

**Emotion Regulation and Executive Function in Children and Adolescents with
Autism Spectrum Disorder and Pathological Demand Avoidance Traits**

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Thesis declaration form

I confirm that the work presented in this thesis is my own. Where information has been derived from other sources, I confirm that this has been indicated in the thesis.

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Overview

This thesis focuses on gaining a greater understanding of some of the difficulties that may be related to Autism Spectrum Disorder (ASD). The literature review (Part 1) uses systematic search techniques to explore whether informant-reported Emotion Regulation (ER) impairments are present in children and young people with ASD. It also has a secondary aim of exploring factors which may impact the levels of ER impairment. Specifically, the impact of participant age, study quality, and parent- versus self-report are investigated.

The empirical research paper (Part 2) was conducted as part of a joint research project with Ellie Bishop, trainee clinical psychologist (see Appendix I for individual contributions). It investigates the relationship between parent-reported Executive Function (EF) and Pathological Demand Avoidance (PDA) behaviours in children with ASD. Firstly, Part 2 specifically focuses on understanding whether there are parent-reported deficits in EF in children with ASD and PDA traits. Next, it investigates the relationship between parent-reported EF impairments and PDA traits in general, then more specific PDA traits such as, affective dysregulation, and non-compliance behaviours.

The critical appraisal (Part 3) describes the process of conducting the literature review and the empirