

Finally, evidence for a link between reduced behavioural control and self-injury comes from research studies on self-restraint. Self-restraint is associated with self-injurious behaviour in some individuals with genetic syndromes. For example, Hyman, Oliver and Hall (2002) sampled 88 individuals with Cornelia de Lange syndrome as part of a questionnaire study exploring associations between self-injury, restraint and compulsive behaviours. Individuals were more likely to engage in restraint behaviours if they engaged in self-injury and had higher levels of compulsive behaviour. Hyman et al (2002) acknowledged that this does not prove a direct relationship between self-restraint and self injury; however, it does lend support for theories that suggest some self-injurious behaviour

might have a compulsive quality indicative of a lack of behavioural control (King, 1993). A link between self-restraint and self injury has also been observed with people with ID without genetic syndromes as well as a link between compulsive behaviour and self-injury (Bodfish, Crawford, Powell, Golden & Lewis, 1995; Buzas, Ayllon & Collins, 1981; Christie et al., 1982; Dossetor, Couryer & Nicol, 1991).

In summary, numerous studies suggest links between behavioural dysregulation (i.e. impulsivity, over-activity) and the presence of self-injury, aggression and property destruction. The majority of these studies were correlational parental report studies, which raises uncertainty about causality, the direction of results and the impact of parental cognitive bias (high reporting across all measures). However, longitudinal studies and studies employing statistical modelling support these factors as predictors of self-injury, aggression and property destruction. This relationship has been noted in individuals with ID of varying degrees of ability and it has been noted across a number of syndrome groups. This suggests that factors falling under the umbrella term 'behavioural dysregulation' may have a role in the development and maintenance of challenging behaviour.

1.6. Executive Function – A Unifying Factor

Behavioural dysregulation as a correlate of self-injury, aggression and property destruction is particularly pertinent given that behavioural dysregulation might indicate underlying cognitive difficulties as opposed to being explained by an operant account of behaviour. Impulsivity and over-activity in individuals with ID could indicate executive functioning deficits (EF) as has been shown in individuals without ID (Brocki & Bohlin, 2006, Barkley, 1997), particularly as EF deficits have been documented in people with ID (Willner, Bailey, Parry & Dymond, 2010).

EF refers to a set of higher cognitive processes, associated with the frontal lobes, that control and regulate behaviour (Suchy, 2009). EF is an umbrella term that includes constructs such as inhibition, working memory and task-switching (Garon, Bryson & Smith., 2008; Miyake et al., 2000). Damage to areas in the frontal lobes can give rise to difficulties with cognition and emotion that may typically be labelled as impulsive or dysregulated. This might include reacting 'automatically' to stimuli in the environment without consideration of the outcome, or failure to stop a behaviour once it has been initiated (Luria, Homskaya, Blinkov & Critchley, 1967; Luria, 1973; MacMillan, 2000).

A link between impulsivity, over-activity and EF is supported by research into the cognitive underpinnings of ADHD. Brocki and Bohlin (2006) investigated how developmental changes in executive functioning impact on impulsivity and hyperactivity in children aged 6-15 years with a diagnosis of ADHD. They found that in younger children poor inhibition is related to ADHD symptomatology. In addition, Nigg (2001) noted a link between motor dysinhibition and ADHD symptoms. ADHD has increasingly been understood as an EF disorder (Barkley, 1997, Gorenstein &

Newman, 1980; Pennington & Ozonoff, 1996; Schachar & Logan, 1990). Willcutt, Doyle, Nigg, Faraone and Pennington (2005), in a meta-review of literature on the relationship between EF deficits and ADHD, argued that EF was supported as a key component of ADHD, although it was likely to interact with other factors.

EF as a unifying underlying factor or cognitive account of behavioural dysregulation is consistent with research that has identified impulsivity and over-activity as correlates of self-injury and aggression. It also fits with findings that impulsivity and over-activity are linked to repetitive behaviours and ASD phenomenology in individuals with ID (Burbidge et al., 2010, Furniss et al., 2010; Emerson, 2001; Rojahn et al., 2004; Matson, Mahan & Fodstad, 2010; Matson & Rivert 2008; Oliver et al., 2012). Repetitive behaviour could be viewed as another behavioural dysregulation factor, not only because of this link with impulsivity, over-activity, and the presence of self-injurious and aggressive behaviours, but because it has been associated with executive functioning deficits in a number of disorder groups (Oliver et al., 2012; Turner, 1997, 1999; Lopez, Lincoln, Ozonoff & Lai, 2005).

Turner (1997) found a link between EF deficits and repetitive behaviour in ASD. For example, restricted interests and adherence to routines were linked to poorer performance on a set-shifting card task relative to controls. In addition, repetitive stereotyped behaviours were associated with perseveration to one location when participants had to guess where a target would appear from a choice of two locations, and perseveration on word generation tasks (Turner, 1997; 1999b). Turner (1997) argued that this perseveration was related to inhibitory control and set-shifting deficits; as well as difficulties generating alternative responses. Lopez et al. (2005) found a similar pattern of results: poorer inhibition was related to repetitive behaviour in high functioning individuals with ASD even when ability level was controlled for.

It is of interest that high levels of impulsivity, hyper-activity and repetitive behaviour are found in ASD relative to individuals with ID of heterogeneous aetiology, and that this co-occurs with self-injury, aggression and property destruction, and EF deficits (Turner, 1997; 1999). Thus, in ASD behavioural dysregulation variables associated with ADHD are often apparent and may point towards commonalities in aetiology of behavioural dysregulation. This is supported by Nydén et al. (2010) who found that high functioning adults with ASD, ADHD and ASD/ADHD had very similar profiles of deficits on a range of cognitive tests, including tests of executive functioning. Nydén et al (2010) argued for a 'continuum of neuropsychological dysfunction' as opposed to separating individuals based on diagnostic categories. The argument for common underlying aetiology in these disorders has been reiterated in a recent review that presented evidence that ADHD is often co-morbid with ASD, that there are genetic commonalities, and that the two disorders may be variants of an underlying disorder (Rommelse, Geurts, Franke, Buitelaar & Hartman, 2011).

The links between executive functioning and repetitive behaviour are not limited to ASD suggesting that the commonalities in aetiology may be relevant to a broad range of genetic syndromes. Evidence of an association between repetitive behaviours and EF has also been found in Prader-Willi syndrome where repetitive questions and adherence to routines have been linked to set-shifting difficulties (Woodcock, Oliver & Humphreys, 2009) and from Rubinstein-Taybi syndrome where repetitive questioning has been linked to inhibition and working memory deficits (Waite, 2012). Outside of the ID literature, repetitive type behaviours in schizophrenia, obsessive compulsive disorder and dementia have also been linked to EF difficulties (Bradshaw, 2001; Cullen et al., 2005; Lawrence et al., 2006; Lysaker, Whitney and Davis, 2009).

In summary, links between cognition (EF) and behaviour dysregulation (impulsivity, hyper-activity, repetitive behaviour), and between behaviour dysregulation and self-injury, aggression and property destruction, are supported by numerous research studies and there is evidence that difficulties with EF correlate of a number of behavioural difficulties including aggression and impulsivity in children.

1.7. Neurobiological Underpinnings – Neural Networks and Neurotransmitters

An alternative but related argument to the EF account of ADHD and behaviour dysregulation is to view ADHD behaviours as arising from disruption to frontostriatal dopaminergic and noradrenergic systems (Bradshaw, 2001; Johnsen, Aase, Meyer & Sagvolden, 2000). Johansen, Aase, Meyer and Sagvolden (2000) argue that reduced dopamine impairs learning in ADHD by altering the reinforcement potential of external stimuli and that impulsivity and hyper-activity may result from children not learning to control their behaviour. This argument is relevant to self-injury, aggression and property destruction because it highlights how the success of operant social interventions could potentially be lower if behavioural dysregulation is also present. In this circumstance, too much emphasis on an operant social account of self-injury, aggression and property destruction without consideration of behavioural dysregulation might limit clinical formulation.

Changes to the reinforcement potential of stimuli is supported by observations that children diagnosed with ADHD tend to be less sensitive to small degrees of positive reinforcement, have shorter delay of reinforcement gradients, which means reinforcement needs to be delivered closer to behaviour to be effective, and be less sensitive to extinction (Sagvolden, Aase, Zeiner & Berger, 1998; Sagvolden & Archer, 1989). Given the dopaminergic mesolimbic systems project to the frontal lobe it is possible that this theory is not entirely incompatible with the executive function hypothesis of behaviour dysregulation, as it is likely that there will be inter-play between downstream biological processes and higher-order cognitive processes. However, it might be that dopaminergic mesolimbic processes can explain executive function deficits and behavioural manifestations without EF being

viewed as the causal mechanism.

A fully integrated theory of neurobiological and cognitive components has yet to be developed; however, overall this research points towards neurobiological disruption and cognitive deficits that are associated with poorer learning and less effective behavioural interventions. These associations fit with the failure of operant theory alone to account for all observations relating to self-injury, aggression and property destruction. For example, operant theory does not provide a satisfactory explanation for self-restraint, nor does it provide a satisfactory explanation for why repetitive behaviour, impulsivity and over-activity tend to cluster together with self-injury, aggression and property destruction. As Hastings (2002) acknowledged social-environmental factors cannot fully explain why behaviours such as self-injury and aggression are heightened in particular genetic syndromes even when degree of disability is controlled for.

Given reinforcing alternative behaviours might be less effective for families of children with behavioural dysregulation due to shorter reinforcement gradients, less sensitivity to rewards, or perseverative/compulsive behaviour, applying interventions based on operant reinforcement might be more stressful for these families because they experience that self-injury, aggression and property destruction are not effectively controlled by environmental manipulation alone. Furthermore, this experience might impact on parental cognitions about managing self-injury, aggression and property destruction and, therefore, behavioural dysregulation could moderate the stress parents experience while caring for a child with ID. This is largely speculative but it is important that these possibilities are not ignored. Furthermore, while self-injury, destruction and aggression are combined with correlates associated with behavioural dysregulation a confound exists that could limit understanding of these relationships with parental well-being. Evidence is now presented from a range of populations to illustrate that behavioural dysregulation is correlated with parental well-being. This is followed by an discussion of the impact of using non-specific composites of 'behavioural difficulties' when exploring relationships with parental well-being.

1.8. Behavioural Dysregulation and Parental Stress

The importance of the contribution of executive function deficits, associated with behavioural dysregulation, to parental stress independently from self-injury, aggression and destruction has been noted in non ID populations. Patel, Wong, Cuevas and Horn (2012) explored associations between EF deficits in child cancer survivors and parental outcomes. Parenting stress significantly correlated with three subscales of the Behaviour Rating Inventory of Executive Function BRIEF (inhibit, shift and emotional control) as well as the overall behaviour regulation index. Furthermore, on direct performance based measures, digit span total score (accounted for by the backwards digit span) had a small to medium correlation with stress. Executive function deficits accounted for an additional 17%

of parental stress once socio-demographic and clinical variables were controlled. ANOVA revealed a similar pattern of results with parental stress being elevated within the group who scored most highly on the BRIEF-P. This study is an example of how executive functioning can be linked to parental stress because it used a combination of parental report and direct assessment, which strengthens the conclusions by reducing concerns about informant bias. Similarly, Paley, O'Connor, Frankel & Marquardt (2006) utilised questionnaire methodology to explore the impact of executive functioning and other factors on parents' stress of caring for a child with Foetal Alcohol Syndrome. A range of factors were related independently to parents' stress including executive functioning deficits, poorer adaptive functioning, externalising and internalising problem behaviours and the parent being an adoptive parent. A strength of this study is that teacher ratings of executive functioning were associated with parental ratings, which suggests parents' ratings were not due to cognitive bias associated with poorer mental health.

In addition, Epstein, Saltzman-Benaiah, O'Hare, Goll and Tuck (2008) explored associations between child EF and stress in parents of children with Asperger's syndrome. Mothers (N = 37) and fathers (N = 24) completed questionnaire measures of stress, child sensory difficulties, and child EF. 78% of mothers and 60% of fathers rated their children as having significant difficulties with EF and sensory difficulties, and mothers' stress levels were strongly positively correlated with the global EF composite. This study adds further support for a link between behavioural dysregulation and stress in mothers. The absence of significant results in fathers may have been due to the small number of fathers or it may be that fathers have different experiences to mothers. The agreement between mothers and fathers on the degree of EF impairment reduces the likelihood that these results occurred due to cognitive bias about children's behaviour due to stress levels; however, the correlational nature of the design means the direction of the result cannot be established.

1.9. Wider Implications of Confounds in the Parental Stress Literature

As noted earlier in reference to Hastings's model, researchers have tried to understand variation in how parents manage and cope with behaviours such as self-injury, aggression and property destruction (Hastings, 2002). It has been argued that parental cognitive variables may mediate or moderate the impact of behaviour difficulties, for example, parental cognitions and attributions about behaviour. MacDonald, Hastings and Fitzsimons (2010) used a cross-sectional questionnaire study to explore an association between generic behavioural difficulties, measured using a composite score from the Strengths and Difficulties Questionnaire, and stress in fathers of children with ID. Psychological acceptance partially mediated this relationship. It was suggested that interventions could focus on changing fathers' attributions, for example, increasing psychological acceptance (MacDonald et al., 2010).

Attributions about challenging behaviour have also been suggested to moderate a relationship between degree of child disability and challenging behaviour. Woolfson, Taylor and Mooney (2011) conducted a questionnaire study with two groups of similar chronological age: parents of children with developmental disabilities and typically developing children. Associations between parental attributions, child behaviour difficulties and stress in parents of children were explored using the Child Behavior Checklist. Extent of aggression, rule-breaking, social problems and 'other' problems were examined. These behaviour difficulties were significantly higher in the developmental disorder group. Parental perceptions of low control over their children were associated with aggressive behaviour and rule breaking behaviour in the developmental disorder group. However, when parents had greater perceived control over their children the differences in degree of general 'behaviour problems' reduced between groups. It was argued that parental attributions of controllability might moderate the association between disability and behaviour.

A possible conclusion arising from studies that link controllability attributions to general 'behavioural difficulties' is that attributions of low controllability might impact on the development and maintenance of aggression. Indeed, there is evidence from studies with support workers that suggest attributions are related to the development and maintenance of aggression, self-injury and property destruction by influencing how they respond to behaviour (Dagnan, Trower & Smith, 1998; Jones & Hastings, 2003). However, research into the relationship between attributions and carer response in ID populations have shown mixed results. For example, in a recent review the attribution model was only partially supported in relation to care-giver helping (Wilner & Smith, 2008). Wilner and Smith (2008) argued that methodological issues may underpin the lack of consistency in the literature, but they also note that additional explanations need to be explored, including alternative theoretical perspectives.

A difficulty may arise from focusing too heavily on parental attributions and the impact on behaviour difficulties at the expense of understanding the impact of behavioural dysregulation on the development of parental attributions. For example, parents may have different attributions about their child's self-injury and aggression and failure to measure behavioural dysregulation variables or the use of challenging behaviour composite scores may mask subtle child behavioural characteristics that underpin these attributions. Thus, a parent whose child is very over-active and impulsive with some incidents of self-injury and aggression may have different attributions to parent of a child with high incidents of self injury and aggression but who is less over-active and impulsive. The child with elevated self-injury may respond well to a behavioural intervention relative to the child with higher levels of over-activity and impulsivity. The conclusion that parental attributions affected the success of the intervention may be erroneous, particularly given that the ADHD literature suggests that children with higher degrees of dysregulation respond less well to extinction and behavioural

modification techniques (Johansen et al., 2002). Hence, the parent of the more over-active child who perceived that their child's behaviour was likely to persist and be less controllable may be making an *accurate* appraisal of their child's behaviour. It is of interest that parents of children with ADHD have been found to hold attributions that fit with the neurobiological explanation for the disorder, namely, they feel behaviour is less controllable and feel less responsible for it and a similar pattern might be observed in parents of children with ID who have behavioural dysregulation characteristics (Johnston & Freeman, 1997). Examining behaviour at a fine grained level would allow for the individual contributions of behavioural dysregulation and behaviours such as self-injury, property destruction and aggression to be explored, and would improve the quality of the claims that can be made from the research.

1.10. An Extended Model of Challenging Behaviour and Parental Stress

Impulsive and hyperactive individuals are likely to be highly dysregulated across everyday activities meaning that parents need to provide a higher degree of monitoring and structure (Murray & Patel, 2001). The accumulating evidence suggests that when clinicians ask parents to conduct intervention strategies for aggression, self-injury and property destruction these parents might already be trying to manage a high degree of pervasive behaviour dysregulation in their children with ID. Carers of individuals with executive function deficits or impulsive dysregulated behaviour may be those individuals who are more likely to experience psychological distress (Harrison & Sofronoff, 2002). The combination of increased caring demands due to behavioural dysregulation and psychological distress might leave parents with fewer internal resources for seeking help and applying interventions, even when interventions are based on a strong theoretical basis. Furthermore, the if parents experience behaviours as difficult to modify with social operant interventions they may experience more stress and be less inclined to follow through with interventions.

An extended model is proposed to incorporate behavioural dysregulation and to operationalise child behaviour problems more clearly. Oliver et al. (2009) argued that challenging behaviour (i.e. self-injury) may persist when it comes into the behavioural repertoire against the background of general behavioural dysregulation. This argument is extended here by proposing that behavioural regulation may both influence parental psychological and emotional reactions, and contribute as a factor maintaining the presence of challenging behaviour (see figure 1.2.).

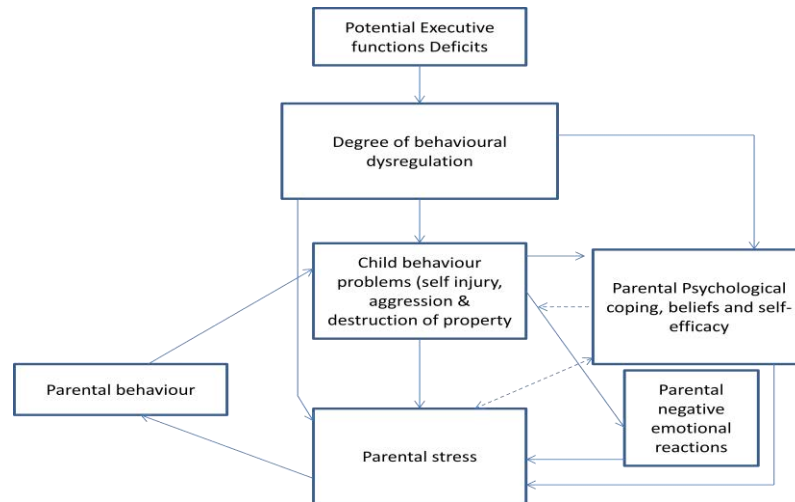


Figure 1.2. Adapted bi-directional model of parental stress, including executive functioning and behavioural dysregulation

1.11. Clinical Implications and Further Research

Further research should explore how parental cognitions and wellbeing relate differentially to child characteristics associated with behavioural dysregulation and self-injury, aggression and property destruction. It is important to understand these relationships as different intervention strategies might be suited to different behavioural difficulties, whether that is targeting parental perceptions or employing multi modal interventions. It should be noted that this extended model suggests a uni-directional pathway between behavioural dysregulation and parental cognitive and emotional factors. This fits with a view that behavioural dysregulation is related to the aetiology of intellectual disability and is supported by evidence that behavioural dysregulation is elevated in particular genetic syndromes (Basile et al., 2007).

1.12. Summary

In summary, this review brought together a wide range of literature to argue behavioural dysregulation in individuals with ID is often a neglected factor in the parental stress literature. Composite scores that merge numerous behaviours together might mask important associations, particularly when they combine overt and demonstrably functional behaviours such as self-injury, aggression and property destruction with the child correlates of these behaviours for which operant accounts may not be applicable: impulsivity, repetitive behaviour, ASD phenomenology and hyperactivity. It may be that child dysregulation characteristics impact on the trajectory of self-injury, aggression and property destruction and parents' responses to these behaviours. Further research could test this model by adopting a fine-grained approach to studying challenging behaviour and parental stress in ID research.

1.13. References

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Parental perceptions of challenging behaviour and the effect of behavioural dysregulation

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Abstract

Background. Parental perceptions of challenging behaviour (CB: self-injury, aggression and property destruction) in adults with intellectual disabilities were explored using the Self-Regulatory Model of Illness Behaviour (Leventhal et al., 1984). Associations between parental perceptions of CB and parental wellbeing were explored, and whether child characteristics affected these associations.

Method. Sixty-five parents completed the Illness Perception Questionnaire-Revised, adapted to measure perceptions of self-injury, aggression or property destruction. Parental locus of control, attributions about behaviour, psychological distress, and the extent of CB were measured. Data on child impulsivity, over-activity, repetitive behaviour and autism spectrum characteristics were included.

Results. Inter-correlations between IPQ-R subscales support the Self Regulatory Model. Few parents endorsed operant reinforcement as a cause of CB. Psychological distress was associated with a belief in *child control over parent*. CB and behavioural dysregulation variables were independently related to parental perceptions.

Conclusions. The Self-Regulatory model is promising for further research into parental perceptions of CB. Research should focus on differentiating between CB related to operant reinforcement and markers of behavioural dysregulation.

Keywords: Challenging behaviour, impulsivity, over-activity, hyper-activity, parental perceptions, well-being.

2.1. Background

Approximately 60% of individuals with intellectual disability (ID) continue to live at home with their parents once they reach adulthood (Department of Health, 2001). Parents who support an adult with a genetic syndrome with associated ID may experience additional challenges as a high proportion (approximately 40-70%) will engage in 'challenging behaviours' such as aggression, self-injury and destruction of property (Arron et al., 2011). In recent years there has been increased interest in investigating the impact of CB such as self-injury, aggression and property destruction on parents' psychological well-being (Hastings, 2002).

CB is often loosely defined in these studies with composite scores of global 'behaviour difficulties' readily employed. These composites often include a wide range of internalising and externalising behaviours and behaviours and psychological characteristics associated with self-injury, aggression and property destruction are not always independently assessed (e.g. impulsivity, hyperactivity, repetitive behaviour). Despite these methodological issues, associations have been established between CB such as self-injury, aggression, property destruction and poorer parental well-being in individuals with autism spectrum conditions, genetic syndromes and mixed aetiology groups (Beck et al., 2004; Eisenhower et al., 2005; Hastings, 2002; Hodapp et al., 1998). For a large proportion of these studies the direction of relationships cannot be established due to the designs. However, when evidence from longitudinal studies is examined, CB such as self-injury, aggression and destruction of property and parental stress appear to be related by a bidirectional positive relationship (Hastings et al., 2006; Orsmond et al., 2003).

The majority of studies that have explored self-injury, aggression and property destruction and the relationship with parental outcomes have sampled families with younger children. Minnes et al. (2007), in one of the few studies with adults with ID, found a link between general 'behaviour difficulties' and depression in parents that was mediated by perceived stress. Hill and Rose (2010) note that there often seems to be an underlying assumption that supporting a person with an ID is stable across the life span. However, families with adult children with neurodevelopmental disorders have been supporting their children with self-injury, aggression and property destruction for a number of years and in longitudinal studies behavioural difficulties have been found to persist over time (Murphy et al. 2005; Taylor et al., 2011). Therefore, further studies are warranted to explore the impact of supporting adults with rare genetic syndromes who have engaged in these behaviours for a number of years.

The observation that not all parents of children develop depression or anxiety is testament to the role of mediating and moderating factors (Hastings, 2002). These factors include social support, additional child characteristics and parental cognitions (MacDonald et al., 2010; McClintock, et al., 2003; Plant & Sanders, 2007). Research into the impact of parental cognitions has

increased steadily with a focus on causal attributions, self efficacy, psychological acceptance and locus of control (Dagnan et al., 2004; Hassall et al., 2005; Hastings & Brown, 2002; MacDonald et al., 2010). Findings demonstrate that the relationship is complex. Lloyd and Hastings (2009) conducted a longitudinal study and found that locus of control impacted on parental well-being but that it did not act as a moderator between parental well-being and 'behaviour problems' measured by a composite score. It was suggested that this may be underpinned by fluctuations in locus of control across situations and time. Hastings and Brown (2002) have found that the relationship between CB, such as self-injury, aggression and property destruction, and poorer parent well-being may be mediated by self-efficacy. Lancaster et al. (2013) found preliminary evidence that parental criticism, associated with attributing behaviour internally to the child, may moderate the relationship between CB, such as self-injury, aggression and property destruction, and psychological distress. In combination, this research indicates that there are a number of cognitive variables that could mediate the relationship between parental outcomes and self-injury, aggression and destruction of property. Williams and Rose (2007) note that while specific cognitive variables have been explored in relation to behaviours such as self-injury and aggression, a large proportion of research has been guided by Attribution Theory and has been conducted with support workers in residential settings (Weiner 1986; 1993). Willner and Smith (2008) argued that the evidence for Attribution Theory in ID was inconsistent and that new theoretical models should be considered. One model that has shown some promising results with residential staff and parents of children with Autism is Leventhal's Self Regulatory model (Al Anbar et al., 2010; Leventhal et al., 1997; Leventhal et al., 1984 ; William & Rose, 2007). The model's strength is that it encompasses a wide range of cognitive variables and the inter-correlations between these variables tend to be consistent across populations (Hagger & Orbell, 2003).

The Self Regulatory Model is used frequently to investigate representations of illness (Hagger & Orbell, 2003). Leventhal proposed that individuals form illness representation schemas about the identity, timeline, consequences, control over, and cause of the illness. These schemas are influenced by three levels of information: 1) cultural and societal norms about the illness, 2) information from professionals and significant others 3) information from personal experience of having the illness. A further reason that the Self Regulatory Model may be useful for studying perceptions of self-injury, aggression and property destruction is because some of the constructs overlap with constructs that have been explored previously in parents of children with ID. For example, personal control as captured by the Self Regulatory Model is a similar construct to locus of control investigated by Lloyd and Hastings (2009).

The constructs in the Self Regulatory Model can be measured by the Illness Perception Questionnaire-Revised (Moss-Morris et al. 2002) which evaluates the five representations originally

described by Leventhal and three subscales that assess cyclical timeline, illness coherence and emotional representations. The addition of these subscales has been supported by numerous studies that have utilised this measure (Moss-Morris et al., 2002). Although this measure was originally developed for physical illness, it has been adapted successfully for carers of individuals with schizophrenia, support workers of adults with ID, and parents and carers of individuals with Autism (Al Anbar et al, 2010; Barrowclough et al., 2001; Mills & Rose, 2011; Williams & Rose, 2007). To date, the measure has not been adapted specifically for use with parents of adults with ID.

The structure of the Self-Regulatory Model has been supported across a number of studies across a range of populations and the constructs have been associated with mood (Fortune et al., 2000; Murphy et al., 1999), adaptive coping and response to the difficulty, and adherence to clinical recommendations (Cooper et al., 1999; Heijmans, 1998; Heijmans and de Ridder, 1998; Moss-Morris et al., 1996; Scharloo et al, 1998, Scharloo et al., 2000; Weinman et al., 2000). Hagger and Orbell (2003) demonstrated that the model was consistent across populations with greater belief in control over illness related to weaker representations of illness identity, fewer perceived negative consequences and less strongly held beliefs about the chronicity of illness. Poorer psychological well-being was related to perceiving greater negative consequences, a stronger illness identity and a belief that the illness was chronic; whereas, perceived control was found to be a protective factor related to active coping and cognitive reappraisal. Moss-Morris et al. (2002) showed that lower illness coherence and a stronger belief in a cyclical time-line were related to beliefs in more severe consequences and stronger emotional representations (Moss-Morris et al., 2002). The merit of using this model in ID populations is that it might be useful for understanding how parents cope, why their levels of distress vary, why they choose to seek support, and how their understanding of the causes of self-injury, aggression and property destruction may influence reactions to these behaviours.

The adoption of the Self Regulatory Model to explore parental perceptions of CB (self-injury, aggression and destruction of property) exhibited by their adult children, and parental well-being in relation to these perceptions are two aims of this study. The third aim is to consider the impact of child characteristics on parental perceptions and psychological well-being. Child characteristics were studied at a fine-grained level in this study. As previously noted, CB is often categorised as a composite behavioural score in which self-injury, aggression and destruction are grouped with other behaviours. This could mask underlying associations between parental perceptions of and specific types of behaviour and the role of particular child characteristics known to be associated with some forms of challenging behaviour as mediating factors (see Waite, this thesis).

There is growing evidence that child characteristics: impulsivity, over-activity, and repetitive behaviour are correlates of behaviours such as self-injury, aggression and destruction of property (Davies, 2010). A recent principal components analysis suggested that these characteristics load onto

one factor that could be indicative of underlying behavioural dysregulation (Oliver et al., 2009). It may be that impulsivity or over-activity underpin or mediate associations between CB and parental perceptions/outcomes. Therefore, in this study behaviours that may indicate underlying behavioural dysregulation were measured independently from self-injury, aggression and property destruction. The role of behavioural dysregulation as a mediator and moderator was considered alongside additional child characteristics that have been associated with CB: mobility, self-help, verbal ability and Autism Spectrum Disorder (ASD) (McClintock et al., 2003).

It was predicted that the Self Regulatory Model would be a good fit for parental perceptions of CB. Therefore, strongly held beliefs about chronic time-line and negative personal consequences were expected to be associated with stronger negative emotional reactions, a lower degree of perceived parental control over the behaviour and weaker beliefs in the availability of effective interventions for behaviour. It was predicted that negative perceptions, lower parental well-being and challenging behaviour would be related, but that behavioural dysregulation may be contributing to these associations.

2.2. Method

2.2.1. Design

This was a cross-sectional correlational study using questionnaire methodology as part of a longitudinal study exploring behavioural and cognitive phenotypes of genetic syndromes. The study began in 2003 and follow-up studies were conducted in 2006 and 2011. All participants had previously completed questionnaires in Autumn 2011 including measures of self-injury, aggression and property destruction, behavioural dysregulation, mood, feeding, ability and Autism Spectrum Disorder. Parents had indicated that they wished to be contacted for future studies and ethical approval was granted by NRES Committee West Midlands - Coventry and Warwickshire (Appendix A).

2.2.2. Recruitment

Invitation letters were sent to 132 parents of children with Angelman (AS: N = 21), Cornelia de Lange (CdLS: N = 34), Cri du Chat (CdC: N = 4), Lowe (LS: N = 14), Prader-Willi (PWS: N = 12), Smith Magenis (SMS: N = 11), and Fragile X syndromes (FXS: N = 35). Parents were selected from a sample of 313 individuals as part of the 2011 follow-up study conducted 8 months prior to this study. Inclusion criteria were that the person they cared for was aged 16 years or over, had a diagnosis of a syndrome by a professional, lived at home with their parent/care-giver, and had been identified as engaging in aggression, self-injury or destruction of property at the most recent follow-up and at least one previous time point.

Questionnaires were returned by 77 parents (CdC: N = 3, 75%; AS: N = 14, 66.67%; CdLS: N = 18, 52.94%; FXS: N = 19, 54.92%; PWS: N = 6, 50%; LS: N = 9, 64.29%; SMS: N = 6, 54.55%). Three participants were excluded because they indicated that in the last 12 months their child had moved into residential accommodation. An additional nine participants were excluded because more than one child with a diagnosis of a genetic syndrome lived in the household.

2.2.3. Participants

The sample consisted of 65 parents/carers (mean age: 52.05 years; range = 34.00 - 76.00); 94.9% were mothers. The mean age of the adults with ID was 26.34 years (range: 16.84-51.93); 53.8% were male. 36.9% of adults with ID had been diagnosed by a paediatrician, 50.8% by a clinical geneticist, 10.8% by a GP, and 1.5% by another professional. Demographic variables from the Wessex Questionnaire (Kushlick, Blunden & Cox, 1973) indicated that 64.6% were mobile, 64.1% had normal vision, 81% had normal hearing, 68.8% were partially verbal or verbal, 55.5% were partially able/able³.

2.2.4. Procedure

Invitation letters offered participation in an online questionnaire study exploring parents' perceptions of CB (Appendix B). A weblink, password and ID number were included so that parents could access the consent forms (Appendix C) and questionnaire via Limesurvey (Appendix D). Participants were notified that they could request a paper copy and 22 participants (29.73%) opted to return a paper copy. Four to six weeks after sending invitation letters, parents were contacted via telephone to enquire whether they had questions or difficulties accessing the survey.

Measures were administered in a fixed order. Participants completed the Challenging Behaviour Questionnaire first (CBQ; Hyman et al., 2002) because following this they were asked to indicate whether self-injury, aggression or property destruction had the greatest impact on their day to day life so that this behaviour could be used as a focus for the following items. Participants then completed the measures in the order described below. To reduce burden, questionnaire results on impulsivity, over-activity, ASD, ability level and repetitive behaviour were taken from informants' previous responses collected in Autumn 2011. These scores are stable across short time-frames (see methods: data analysis).

³ Partially able/able is defined as a score of 6 or above on the Wessex self-help score (total max score = 9). This score includes feeding, dressing and washing, which are individually assessed on a three point scale: not at all, with help or without help (max score = 3) and then summed.

2.2.5. Measures.

2.2.5.1. The CB Questionnaire (CBQ; Hyman et al., 2002). This brief, informant based questionnaire measures self-injury, physical aggression, destruction of property and repetitive behaviour. Informants indicate the presence of a behaviour and then rate frequency, severity and duration on five-point Likert scales. In the original measure the Likert scales are only administered for the self-injury item; however, in the present study the measure was adapted by duplicating these scales for physical aggression and destruction of property. The repetitive behaviour item was excluded. Previous examination of the psychometric properties of the questionnaire has demonstrated good inter-rater reliability (Kappa value = .92). Higher scores indicate more severe behaviour.

2.2.5.2. Modified Illness Perception Questionnaire-Revised (IPQ-R; Moss-Morris et al., 2002). The Illness Perception Questionnaire captures the five components of the Self-Regulation Model of Illness Behaviour (Leventhal, 1987). The subscales are: identity, timeline (acute/chronic), timeline (cyclical), consequences, personal control, treatment control, illness coherence, emotional representations and cause. For all subscales, apart from the *identity* and *cause* subscale, informants use a 5-point Likert scale ranging from strongly disagree to strongly agree. The measure has shown good predictive validity, for example, adjustment to illness in multiple sclerosis (Moss-Morris et al., 2002). The subscales of the Illness Perception Questionnaire have good internal reliability (range .75-.89) and acceptable test-retest reliability (range .46 to .88; 75% > .70).

The subscales with Likert scales were adapted to measure parents' views of CB. Items were modified by changing the word 'illness' to a type of CB (e.g. aggression) in agreement with guidance for using the IPQ-R (www.uib.no/ipq/ accessed 06.03.12). The type of CB substituted for the word 'illness' was determined by asking parents to indicate the behaviour that had the most impact on their day to day lives. Slight modifications were made to the wording to make items applicable and transparent to parents (e.g. the word 'my' was changed to 'his/her', the first three questions were prefaced with 'looking ahead to the future'). Overall, there were 38 items. The *identity* subscale was omitted because this scale typically includes a list of symptoms that form part of an illness. This subscale did not fit with the aims of the study and would require extensive development and validation.

A new *cause* scale was developed. Hagger and Orbell (2003) have identified a number of causal factors associated with illness representations and that these fall into four domains: biological, emotional, psychological and environmental. The *cause* subscale was developed for this study measured a range of causal factors similar to those found in the health psychology literature, and was informed by research that carers often endorse internal emotional or internal organic causes for challenging behaviour while lacking specific behavioural knowledge about how behaviour is reinforced (Oliver et al., 1996). Parents rated agreement that aggression/self-injury/destruction was

caused by: syndrome, atypical brain development, mental health, mood/emotions, situation/environment, shaping/reinforcement and pain (Beail, 2003; Breau et al., 2003; Esbensen, 2011; Iwata et al., 1994; McGill & Langthorne, 2011; Oliver et al., 2009). There were two items per factor that were rated on a five-point Likert scale (strongly disagree to strongly agree).

Higher scores on the IPQ subscales indicate more strongly held beliefs, so, on the *consequences* subscale it indicates a stronger belief in more negative consequences for the parent, on the *timeline chronic* subscale a stronger belief that behaviour will persist, on the *timeline cyclical* subscale a stronger belief that the behaviour changes day to day and is unpredictable, on the *emotional representations* subscale a higher degree of negative emotional responses to the behaviour, on the *person control* subscale a greater belief in the ability to influence the likelihood of behaviour occurring, on the *treatment control* subscale it indicates a belief in effective interventions, on the *illness coherence* subscale a greater belief that the behaviour is understandable, and on *cause* subscale a stronger belief that a particular cause explains the behaviour.

2.2.5.3. Controllability Beliefs Scale (Dagnan et al., 2004). The controllability beliefs scale was originally designed to measure staff's attributions concerning service users' CB. The measure was designed for use in Dementia services. However, items are applicable to individuals with ID. Informants use a 5-point scale to indicate agreement with statements concerning the reasons why CB occurs. The scale has good internal reliability ($\alpha = .89$). It has been employed in studies with staff teams who support service users with ID (Kalsy et al., 2007; Mills & Rose, 2011). A higher score signifies that the parent believes their child has greater internal control over their challenging behaviour.

2.2.5.4. Parental Locus of Control Scale (short-form) (Hassell et al., 2005). The Parental Locus of Control Scale (short form) is a 24 item questionnaire. Informants respond to statements on five-point Likert scales (disagree to strongly agree). Reported Alpha coefficients for internal consistency range from .65 to .77 for the subscales and .92 for the total scale (Campis et al., 1986). Test-retest reliability for the entire scale is 0.83. In this study a short form of the measure was used. Hassell et al., (2005) selected the six items with the highest factor loadings on each subscale yielding four subscales: parental efficacy, parental responsibility, parents' control over child, child's control over parent; and a total score. A higher score on this measure indicates a greater external locus of control. It has been used in studies with individuals with ID (Hassall et al., 2005; Hill & Rose, 2009).

2.2.5.5. Hospital Anxiety and Depression Scale (Zigmond & Snaifh, 1983). This is a widely used measure of anxiety and depression (Bjelland et al., 2002). It has 14 items with four possible responses. Informants rate their experience over the last few days. It yields an anxiety score, a depression score and a composite anxiety-depression score. The total score has been argued to be a

measure of general psychological distress (Johnston et al., 2000). Individuals scoring above eight on the anxiety and depression scales are categorised as falling above a clinical cut-off, with scores classified as mild (8-10), moderate (11-14) and severe (14+). The measure has excellent test retest reliability ($r = .84$ & $.85$) and internal consistency (Cronbach's Alpha: $.85$ for anxiety & $.79$ for depression). Studies support the use of the HADS for non-clinical populations (Bjelland et al, 2002).

2.2.6. Additional Child Characteristic Measures

A parsimonious approach was used throughout whereby specific subscales and total scale scores were selected from the 2011 follow-up study to explore the role of child characteristics, and in particular, characteristics associated with behavioural dysregulation. Mobility, self-help ability, verbal ability and ASD characteristics were included based on previous research findings of an association with CB (McClintock et al., 2003). Four characteristics previously hypothesised to be indicative of behavioural dysregulation were included: impulsivity, impulsive speech, over-activity and repetitive behaviour (Arron et al., 2011). These were taken from the following measures:

2.2.6.1. Wessex Scale (Kushlick et al., 1973; Appendix E). The Wessex Questionnaire is a short informant-based measure that assesses physical and social ability. Domains covered include mobility, vision, literacy, hearing, continence, speech and self-help. The questionnaire has been used in a wide range of published studies with people with ID (Dagnan et al., 1998; Moss et al., 2000; Oliver et al., 2008). The scale has modest reliability (Kappa value = $.62$ for overall scale and mean of $.54$ for item level). Palmer and Jenkins (1982) argue it is an appropriate measure for large scale questionnaire studies. Higher scores on this measure indicates greater ability.

2.2.6.2. The Activity Questionnaire (TAQ, Burbidge & Oliver, 2008, Appendix F). The Activity Questionnaire consists of 18 items that form three subscales: impulsivity, over-activity and impulsive speech. The questionnaire was developed specifically for people with ID. Factor analyses have supported the structure of the measure (Burbidge et al., 2010). The subscales have good inter-rater and test-retest scores (all correlations $\geq .70$). The measure is suitable for mobile and immobile participants as a prorated scale is used for immobile participants. A higher score indicated greater levels of behaviour.

2.2.6.3. Social Communication Questionnaire (SCQ; Rutter, et al., 2003; Appendix G). The SCQ is a 40 item ASD screening tool. The subscales measure communication deficits, social interaction deficits and repetitive behaviour. A higher score on these subscales indicate a higher level of impairment. The SCQ has been validated with clinical populations (sensitivity = $.85$; specificity = $.75$) and against the Autism Diagnostic Interview-Revised (Berument et al., 1999; Lord et al., 1994). Internal consistency is good ($\alpha = .90$ for full scale, Berument et al., 1999). A proportional

communication subscale (score on communication subscale/8 x 13) was used in the current study to avoid non-verbal participants having unrepresentative low scores due to a proportion of items on this scale not being applicable to these participants.

2.3. Data Analysis

2.3.1. Composite Scores

2.3.1.1. CBQ Composite Score. A total CBQ score was calculated combining self-injury, aggression and property destruction. Prior to calculating this score, participants who had responded that self-injury, aggression, and/or destruction of property had not occurred in the last month were assigned a score of zero for the corresponding severity, duration and frequency items so that a total score could be calculated for each person. Combining self-injury, aggression and property destruction into a total score was justified as the aim was explore parental representations of challenging behaviours frequently associated with operant reinforcement, while ensuring that behavioural dysregulation characteristics were not combined into the challenging behaviour composite score. Parents focused on a specific behaviour, self-injury, aggression or property destruction, while completing the IPQ but Mann-Whitney U tests comparing participants who focused on self-injury and those who focused on aggression⁴ (see Appendix H) indicated that there were no group differences on any parental perception/well-being measure, which further supported using a total score.

Moderate positive correlations⁵ between the severity, duration and frequency scores for self-injury, aggression, and property destruction supported combining scores (see Appendix I). Some correlations that were weaker and did not reach significance (for example, correlations with the *destruction of property duration* score); however these were still combined as the purpose of the subscale scores was to capture the overall experience of these challenging behaviours, and frequency, severity and duration might not always be equal for individuals.

The total CB score combining aggression, self-injury and property destruction was derived following the method reported by Lopez et al. (2005). The scores for frequency, duration and severity for each behaviour were standardised by conversion into z-scores and then these were averaged to yield the CBQ total score. These subscale scores were used in an analyses of parental perceptions of challenging behaviour.

⁴ Mann-Whitney U tests were not conducted for destruction of property due to only a small number of participants who indicating that this was the behaviour that had most impact on their day to day lives (N=7).

⁵ The analysis that was conducted to check aggression, self-injury and property destruction scores correlated prior to combining these into the composite score was conducted without the participants who were assigned a score of zero across all three behaviours (no behaviour in last month). This was deemed more conservative as the scores from these participants would have been perfectly correlated and therefore any overall associations might have been inflated by their inclusion. These participants were included in all other analyses in this study.

2.3.1.2. IPQ Subscales and Cause Scale Composite Scores. The IPQ was adapted for this study so the internal consistency of each subscale was examined. Cronbach's Alpha was good to excellent for six out of seven subscales (.81 - .93). Alpha for the *timeline-cyclic* subscale was adequate (.68) (Appendix J). The *cause* subscale was developed for this study and initial correlations revealed that the two-item scales were inadequately correlated to justify merging the scales into a composite score for each causal factor. However, inspection of scatter plots for each pair revealed a small subset of participants whose scores were at opposite extremes on the two items (Likert scale difference +/- 3). The number of participants for each pair whose scores fell in this pattern were: mental health: N = 3; atypical brain development: N = 3; reinforcement/operant: N = 3; situation/environment: N = 3; mood/emotions: N = 2; pain: N = 0; syndrome: N = 2. These participants were removed as they were outliers. The Spearman Brown's coefficients were recalculated for each scale and were good (range .51-.84; five out of seven items > .70) (Appendix J).

Composite scores were calculated for pairs of items. If participants agreed with each item they were assigned a score of five, if they agreed with one and partially agreed with another they were assigned a score of four, if they partially agreed with both or agree and disagreed they were assigned a score of three, if the partially agreed and disagreed they were assigned a score of two, if they disagreed with both items they were assigned a score of one. The outliers were excluded from this composite score. The five point scale was used in analyses.

2.3.2. Data Analysis Strategy. All analyses were conducted at group level due to the small N for each syndrome group and to reduce the number of correlations and possibility of a type-I error. The order of the analyses followed the aims of this study. The *cause* subscale of the IPQ was analysed in more detail than the other IPQ subscales to explore the causal factors parents endorsed most frequently on this scale and how this fitted with previous research studies (e.g. Oliver et al., 1996).

Inspection of normality plots and tests revealed that a large proportion of the subscales were not normally distributed. The data could not be transformed so non-parametric correlations, Friedman, Wilcoxon signed ranks and Mann-Whitney U tests were employed. Simultaneous linear regressions were conducted to explore the association between parental perceptions and parental well-being, and child characteristics (i.e. behavioural dysregulation variables) and parental perceptions/outcomes. Prior to the regression analyses, inspection of the residual plots indicated that regression should be robust.

Due to the exploratory nature of the analysis a p-value of .005 was adopted for multiple t-tests. Correlations are reported at three levels .005, .01, and .05. Only a small subset of regression analyses were conducted to examine relationships in more detail so a less conservative *p*-value is adopted for these analyses to reduce the likelihood of making a type II error.

2.3.3. Stability of TAQ and SCQ variables. As subscales were used in this analysis from the previous follow-up in 2011, data collected at this time might not have been an accurate representation of levels of impulsivity, over-activity, impulsive speech, repetitive behaviour and ASD phenomenology at the time of the current study. Prior to the analyses including these variables, an analysis was conducted to compare scores obtained in 2011 to scores in 2006 to check that behaviours were relatively stable. Spearman's correlations revealed that there was adequate stability for over-activity, impulsive speech, repetitive behaviour and social interaction ($r_s > .6$). Impulsivity approached this cut-off ($r = .56$). Wilcoxon tests revealed that scores did not differ across time-points except for the SCQ communication and social interaction subscales where there was a slight decrease in scores ($z = -.26, p = .008$; $z = -.39, p < .001$). These results suggest that the behaviour regulation variables of interest in this study are likely to be fairly stable and that 2011 estimates are appropriate (see Appendix K for full statistics).

2.4. Results

2.4.1. Descriptive statistics

The descriptive statistics for all variables are displayed in Table 2.1. The inter-quartile ranges indicate that there is a reasonable spread of data for the majority of the items. Smaller inter-quartile ranges for variables such as the *situation* subscale from the *cause* scale indicate highly consistent responses from parents.

Table 2.1.
Descriptive statistics for all variables included in the analysis.

Measure	Variable	N	Max Score	Median	Inter-Quartile Range	Percentage > clinical cut-off
IPQ	Timeline Chronic	65	30	21	6	
	Timeline Cyclical	65	25	14	3	
	Consequence	65	30	18	6	
	Personal Control	65	30	18	7	
	Treatment Control	65	25	15	5.5	
	Illness Coherence	65	25	16	8	
	Emotional Representations	65	30	19	7.5	
IPQ Cause Scale	Syndrome	62	5	4	2	
	Brain Difference	61	5	4	2	
	Mental Health	61	5	2	1.5	
	Mood	62	5	4	1	
	Situation	61	5	5	0	
	Reinforcement	61	5	1	2	
	Pain	64	5	3	3	
PLOC	Parental Efficacy	65	35	14	4	
	Child Control	65	35	18	4	
	Parental Control	64	35	18.5	5	
	Parental Responsibility	65	35	23	2	
	Total	64	140	70.5	14.75	
CBS	Total	65	64	18	14	
HADS	Anxiety	65	21	8	6.7	Mild: 21.5%; Moderate: 32.3%; Severe: 3.1%
	Depression	65	21	5	3.2	Mild: 15.4%; Moderate: 13.8%; Severe: 1.5%
	Psychological Distress	65	42	14	12	
Wessex Scale	Mobility	65	3	3	1	64.6% mobile
	Speech	65	3	2	2	68.8% verbal/partially verbal
	Self Help	64	9	6	3.5	55% able/partially able
SCQ	Communication	65	13	7	4.23	
	Sociability	65	15	7	5	
	Repetitive Behaviour	65	8	4	2.5	
TAQ	Impulsivity	65	24	18	9.75	
	Over-activity	65	36	14	16.5	
	Impulsive Speech	39	12	5	8	
CBQ ^a	Self injury severity	48	5	2	3	
	Self injury duration	48	5	2	3	
	Self injury frequency	48	5	3	2	
	Aggression severity	40	5	3	1	
	Aggression duration	40	5	3	3	
	Aggression frequency	40	5	2	2	
	Destruction severity	36	5	3	1	
	Destruction duration	36	5	2	2	
	Destruction frequency	36	5	2	1	

^a Median scores presented at subscale level prior to CB total score computation with z-score transformation.

2.4.2. Parental Perceptions of CB (Self-Injury, aggression, property destruction)

The first aim of this study was analysis of inter-correlations of the IPQ subscales to explore the utility of the Self Regulatory Model and associations between measures of parental perceptions/outcomes. Associations between the IPQ, the PLOC, CBS and HADS are displayed in Table 2.2. As predicted, perceiving that CB (self-injury/aggression/destruction) would persist for a long time was associated with a stronger belief in negative consequences of the behaviour on parents, perceptions of lower personal control over CB and a less strong belief in the availability of effective interventions. A stronger belief in negative consequences was associated with a stronger belief in a cyclical time-line for CB and stronger negative emotional reactions to the behaviour. Finally, a stronger belief in personal control was strongly related to a belief in availability of interventions.

The IPQ subscales related consistently to the PLOC subscales. Higher PLOC external locus of control was significantly positively associated with a stronger belief in negative consequences on the IPQ, a lower sense of personal control, less strong beliefs in the availability of interventions and negative emotional reactions. Child control over parent was associated with a belief in negative consequences, negative emotion representations and lower perceived personal control on the IPQ. Lower self efficacy on the PLOC was related to lower personal control on the IPQ, less strong beliefs in available interventions and a poorer sense of personal understanding of CB.

The HADS anxiety and depression scores were associated with more external locus of control, in particular: child control over parent, and less parent control over child; and negative emotional representations. Heightened anxiety was also related to a stronger belief in a chronic timeline for CB. The majority of associations were moderate.

A belief that CB was caused by atypical brain development on the *cause* scale was moderately associated with a weaker sense of illness coherence, and weakly associated with increased levels of anxiety, external self efficacy, external parental control, and a weaker belief that their child has control over behaviour (CBS Score). Stronger beliefs that the child's syndrome was a causal factor replicated this pattern with parental control and child control over behaviour.

A belief that environment/situational factors cause CB was moderately associated with greater personal control and self-efficacy; and weakly associated with greater parental responsibility for child's general behaviour. Reinforcement/shaping as a causal factor was positively associated with parental responsibility as well as a greater belief in available interventions. Finally, a strong association was found between higher scores on the pain items and perceiving negative parental consequences and a belief in situational factors impacting on behaviour.

Table 2.2. Correlations between parental perception/outcome measures (IPQ-R, Cause Scale, PLOC-Short Form, CBS & HADS)

	IPQ-R – Scaled Subscales							Cause Scale							PLOC					CBS	HADS		
	2.	3.	4.	5.	6.	7.	8.	9.	10.	11.	12.	13.	14.	15.	16.	17.	18.	19.	20.	21.	22.	23.	
1.IPQ-R Timeline	.16	.37***	-.37**	-.37**	-.01	.18	.35*	.14	-.08	-.08	-.15	-.20	-.06	.08	.29	.21	.18	.29*	-.23	.30*	.16	.20	
2.IPQ-R Timeline Cyclic		.38***	.04	-.13	-.29	.12	.15	.05	.14	-.04	.15	.10	.20	-.00	.04	.07	.13	.08	-.08	-.05	-.10	-.08	
3.IPQ-R Consequences			-.22	-.09	-.22	.40***	-.01	-.06	.06	.00	-.10	.06	.20	.02	.45***	.36***	.19	.37***	.11	.06	.11	.06	
4.IPQ-R Personal Control				.56***	.16	-.21	-.19	.14	.03	.20	.30*	.20	.28*	-.31*	-.25*	-.25	-.24	-.37***	.21	-.21	-.20	-.16	
5. IPQ-R Treatment Control					.28	-.21	-.18	-.16	.23	.04	.14	.33**	.18	-.25*	-.04	-.37***	-.17	-.31*	.16	-.14	-.17	-.09	
6. IPQ-R Illness Coherence						-.06	-.16	-.30*	-.11	-.06	.16	.05	-.10	-.38***	.02	-.22	-.16	-.30*	.13	.19	.19	.23	
7.IPQ-R Emotion Representation							.03	-.05	.01	-.10	.10	.07	.06	.21	.35**	.65***	.05	.49***	-.01	.44***	.41***	.46**	
8. Cause: Syndrome								.38***	-.03	-.29*	-.12	-.08	.05	.21	.05	.28*	.05	.20	-.30*	.12	-.09	.01	
9. Cause: Brain Development									.35**	.08	-.29*	.01	.06	.28*	-.05	.29*	.10	.18	-.27*	.27*	.03	.10	
10. Cause: Mental Health										.31*	.04	.11	.14	.03	-.08	-.02	.07	.03	.05	.01	-.09	-.06	
11. Cause: Mood											.28*	.13	.07	-.25	-.13	-.01	-.02	-.09	.17	-.06	-.23	-.18	
12. Cause: Environment												.07	-.09	-.31*	-.12	-.12	-.28*	-.32*	.03	.07	-.04	.03	
13. Cause: Reinforcement													.18	-.06	.06	.12	-.30*	-.06	.14	-.1	-.10	-.09	
14. Cause: Pain														-.15	.07	.04	-.06	.00	.11	.06	-.05	.03	
15. PLOC Parental Efficacy															.17	.41***	.24	.65***	-.12	-.12	.16	.13	
16. PLOC Child Control																.51***	.11	.67***	.08	.40***	.40***	.43**	
17. PLOC Parental Control																	.21	.80***	.01	.50***	.41***	.34**	
18. PLOC Parent Responsibility																		.51***	-.18	.09	.01	.03	
19. PLOC Total																			-.07	.39***	.32*	.34**	
20. CBS Total																				-.13	-.00	-.02	
21. HADS Anxiety																					.70***	.91***	
22. HADS Depression																						.92***	
23. HADS Personal Distress																							

*** $p < .005$ ** $p < .01$ * $p < .05$

2.4.3. Causal Factors Endorsed by Parents on Cause Scale

Further analyses were conducted to explore the causal factors that parents endorse for CB (self-injury/destruction/aggression). A Friedman test revealed that there was a significant difference between parents' responses on subscales of the *causal* scale ($\chi^2(6) = 119.17, p < .001$). Post hoc tests are depicted in figure 2.1.

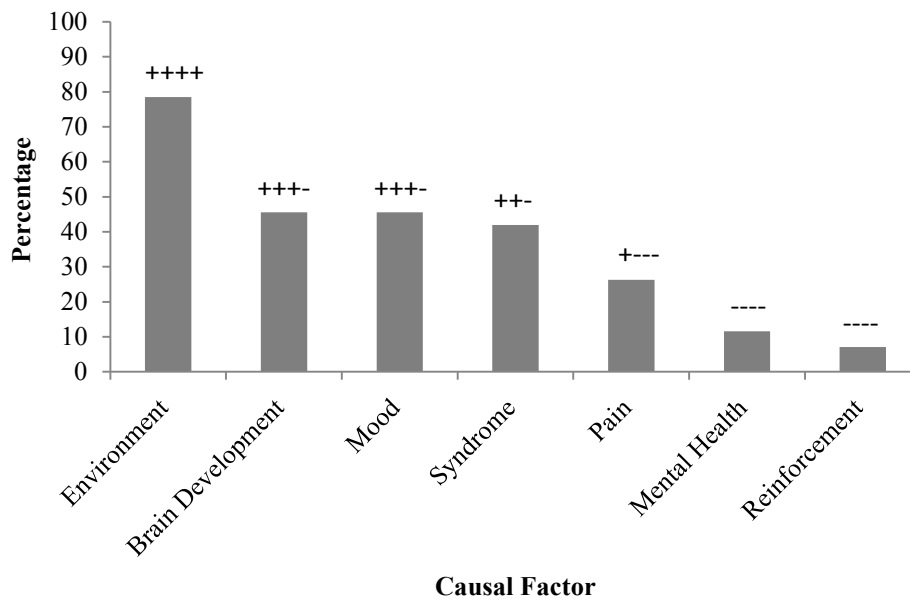


Figure 2.1. Percentage of people who agreed with both items from each causal scale.

+ greater than one other causal factor

- less than one other causal factor

Note. Percentage of participants who agreed that causal factor impacted on behaviour (agreed to both items measuring the causal factor). 'Percentage agreed' is displayed to aid interpretation as the mean score could not be presented due to the non-parametric nature of the data and the median score did not capture subtle differences between syndrome and mood/brain development items. However, the statistical analyses were conducted using the total composite score for the *cause* scale..

Note. CB total score (self-injury, aggression & destruction) was appropriate for these analyses as no differences were found between those who indicated prior to completing the IPQ that self-injury was the behaviour of most concern and those where aggression was of most concern (see data analysis section).

Parents more frequently agreed that situational factors, their child's mood and atypical brain development underpinned CB (self-injury, aggression, property destruction). They were least likely to agree that the behaviour was reinforced/shaped, related to poor mental health or pain.

2.4.4. Parental Clinical Outcomes: Anxiety and Depression

The second aim was to explore global parental well-being. Thirty-seven (56.9%) parents fell above the cut-off for anxiety and nineteen (30.8%) fell above the cut-off for depression. As previously observed in Table 2.2., poorer well-being was strongly associated ($\geq .40$) with perceived child control over parent, lower parental control over child and stronger emotional representations (negative emotional responses). As the relationship between these parental perception variables with depression and anxiety appeared equivocal, regression analyses were conducted for the total score (psychological distress index) to identify factors most predictive of psychological distress. A summary of all results is displayed in Table 2.3. The results of the regression indicated that the model accounted for 29% of the variance ($R^2 = .29$, $F(3,63) = 8.03$, $p < .001$). Child control and timeline cyclical were significant predictors of psychological distress when the other variables were entered.

Table 2.3.
Simultaneous regression analysis of variables associated with psychological distress

DV	R ²	IVs	B	SE B	β	t	p
Psychological Distress	.29						
		(constant)	-6.48			-1.48	.144
		Emotional Representations	.29	.23	.19	1.27	.211
		Child control over parent	.57	.24	.30	2.40	.020
		Parent control over child	.31	.29	.17	1.08	.283

2.4.5. The Impact of Child Characteristics on Parental Perceptions and Outcomes.

The final aim of this study was to explore the impact of degree of CB (self-injury/aggression/destruction) on parental perceptions and explore the association with behavioural dysregulation. Firstly, the associations between challenging behaviour and child characteristics were explored. Associations between subscales of the TAQ, SCQ, CBQ, child age, verbal ability, self help score and parental perceptions of CB are displayed in Table 2.4. There were moderate positive correlations between CB, verbal ability, impulsivity, over-activity and repetitive behaviour.

Principal components analysis was conducted to see whether impulsivity, over-activity and repetitive behaviour (from the RBQ and SCQ) loaded onto one factor in line with our hypothesis of underlying behavioural dysregulation. Kaiser-Meyer-Olkin test and Barlett's test indicated that it was appropriate to perform this test even though the sample was small ($.62$ & $p < .001$ respectively). 59.08% of the variance was explained by a one-factor solution that was labelled 'behavioural dysregulation' (eigenvalue = 2.36). All factor loadings $> .7$. This behavioural dysregulation factor was then used in subsequent analyses alongside the individual variables. Verbal ability did not fit this model.

Table 2.4.

Correlations between CB total score (self-injury, aggression & property destruction) and child characteristics including behavioural dysregulation variables.

	Total CB Score
Age	.01
Mobility	-.08
Verbal Ability ^a	-.35***
Self Help Score	-.19
Impulsivity	.36***
Over-activity	.43***
Impulsive-Speech	-.04
SCQ Communication Score	-.07
SCQ Sociability Score	.19
SCQ Repetitive Behaviour Score	.33**
Behavioural Regulation Score	.46***

*** Significant at $p < .005$ ** Significant at $p < .01$ * Significant at $p < .05$

^a Two participants scores were re-coded from 4 (has speech but chooses not talk) to 3 (has speech).

Note. Behavioural regulation was added to this table after this factor was calculated from the principal components analysis.

Secondly, associations between challenging behaviour and parental variables were explored. Correlations between CB (self-injury/aggression/destruction), child variables and parental perceptions/outcomes are shown in Table 2.5. Only child variables that were correlated with CB were carried over to this next stage in the analyses, which was to explore the role of these child variables on the associations between CB and parental representations/outcomes.

Higher levels of impulsivity were moderately associated with higher degrees of external parental responsibility, a stronger belief in syndrome as a causal factor, and a stronger belief in a cyclical time-line. Higher scores on the challenging behaviour composite were strongly associated with more beliefs about the severity of the consequences of behaviour. CB was moderately correlated with beliefs about chronicity of the timeline, and weakly positively associated with greater child control over parent, belief in cyclical timeline, and external parental responsibility.

Table 2.5.
 Parental Variables and Child Characteristics

	CB Score	Verbal Ability	Impulsivity	Over- activity	SCQ Repetitive Behaviour	SCQ Sociability Score	Behavioural Dysregulation
IPQ Timeline	.38***	-.13	.17	-.13	.09	.15	.15
IPQ Timeline Cyclic	.28*	-.13	.38***	.23	.07	.22	.28*
IPQ	.54***	-.01	.16	.02	.11	-.04	.14
Consequences							
IPQ Personal Control	-.12	.01	-.02	.09	-.07	-.03	-.04
IPQ Treatment Control	.01	-.21	-.15	.19	.03	.04	.00
IPQ Illness Coherence	-.15	-.10	-.14	-.06	.02	-.15	.02
IPQ Emotional Representation	.03	.20	.07	-.25 ^a	-.20	-.06	-.14
IPQ Cause: Syndrome	-.25 ^b	-.05	.31*	.15	.09	.11	.18
IPQ Cause: Brain Difference	.11	-.20	.08	.12	.16	.08	-.10
IPQ Cause: Poor MH	-.05	.04	.12	.04	.00	-.01	.04
IPQ Cause: Mood	-.22	.24	-.05	-.10	-.02	-.20	-.07
IPQ Cause: Environment	-.24	-.14	-.22	.16	-.25 ^c	-.18	-.25
IPQ Cause: Reinforcement	-.10	.13	-.09	-.09	.01	-.17	-.09
IPQ Cause: Pain	.07	-.19	-.13	-.11	.10	.28*	.13
PLOC Parental Efficacy	.12	-.10	.01	-.01	-.22	.04	-.11
PLOC Child Control	.29*	-.08	.05	.01	.11	.14	.16
PLOC Parental Control	.19	.24*	.12	-.15	-.10	-.13	.04
PLOC Parental Responsibility	.25*	-.03	.41***	.10	-.05	-.17	.11
PLOC Total	.32*	.00	.18	-.05	-.06	.01	.04
CBS Total	.00	.22	.15	-.22	.12	-.28*	-.02
HADS Anxiety	.05	.22	-.04	-.08	.04	.00	.04
HADS Depression	.02	.15	.07	-.21	-.25*	.04	-.14
HADS Total	-.04	.17	-.03	-.17	-.15	-.03	.06

 *** Significant at $p < .005$ ** Significant at $p < .01$ * Significant at $p < .05$

^a Inspection of the scatter-plot revealed a subset of four participants (Angelman Syndrome: N = 3; Lowe: N = 1) who were potentially outliers. Parents reported very low scores on the emotional representations subscale of the IPQ that asks about emotional reactions to challenging behaviour and examination of scores on the CBQ suggested only low levels of challenging behaviour, primary destruction of property. The AS participants over-activity scores were very high and given that a phenotypic behaviour of AS (Berry et al., 2005) so this correlation was likely to have been driven by these participants. If these participants were removed the correlation did not reach significance.

^b Approached significance at $p = .05$ ^c Approached significance at $p = .06$

Note. Variables were only included if they correlated with CB in the previous analyses.

Simultaneous regression analyses were conducted for parental responsibility and cyclical time-line from the IPQ given both CB (self-injury/aggression/destruction) and impulsivity were related to these variables (see Table 2.6).

The results of the regression with *timeline cyclical* as the dependent variable (DV) indicated that the model accounted for 15% of the variance ($R^2 = .15$, $F(2,62) = 5.64$, $p = .006$). The results of the regression for with *parental responsibility* as the DV indicated that the model accounted for 16% of the variance ($R^2 = .16$, $F(2,62) = 5.88$, $p = .005$). Impulsivity was the only factor that remained significant in both regressions.

Table 2.6.

Simultaneous regression analyses of associations between impulsivity and challenging behaviour (self-injury, aggression and property destruction) on parental perceptions.

DV	R ²	IVs	B	SE (B)	β	t	p
Timeline Cyclical	.15	(constant)	11.18	1.07	11.18	10.49	< .001
		Impulsivity	.17	.06	.17	2.74	.008
		CB	.39	.51	.29	.77	.444
Parental Responsibility	.16	(constant)	20.00		20.00	15.87	< .001
		Impulsivity	.16	.07	.16	2.23	.029
		CB	.94	.60	.94	1.56	.123

2.5. Discussion

There were three aims in this study; firstly, to explore the utility of Leventhal's Self Regulatory Model for studying CB in parents of adult children with ID; secondly, to explore associations between variables on the IPQ and other measures of parental perceptions and parental well-being (depression and anxiety); finally, to explore the associations between CB and parents' perceptions and the impact of child characteristics on these associations. In particular, the role of factors that fall under the broad umbrella of 'behavioural dysregulation' were explored i.e. impulsivity, over-activity, and repetitive behaviour. This study's strength is that it employed a robust measure of parental perceptions (IPQ) and separated behavioural dysregulation characteristics from self-injury, aggression and property destruction so specific associations could be explored.

It was predicted that the Self Regulatory Model would have utility within this population and that a similar pattern of inter-correlations would be found to other populations where this measure has been applied (Hagger & Orbell, 2003). Specifically, it was anticipated that beliefs in a chronic timeline, more severe consequences and lack of personal control would be associated with CB scores

(self-injury/aggression/property destruction), but that this link might be partially explained by behavioural dysregulation.

It was demonstrated that the Self Regulatory Model might be a useful measure for exploring parents' perceptions of self-injury, aggression and property destruction in their adult children. Patterns of inter-correlations between IPQ subscales were aligned with other populations (Hagger & Orbell, 2003). For example, stronger beliefs in negative consequences of CB on parents were found to be related to a stronger belief in chronic and cyclical timeline and more negative emotional representations. However, the inter-correlations did not completely replicate patterns from other populations, for example, scores on the *consequences* subscale were not associated with either the *personal* or *treatment* control subscales and the *illness coherence* subscale was not related to the *consequences* or *emotional representations* subscales as would be expected (Moss-Morris et al., 2002). Despite this the trends in the data are in the expected direction between these IPQ subscales so it may be that some of these correlations are underpowered due to the small sample size. Overall, these inter-correlations are promising and suggest that adopting models from health psychology may be useful for understanding how parents make sense of and understand self-injury, aggression and property destruction.

Previous studies have shown that IPQ subscales, such as *consequences*, predict poor psychological well-being (Hagger & Orbell, 2003). Inter-correlations on the IPQ highlight a relationship between greater perceived negative consequences for the parent and greater emotional representations. The emotional representations subscale is different from the HADS because it measures reactions about self-injury, aggression and destruction specifically whereas the HADS is a measure of global psychological distress (Moss-Morris et al., 2002; Zigmond & Snaith, 1983). Cause and effect cannot be established due to the correlational design; however, these results suggest that reducing the impact of self-injury, aggression and destruction on parents might improve emotional responses to CB, or that helping parents build resilience to these ongoing behaviours may reduce their sense of negative consequences. It may be that parents with poorer emotional representations perceive greater consequences of behaviour due to a cognitive bias but that these consequences are not objectively different from those faced by other parents. The direction of the results could be established by attempting to measure variables such as average time spent managing behaviour per day, degree of social exclusion due to behaviour, risk of physical assault, financial impact, and comparing these measures to perceived negative consequences measured by the IPQ.

In terms of global psychological distress, a high proportion of parents reached the cut-off for anxiety (56.9%) and depression (30.7%) on the HADS. Heightened anxiety and depression in this group is consistent with studies and appears typical for ID populations. For example, White and Hastings (2004) found that in parents of adolescents with severe ID 61% fell above the cut-off for

anxiety and 36% for depression on the HADS. Hastings and Brown (2002) also employed the HADS and found a similar profile of mental health difficulties in mothers of children with Autism and ID (54% anxiety; 38% depression). As the current study reports on an older cohort of individuals, it suggests anxiety and depression might be chronic in parents with children with ID. Overall, it highlights the vulnerability of this group and the need for interventions that are targeted specifically at the mental health of parents with older children.

The variable that was most strongly positively associated with global psychological distress measured by the HADS was parents' rating of their child's control over them, measured by the PLOC. The PLOC does not assess perceptions of self-injury, aggression and destruction, but is a generic measure of parenting locus of control. The relationship between *child control over parent* and psychological distress fits with research that highlights the role of parental locus of control in predicting parental distress (Lloyd & Hastings, 2009). The PLOC was moderately associated with all subscales on the IPQ except for *timeline-cyclic* suggesting that locus of control might be partially important when studying perceptions about self-injury, aggression and destruction. Parents with more external locus of control had beliefs about behaviour on the IPQ that have been associated with poorer coping and adherence to intervention strategies in the wider literature (Cooper et al., 1999; Heijmans, 1998; Heijmans and de Ridder, 1998; Moss-Morris et al., 1996; Scharloo et al., 1998, 2000; Weinman et al., 2000). It may be that targeting parental locus of control or its correlates (e.g. perceived negative consequences of CB, beliefs in available treatment and emotional representations) may be beneficial clinically when attempting to reduce anxiety and depression. These results fit with broader literature that points to a link between locus of control and depression (Bensassi et al., 1988).

The *cause* scale was developed for this study and needs further validation to explore test-retest reliability and construct validity. However, the patterns of results from this measure appear logical. For example, parents who perceived CB as being caused by situational factors scored more highly on personal control, (IPQ), were more likely to have greater internal self-efficacy and a sense that the behaviour was their responsibility relative to other parents. This might be because parents have experienced some mastery over behaviour, or because these beliefs give them a stronger sense that the behaviour is controllable even when the behaviour continues. Conversely, parents who believed that the behaviour was caused by underlying brain difference were less likely to have a coherent sense of self-injury, aggression and destruction, were more likely to have external self-efficacy and there is tentative evidence to suggest they are more likely to rate their children as having less control over behaviour (measured by the CBS). This appears to indicate a more deterministic view of challenging behaviour. Similar associations between parent beliefs in a biological cause and lower controllability have been found in ADHD populations without ID (Johnston & Freeman, 1997)

It is of interest that a belief in operant reinforcement was the only causal factor associated with

a belief that there are effective treatments for CB and this was related to a heightened sense of parental responsibility. Despite this, parents were least likely to view operant conditioning as a cause of aggression/self-injury/destruction, which suggests that they do not endorse a functional account of behaviour. Overall, parents tended to endorse situation/environment as the main cause of CB, along with atypical brain development and mood. Given that families are most likely to endorse situational factors as underpinning behaviour, it appears they might accept the 'antecedent' aspect of a functional account of behaviour while not linking this with the impact of reinforcement through consequences. This result fits with Oliver et al. (1996) who found that carers often lack specific behavioural knowledge. This pattern of results points towards one of three explanations: that families have not received information about reinforcement and learning theory; that families have had information but do not feel it can explain their child's behaviour; or families find it difficult to respond honestly to direct questions about behaviour being learnt or taught. This is clearly an area where further investigation is warranted because each one of these explanations is likely to have an impact on the chronicity of CB overtime, the likelihood of parents seeking help from professionals, and the likelihood of the advice parents receive from professionals fitting with their views about behaviour. If it were found that parents have had no exposure to information about reinforcement of challenging behaviour it would fit with accounts that families with older children may be members of a generation who have not had access to the support from services and who may have encountered a lack of professional awareness as to the needs of their children with genetic syndromes (Quine & Pahl, 1986; Quine & Rutter, 1994). Again, this highlights the need for improved intervention strategies to target the needs of this vulnerable group.

The final aim was to explore the influence of child characteristics on parental perceptions of CB, self-injury, aggression and destruction, and parental well-being. Analyses supported previous research findings linking impulsivity, hyperactivity and repetitive behaviour with self-injury, aggression and destruction of property (Davies, 2010). Repetitive behaviour was the only aspect of the triad of impairments measured by the SCQ that was consistently associated with these behaviours, which fits with arguments that repetitive behaviour may be linked to underlying behavioural dysregulation (Oliver et al., 2009). Furthermore, the principal components analysis replicated the findings that impulsivity, over-activity and repetitive behaviour do load onto one factor (Oliver et al., 2009). Associations were also found between verbal ability, self-injury and aggression, which fits with previous research findings (McClintock et al., 2003). It is clear that a fine-grained approach that acknowledges the role of each of these individual factors is warranted.

When associations between child factors and parental perceptions were explored the complexity of the relationships between variables became apparent. Contrary to previous findings, more severe CB (self-injury, aggression and property destruction) was not found to be related directly

to anxiety and depression (Minnes et al., 2007). These behaviours were, however, related to more strongly held beliefs about negative consequences for the parents, chronic and cyclic timeline, child control over parent, and lower parental responsibility. As previously noted, a belief that their child has more control over their actions is associated with parental depression and anxiety. This suggests that while self-injury, aggression and destruction were not associated directly with anxiety and depression, the extent of these behaviours contributes to factors that are directly associated with poorer well-being. An alternative explanation for why higher CB score was not associated with lower parental well-being is that parents were only recruited if they had indicated at two previous time points that their child engaged in self-injury, aggression or destruction. This meant there might have been fewer parents in the sample experiencing low levels or absence of CB coupled with higher well-being and the inclusion of these parents might have strengthened the association.

The behavioural dysregulation hypothesis was explored and it was found that impulsivity was moderately correlated with a belief that behaviour is unpredictable/comes and goes (*time-line cyclical*), lower parental responsibility and belief in syndrome as a causal factor. Regression analyses indicated that, while the CB composite score was related to timeline-cyclical and parental responsibility, only impulsivity remained significant when entered into the regression. This was true even though impulsivity was measured at an earlier time-point, an average of eight months earlier. Overall, it appears that impulsivity might independently predict aspects of the Self Regulation Model. Although results should be interpreted with caution due to the number of correlations conducted, this fits with the predictions of this study and warrants further investigation. Impulsivity might be related to lower levels of parental responsibility and a belief in an unpredictable cyclical timeline because parents have experienced that their child's behaviour is related to other factors apart from their actions or the environment, such as their child's level of dysregulation, and do not feel solely responsible for their child's behaviour. Lowered levels of parental responsibility has been observed in parents of children with a diagnosis of ADHD without ID, and it has been argued that knowledge of this diagnosis might have affected parents' perceptions (Johnston & Freeman, 1997). However, children in the current sample did not have a diagnosis of ADHD suggesting that parents' direct *experiences* of their children's behaviour may have affected their perceptions. Alternatively, as direction cannot be established in this study, the association between parental responsibility and impulsivity may be bi-directional and fit into an attachment model of behavioural regulation whereby early attachment difficulties may proceed impulsive behaviour (Janssen et al., 2002; Olson et al., 1990). These findings illustrate the importance of specificity when measuring child characteristics.

The limitations of this study are the correlation design and the specific focus on adults with ID. The number of correlations and the use of non-parametric statistics means that both type-I and type-II errors are more likely in this type of study. It is also not possible to know the direction of

associations. The study could be improved by longitudinal methodology or by a cross sectional approach that incorporates a younger group of individuals so that the parental perceptions can be tracked developmentally. If it were possible to increase sample size it would be interesting to explore whether these patterns are representative of all syndrome groups included as there was some evidence that individual syndrome groups, such as Angelman syndrome, may drive particular associations. Additional limitations relate to the measures used in this study and the adaptations made to them for use with this population. While the measures have been used in previous research and have been shown to be robust, alterations may have changed their psychometric properties and further work is needed to validate these measures in ID populations. However, the newly developed *cause* subscale appears particularly promising as a measure, and all measures employed had good face validity after the adaptations. Finally, the results are likely to only be generalisable to mothers in this age band due to the high proportion of the sample that were female. Previous research has shown differences between fathers and mothers and if a higher proportion of the sample had been fathers it would have been possible to explore this statistically (Hastings & Brown, 2002; Hastings et al., 2005).

Overall, these results highlight the importance of continuing to study factors associated with behavioural dysregulation separately from self-injury, aggression and property destruction in parents and carers of people with genetic syndromes and ID. In addition, they point towards utilising models from health psychology to understand parents' perceptions of behaviour.

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PUBLIC DOMAIN BRIEFING PAPER

Parents' thoughts about self-injury, aggression and property destruction in their children with intellectual disabilities, and the influence of behavioural dysregulation

Jane Waite

University of Birmingham

What was this study about?

Prior to starting this study, previous research was examined that has explored well-being of parents of children with intellectual disabilities. It was found that child behavioural difficulties are often linked to poorer parental well-being (Hastings, 2002). However, 'behavioural difficulties' is often a very broad term and includes many types of behaviours. These behaviours are regularly grouped together in studies and it is difficult to know which behaviours contribute most to poorer parental well-being. For example, impulsivity, hyper-activity, repetitive behaviour, self-injury, aggression, and destruction of property might be all measured together and combined into one score that is then used in the analysis. However, it is important to know which behaviours contribute most to parental well-being because factors associated with behavioural dysregulation (impulsivity, hyper-activity, repetitive behaviour) might impact on parental well-being in a different way to self-injury, aggression or property destruction, or might influence how parents think and feel about these behaviours.

An empirical study was conducted to explore the impact of behaviours associated with behavioural dysregulation on parents' perceptions of self-injury, aggression and property destruction. Parental perceptions were examined using a framework called the Self Regulatory Model of Illness Behaviour (Leventhal, Nerenz & Steele, 1984). This model captures a wide range of perceptions and seeks to explain how individuals form an internal representation of an illness or disorder. The model has been adapted in the past to explore carers' experiences of their relatives illness but has also been adapted to explore Autism Spectrum Disorder and behavioural difficulties in individuals with intellectual disabilities (Williams & Rose, 2007; Al Anbar, Dardennes, Prado-Netto, Kaye & Contejean, 2010). An additional aim was to explore whether the Self Regulatory Model could be used with parents of adults with intellectual disabilities and to learn about parents' perceptions of self-injury, aggression and property destruction.

It was predicted that the Self Regulatory Model would be a useful model for understanding parental perceptions of challenging behaviour in intellectual disability populations and that similar relationships between parts of the model would be present as in other populations where it has been

used. In addition, it was predicted that behavioural dysregulation would impact on parental perceptions of self-injury, aggression and property destruction.

How was the study conducted?

The empirical study separated behaviours that might be indicative of behavioural dysregulation (impulsivity, repetitive behaviour and over-activity) from behaviours most frequently reported as challenging by parents: self-injury, aggression and property destruction. Sixty-five parents of adult children with intellectual disabilities completed questionnaires about self-injury, aggression and property destruction and answered questions about their perceptions of these behaviours. Perceptions included parents' views about the timeline of the behaviour, the likelihood of it having a negative consequence for them, the availability of effective interventions, whether parents felt the behaviour made sense, their emotional reactions to the behaviour, their beliefs about the cause of behaviour and their sense of personal control over the behaviour. The questionnaires also measured parents' beliefs about the controllability of the behaviour and their mental health. The responses to these questionnaires were analysed with reference to information parents had given us several months earlier about impulsivity, over-activity and repetitive behaviour in their children.

What did the study find?

In agreement with the study predictions, the Self Regulatory Model appears to be useful for studying parents' perceptions of self-injury, aggression and property destruction. This is because associations between different parental perceptions showed a similar pattern to other groups of people where it has been applied (Hagger and Orbell, 2003) . In particular, if parents believed that self-injury, aggression or property destruction would last a long time they were more likely to believe it would have negative consequences for them. A weaker belief in *child control over parent* was related to better mental health and fewer perceived negative consequences; however, overall a large proportion of the parents' that took part experienced clinically significant levels of anxiety and depression (58.9% and 30.8% respectively). Finally, we know from previous research that self-injury, aggression and destruction of property are often learnt over time because the behaviour has particular reinforcing consequences for an individual (Carr & Durand, 1985). However, when perceptions of the cause of the behaviour were measured very few parents agreed with this.

Behavioural dysregulation variables, impulsivity, repetitive behaviour and over-activity, were found to be strongly related to each other. This suggests that these behaviours might be underpinned by a

common cause. Behavioural dysregulation was strongly correlated with behaviours such as self-injury, aggression and property destruction. This replicates previous findings that the presence of markers for behavioural dysregulation increases likelihood of the presence of self-injury, aggression and property destruction (e.g. Oliver, Petty, Ruddick, Bacarese-Hamilton, 2012). When the impact of behavioural dysregulation variables on parents' perceptions were explored it was found that impulsivity was related to a belief that self-injury, aggression or property destruction is less predictable, and lower responsibility for their child's general behaviour. The degree of self-injury, aggression or property destruction experienced by families was independently related to other parental perceptions such as a belief that self-injury, aggression or property destruction would last a long time and a belief in more negative consequences for the parent.

Why is this important?

These results have identified that mental health difficulties are common in parents of older children with disabilities and that more resources need to be targeted towards alleviating psychological distress. In addition, the results are important because parents thoughts about behaviour are likely to be closely related to parental stress. Often it is argued that parental thoughts about behaviour can explain why some parents experience greater stress than others, and that changing parents' thoughts about self-injury, aggression and property destruction might then lead to changes how parents manage these behaviours. However, the link between impulsivity and lower parental responsibility and a belief that the behaviour is unpredictable might indicate that parental perceptions can also be shaped by their experiences of their child's behavioural dysregulation. This suggests that if behavioural dysregulation is taken into account when working with families of individuals with intellectual disabilities clinician's might develop a better understanding of the families' needs. In addition, the finding that very few parents believed that self-injury, aggression and property destruction could be shaped over time suggests a gap between professional knowledge and dissemination to parents that could be addressed to increase the likelihood of successful interventions.

What should happen next?

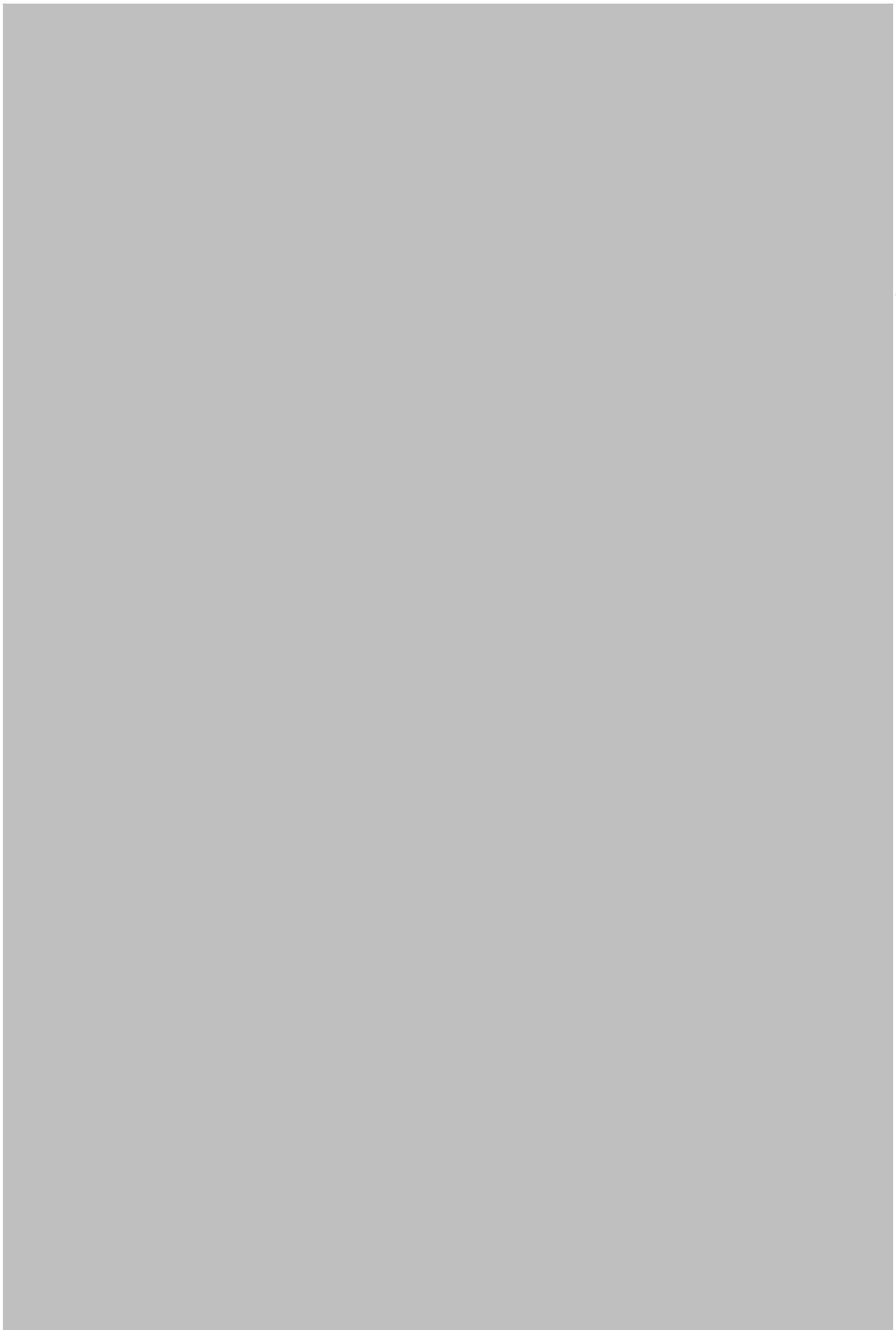
It is clear from this research that further studies exploring associations between parental well-being and self-injury, aggression and property destruction should measure these behaviours carefully and separate them clearly from impulsivity, over-activity and repetitive behaviour. Furthermore, behavioural dysregulation should be considered when clinician's are assessing self-injury, aggression and property destruction as it may influence outcomes for families. The Self Regulatory Model provides a promising framework for understanding parents perceptions of behaviour and could be used in further research.

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APPENDIX A









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APPENDIX B



UNIVERSITY OF
BIRMINGHAM

Address Block]

[REF: Participant

Number]

[Date]

Re: [Child's Name]

Dear [Parent's Name]

You may remember that you have taken part in our research before by completing questionnaires about [child's name]. We hope you found the feedback that we sent to you helpful.

We are writing to you because your responses on these questionnaires indicate that [child's name] has been engaging in challenging behaviour over a number of years. We are interested in learning more about your experiences. In particular, we are interested in learning more about the characteristics of children with challenging behaviour, your views about challenging behaviour, and the impact of challenging behaviour on your life. This is the first study to explore these questions in adults with [syndrome name] syndrome who have engaged in challenging behaviour over a long period of time and the results of this study will be important for understanding challenging behaviour and developing future interventions. The more people that take part in the research, then the more meaningful the results will be.

We are contacting you because you have agreed for your personal details to be kept at the Cerebra Centre for Neurodevelopmental Disorders at the University of Birmingham, and to be contacted with information about future research at the centre. We would like to invite you and [child's name] to complete a brief online questionnaire study specifically about challenging behaviour. This can be accessed using the web address below, or you can request a paper copy by calling [redacted]. If you have provided us with an email address we will also be sending you this link via email.


INSERT LINK

Password: [redacted]

ID number: [participants number]

Please ensure that you enter your unique ID number [participant number] on the online questionnaire so that we can trace it back to you. You will be asked to enter this number when you begin the questionnaire.

When we have analysed what you tell us, we will provide feedback about our findings. There is an information sheet enclosed that gives you more details about why the research is being carried out and what it will involve.

Please read the information sheets before completing the questionnaire and if you are unclear about any aspect of the study or have any questions then contact Professor Chris Oliver at the address below or on 

Thank you for your time and we look forward to hearing from you.

Yours sincerely



Chris Oliver

Professor of Neurodevelopmental Disorders

Understanding Challenging Behaviour in Neurodevelopmental Disorders: Information Sheet

Who should complete the questionnaire

We are recruiting parents/caregivers of people with challenging behaviour aged between 4 and 60 years inclusive.

Background and aims of the study

We would like to invite you to take part in a questionnaire study being conducted at the Centre for Neurodevelopmental Disorders, University of Birmingham. The aim of this research is to improve our understanding of the causes of challenging behaviour and its impact on parents.

What will happen if you and your child/the person you care for decide(s) to participate?

Where will the research take place?

The research will involve completing the online questionnaire at:

[insert link]

This can be completed by you in your own time. If you would like to fill out a paper copy, please contact Dr Jane Waite or Professor Chris Oliver on [redacted].

How long will participation in the study take?

The questionnaire will take approximately 20-30 minutes to complete.

What will participants be required to do during the study?

If you decide to take part you would be asked to complete the online consent form and questionnaire pack. At the end of the questionnaire you will be asked whether you would like to participate in an interview at a later date about help you may or may not have received for challenging behaviour. This interview is entirely optional so you can make up your mind about this later on without it impacting on your participation in the online questionnaire study.

Are there any risks that individuals taking part in the study might face?

There will not be any risks associated with participation in this study.

What are the potential benefits for participants from taking part?

This study will help us to find out more about the causes of challenging behaviour and parents' experiences of this behaviour. The findings from this research may be very important in helping us to develop future behavioural intervention programmes.

Where will data be stored?

The data collected will be kept in password protected storage at the University of Birmingham and on high security servers at the University of Birmingham. Only members of the research team at the University of Birmingham and our colleagues at NAME will have access to information that we collect about you. Information will be treated as strictly confidential and handled in accordance with the provisions of the Data Protection Act 1998.

If you/ the person you care for decide(s) to participate, what will happen after that participation?

You and your child or person you care for will receive an individual feedback report describing the results of all of the assessments that were carried out during the study. Descriptions of research findings will be published in newsletters of the relevant family support groups and educational

institutions involved. Any request for advice concerning the person you care for will be referred to Professor Chris Oliver, Clinical Psychologist. The researchers will publish the findings from the study in scientific journals and will present the results at relevant conferences.

What will happen to the data afterwards?

The information that you provide will be held on a password protected database and on servers in a high security data centre. As you have previously indicated that you would like us to retain your personal details for future studies we will not destroy your details unless you tell us otherwise.

Consent

When you log on to the online study you will be asked to give your consent to participate in the study if you decide that you do wish to participate. We need to receive consent from you and the person you care for before you can participate. If the person you care for does not have the capacity to consent you can consent by yourself (for under 16s only) or act as a personal consultee for this individual (over 16s).

Withdrawal

Even after consent has been granted, you or your child/person you care for can request to be withdrawn from the study up to 6 months after participation, without giving a reason. This will not restrict the access to other services and will not affect the right to treatment.

Confidentiality

Every effort will be taken to protect the information that you provide. If published, information on participants will be presented without reference to their name or any other identifying information. All personal details held at the University of Birmingham will be kept separately from the data collected so that it will only be possible to connect results to individuals via a special code. This will ensure that results are kept anonymous. We will only disclose personal information to appropriate authorities if we feel that either you or the person you care for is at risk of harm above and beyond what would be anticipated given that your child engages in challenging behavior.

The company with whom we have chosen to host our questionnaires (NAME) adheres to stringent security practices. However, as is always the case when using the internet, there is a possibility that agents (e.g. 'hackers') might attempt to access the information that you provide. Please participate in this research only if you are comfortable with this risk. In the unlikely event of any evidence of abuse being identified, this information will be disclosed by the research workers.

Review

The study has been approved by the NRES Committee West Midlands - Coventry & Warwickshire. The study forms part of an educational project and has been reviewed by Research Governance & Ethics, Research Support Group, University of Birmingham.

Further information

If you would like any more information about the study please contact Dr Jane Waite or Prof Chris Oliver on [REDACTED]. Alternatively, write to Chris Oliver, School of Psychology, University of Birmingham, Edgbaston, Birmingham, B15 2TT.

If you would like to make a complaint about this research please contact Brendan Lavery, Head of Research Governance & Ethics, Research Support Group, University of Birmingham. [REDACTED]

[REDACTED]

APPENDIX C

CAPACITY TO CONSENT

You need to decide whether your child/the person you care for is able to understand enough about the study to make an 'informed' decision independently about whether or not they would like you to participate and to communicate this decision to you. If you are unsure whether or not your child/person you care for is able to understand enough to make a decision independently then we can provide you with some guidelines to help you to assess this. A symbol information sheet can also be made available to you if this would be of help.

*Please contact Dr Jane Waite [redacted]
[redacted] to request a copy of this.*

If your child has the capacity to consent just complete
consent form A

If your child **does not** have the capacity to consent
please read the 'Acting as a Personal Consultee'
information sheet and just complete **consent form B**

By ticking the boxes and clicking on 'Next' at the bottom of the page you are consenting to participate in this survey.

These questions are to be completed by the **person you care for.**

Please read the following statements and tick each one you agree with. **This section is required.**

- Somebody has explained the project to me or I have read the information ☐
- I understand what the project is about ☐
- I have asked all of the questions they want ☐
- I have had my questions answered in a way they understand ☐
- I know it is OK to stop taking part at any time ☐
- I am happy to take part ☐

These questions are about you (the person who will be completing the questionnaires). Please read the following statements and tick each one you agree with. **This section is required.**

- I have read and understood the 'Understanding Challenging Behaviour in Neurodevelopmental Disorders' information sheet (ref: ISW/V1/16.04/2012) [\[weblink to information sheet\]](#). ☐
- I understand that I do not have to take part in the study if I do not want to ☐
- I understand that my participation is voluntary and that I am free to withdraw at any time without giving any reason, without my or that of my child's/person I care for's medical care or legal rights being affected ☐
- I understand that all information collected during the study will be confidential. All information collected during the study will be held on secure servers by the hosting website and then transferred to locked cabinets that only members of the research team will have access to. No names will be published in any reports. Information will be treated as strictly confidential and handled in accordance with the provisions of the Data Protection Act 1998. ☐

- I understand that the company who are hosting the questionnaires (NAME) adheres to stringent security practices. However, as is always the case when using the internet, there is a possibility that agents (e.g. 'hackers') might attempt to access the information that you provide. I understand these risks and am comfortable with them.

☐

- I would like to give my 'informed' consent to take part in the study 'Understanding challenging behaviour' ☐

Before deciding whether to participate, please ensure you read the information on acting as a [personal consultee \[insert link\]](#) for the person you care for. By ticking the boxes and clicking on 'Next' at the bottom of the page you are acting as a personal consultee and consenting on behalf of the person you care for to participate in this survey.

Please read the following statements and tick each one you agree with. ***This question is required**

1. I have been consulted about the person I care for's participation in the research project titled 'Understanding Challenging Behaviour in Neurodevelopmental Disorders'. I have read the information sheets (refs: ISW/V1/16.04.2012 & PC/V1/16.04.2012) [\[weblink to information sheets\]](#) and had the opportunity to ask questions about the study and understand what is involved. ☐
2. In my opinion he/she would have no objection to taking part in the above study. ☐
3. I understand that I can request he/she is withdrawn from the study at any time without giving any reason and without his/her care or legal rights being affected. ☐
4. I understand that all information collected during the study will be confidential. All information collected during the study will be held on secure servers by the hosting website and then transferred to locked cabinets that only members of the research team will have access to. No names will be published in any reports. Information will be treated as strictly confidential and handled in accordance with the provisions of the Data Protection Act 1998. ☐
5. I understand that the company who are hosting the questionnaires (NAME) adheres to stringent security practices. However, as is always the case when using the internet, there is a possibility that agents (e.g. 'hackers') might attempt to access the information that you provide. I understand these risks and am comfortable with them. ☐
6. I agree to take part in the study titled 'Understanding Challenging Behaviour in Neurodevelopmental Disorders'. ☐

Ref: PC/V1/16.04.12

Personal Consultee Information Sheet

Please read this information sheet if you care for a person who you have judged *is not* able to make an 'informed' decision about whether or not they would like to take part in the study or *is not* able to communicate that decision to you.

We would like to invite you to act as a **personal consultee** for the person that you care for.

Information for Personal Consultees

What is a Personal Consultee?

In order to understand illness and disability, and to improve treatment and care, research is essential. That research may focus on the people with the illness or disability or on children under the age of 16, and may invite those people to participate. Some people will have capacity to make their own decision whether to take part in the research.

Others, possibly those most affected by the illness or disability, may not have that capacity. They may not be able to understand enough of the research to be able to give 'informed consent'. They may not be able to communicate a decision. The research provisions of the Mental Capacity Act are designed to allow such people to take part in research even though they cannot give valid consent of their own.

First, the research has to be approved by a Research Ethics Committee. Then, instead of asking the research participant for consent, the researcher must ask a consultee for an opinion whether the research participant would have wished to take part in the research.

Who can be a personal consultee?

Any person interested in the welfare of the proposed participant, for example:

- A family member, unpaid carer or friend
- A person acting under a Lasting Power of Attorney
- A court appointed deputy

Who cannot be a personal consultee?

- Paid carers and professionals
- People connected with the research (e.g. members of the research team)

Why have I been asked?

You have been asked to act as a personal consultee by a researcher because the researcher thinks you might be willing and able to do this because of your close relation with the proposed research participant.

If I agree to be a personal consultee, what will I have to do?

You will need to think about what the proposed participant's wishes and feelings about the research would be if they had capacity to make an informed decision and decide whether in your view the person should be involved in the research or not. This means you need to

- Look at the study information sheet [\[weblink to information sheet\]](#)
- Think about whether or not the person would want to be involved in the research project if he or she had the capacity to make that decision.

You should not put forward your personal views on participation in the specific project or research in general, you must consider only what the person's views and interests are or would likely be. You should think about:

- What the broad aims of the research and the practicalities of taking part will mean for the proposed participant.
- How the specific activities in the research might impact the participant.
- Any view previously expressed by the person on the overall nature of the research.

If you advise that the proposed participant would not have wanted to be involved in the research, they cannot be included in the research.

If you advise that the proposed participant would want to be involved, they may be included in the research. If the research commences but the person shows any sign at any stage that they are not happy to be involved in the research you can change your advice at any time without giving a reason, whereby the researcher must withdraw the person from the research. If the person seems unhappy at any point or shows any signs of objection, then they will be withdrawn from the research.

The research project has been approved by the Coventry NHS Research Ethics Committee. If you wish to see proof of approval from this body, or you wish to discuss any concerns about acting as a personal consultee for the person that you care for, please contact Chris Oliver

Where can I get more information and guidance?

More information is available from:

Department for Constitutional Affairs (2007) *Mental Capacity Act 2005 Code of Practice*

<http://www.dca.gov.uk/legal-policy/mental-capacity/mca-cp.pdf>

Department of Health (2007) *Guidance on nominating a consultee for research involving adults who lack capacity to consent* (consultation)

http://www.dh.gov.uk/en/Publicationsandstatistics/Publications/PublicationsPolicyAndGuidance/DH_076207

Mental Capacity Implementation Programme (2007) *Making Decisions: a guide for family, friends and unpaid carers. Second edition*

<http://www.dca.gov.uk/legal-policy/mental-capacity/mibooklets/booklet02.pdf>

A printed copy of this booklet is available by telephoning .

APPENDIX D

Questionnaire pack removed to protect details. Please contact first author.

APPENDIX E

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APPENDIX F

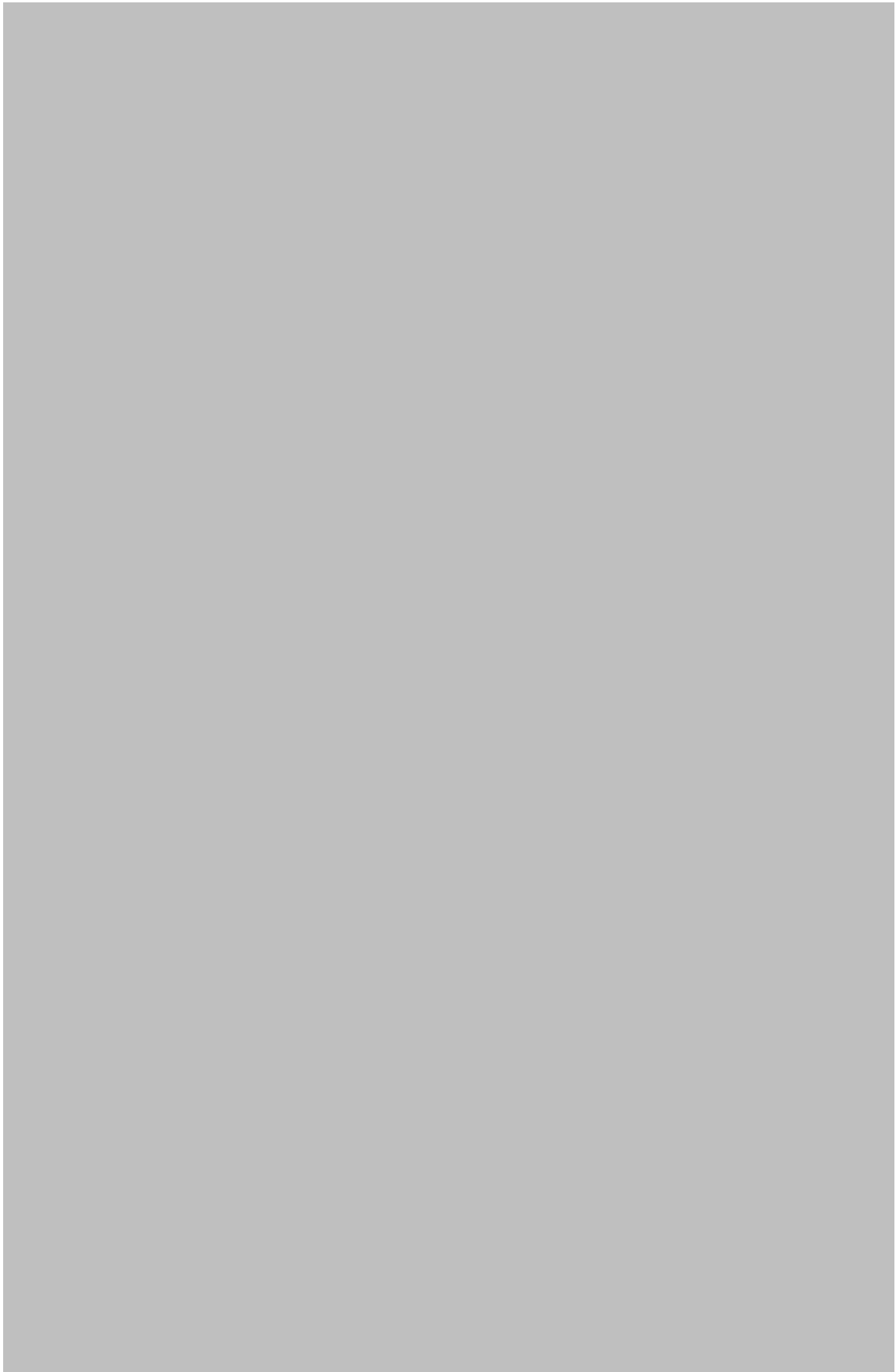
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APPENDIX G

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APPENDIX H

APPENDIX H



APPENDIX I



APPENDIX J

APPENDIX J

Internal consistency of the IPQ-R subscales and Cause Scale



APPENDIX K

APPENDIX K

Comparison of child characteristics in 2006 to 2011: Stability over time

Child Characteristic	2006 N	2006 Median (IR)	2011 N	2011 Median (IR)	Z	<i>p</i>	<i>r</i>	<i>p</i>
Impulsivity (TAQ)	49	19.00 (11.00)	64	18.00 (9.25)	-.996	.319	.56	< .001
Over activity (TAQ)	49	17.00 (14.00)	65	14.00 (17.00)	-1.50	.134	.76	< .001
Impulsive Speech TAQ	31	4.00 (8.00)	39	5.00 (8.00)	-.52	.607	.91	< .001
SCQ	45	8.12 (4.88)	65	7.00 (4.23)	-.265	.008	.54	< .001
Communication SCQ Social interaction	45	9.00 (5.50)	65	7.00 (5.00)	-.389	< .001	.67	< .001
SCQ Repetitive Behaviour	44	5.00 (2.00)	65	4.00 (2.50)	-1.71	.088	.61	< .001

Note. N varies due to missing data.

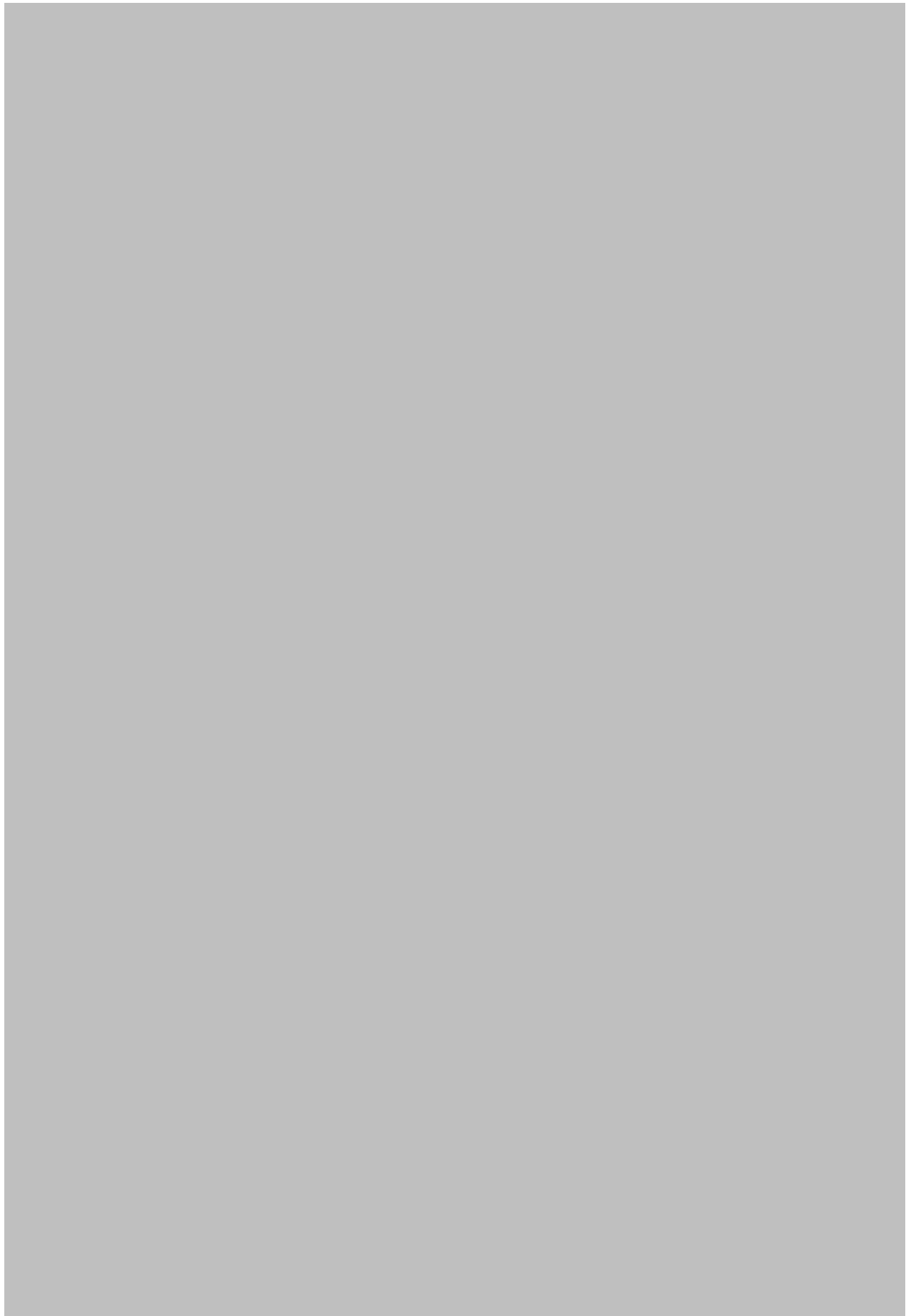
APPENDIX L

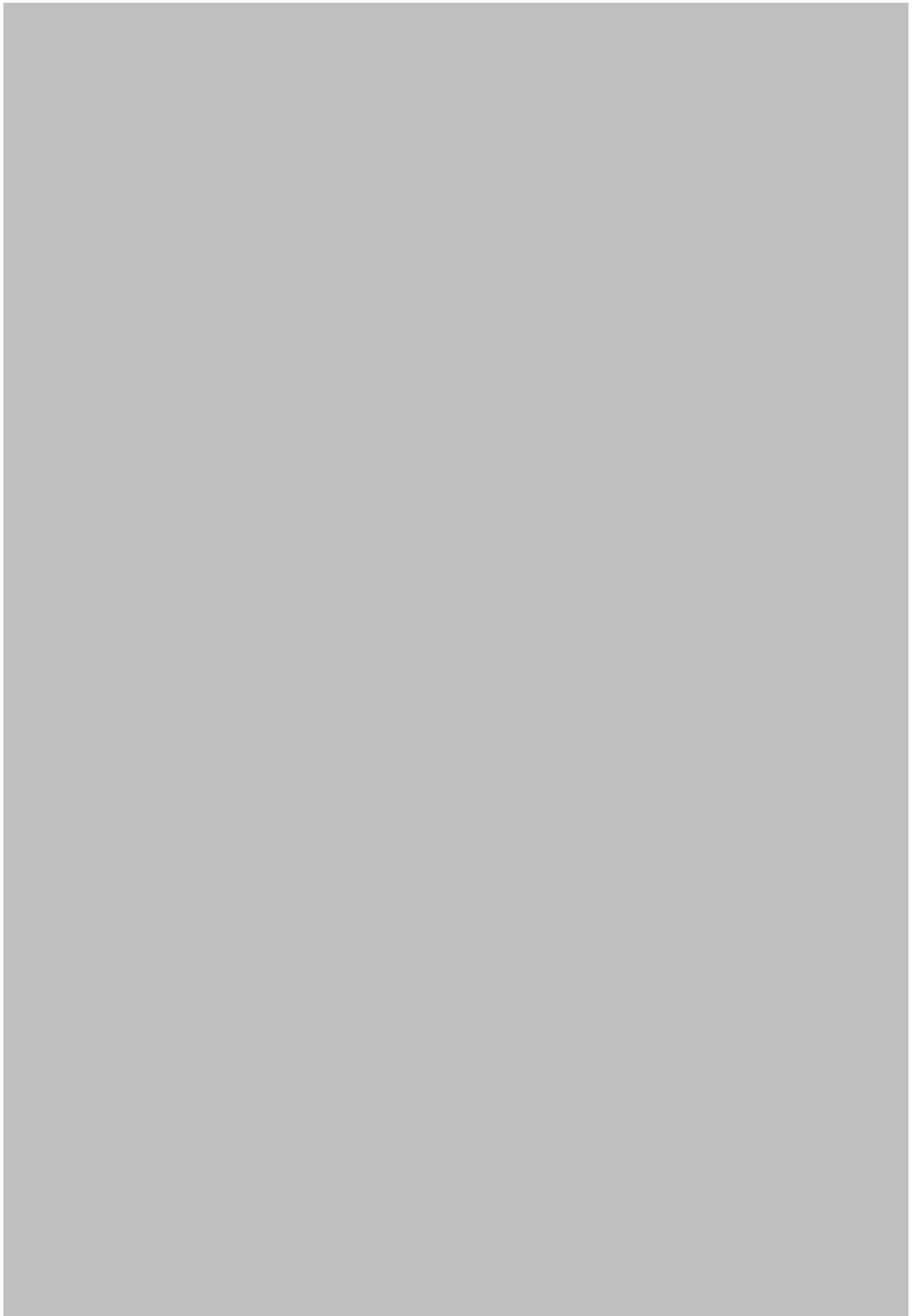
Research in Development Disabilities Guidelines

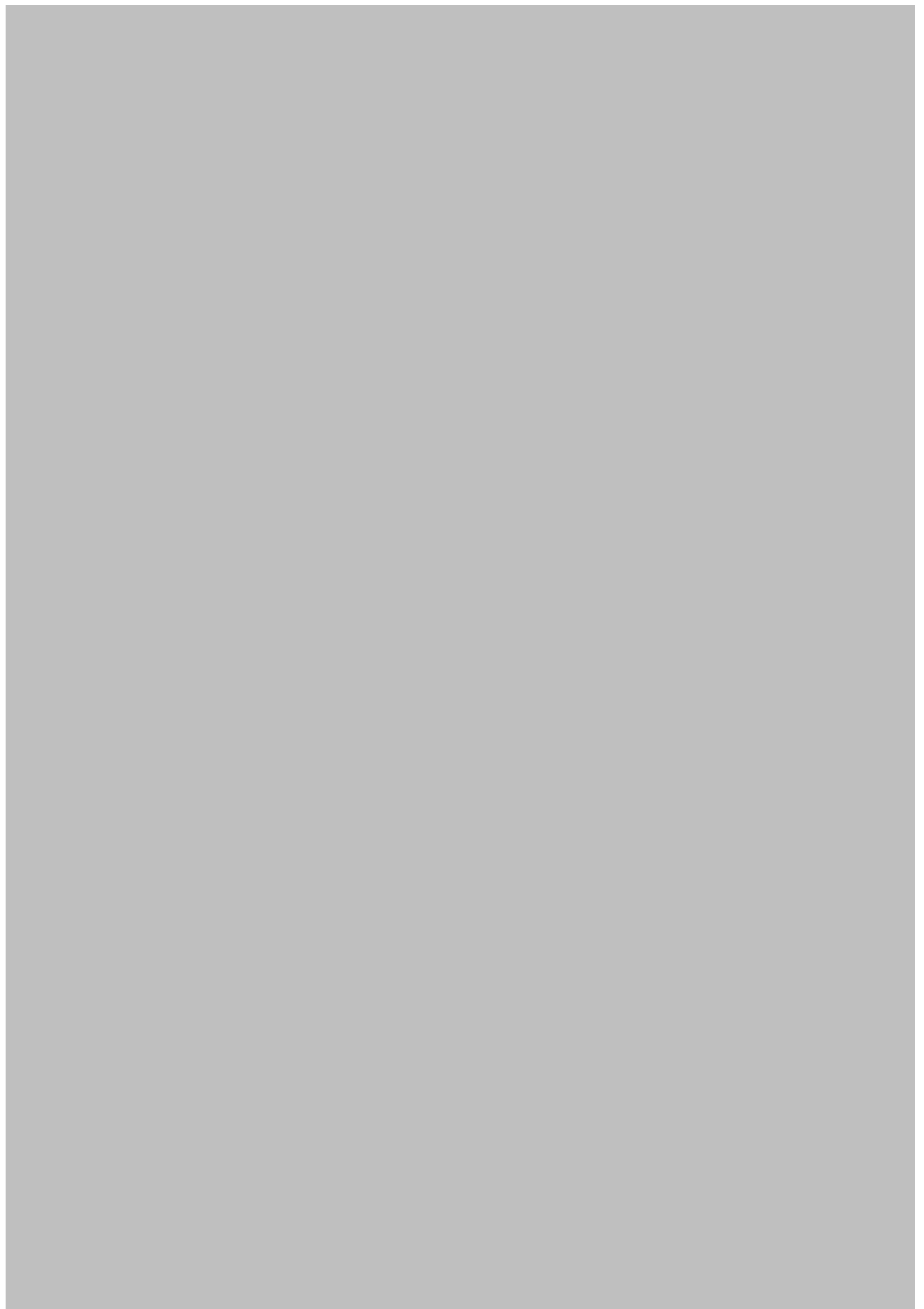
GUIDE FOR AUTHORS

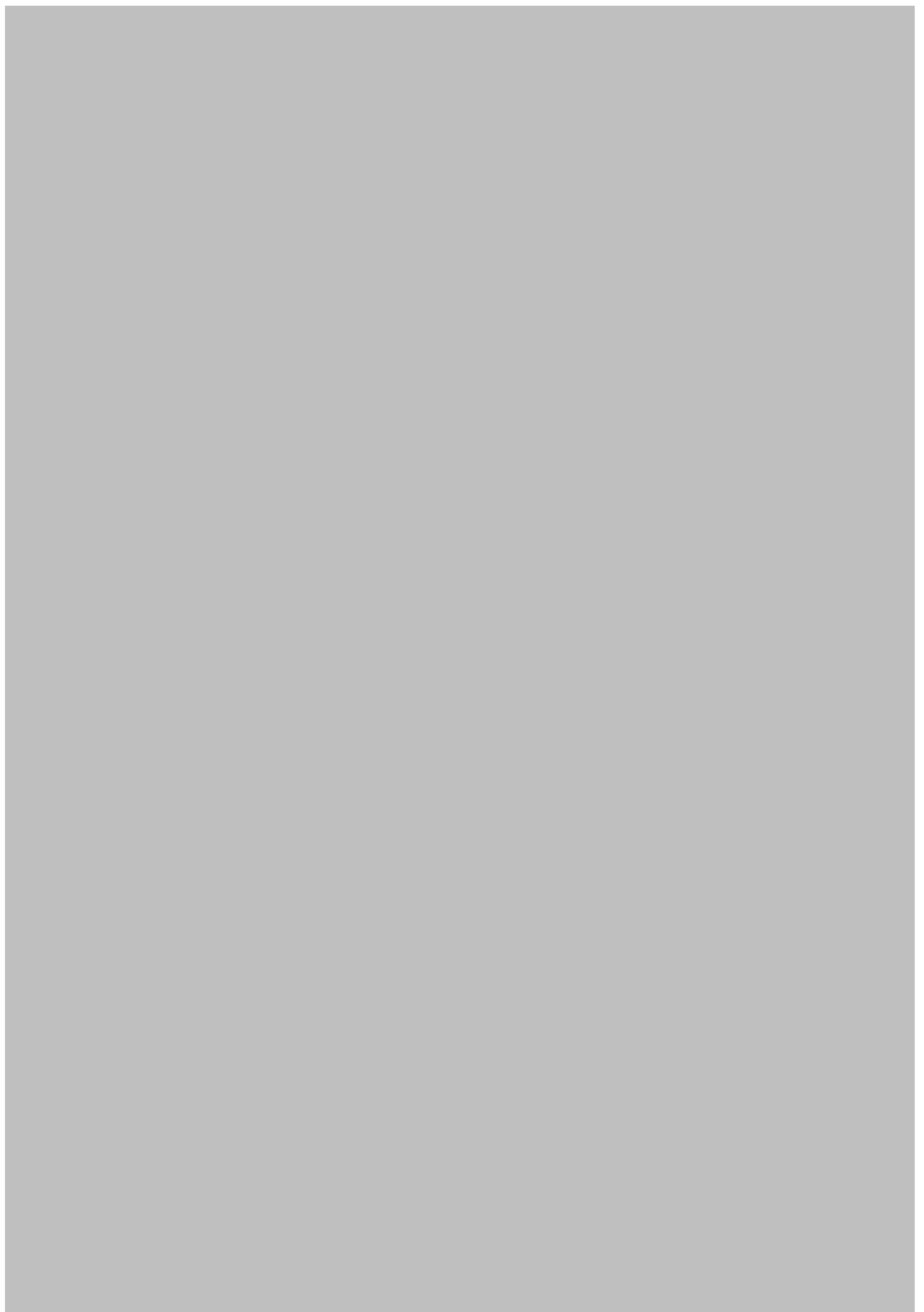


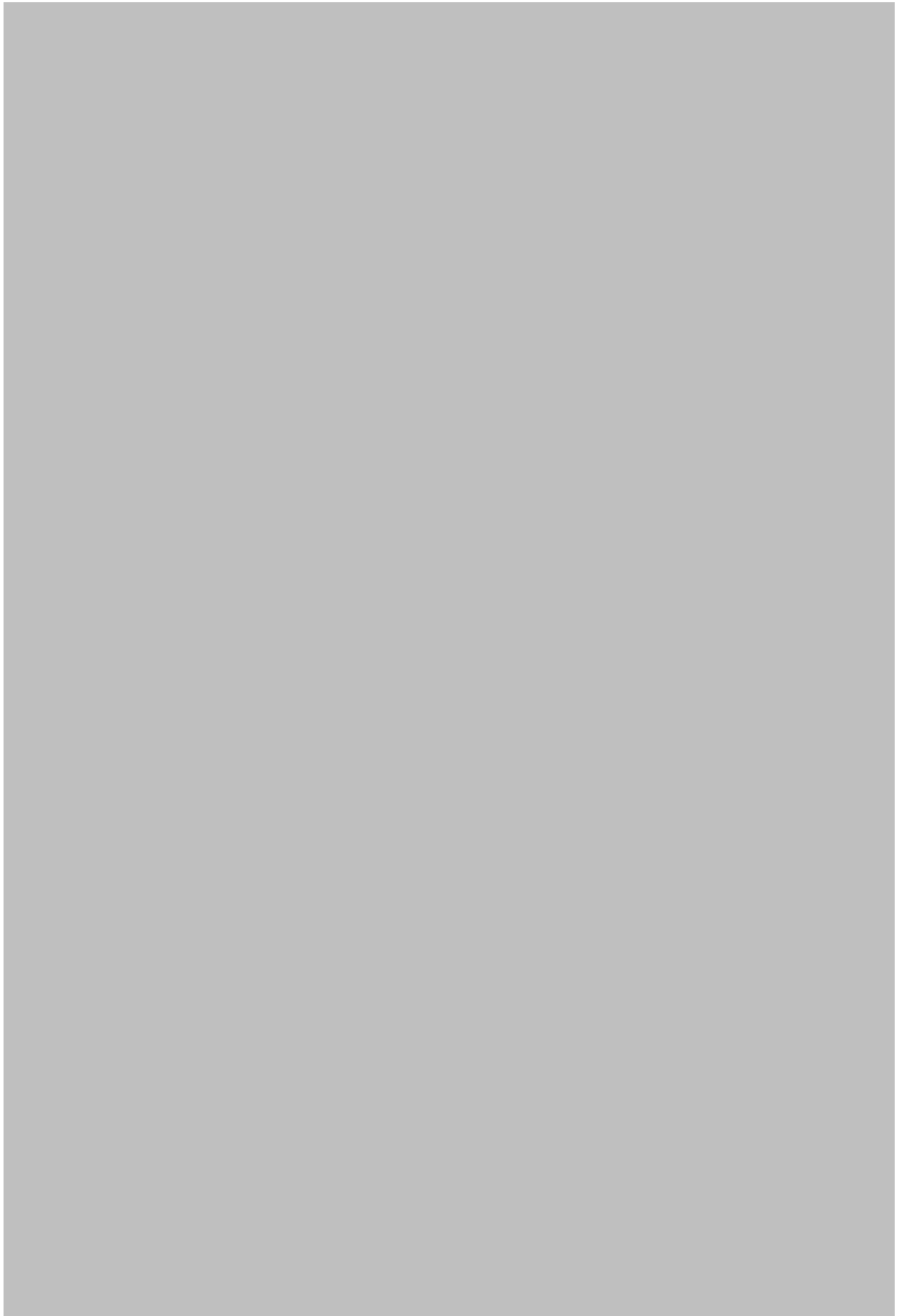














APPENDIX M

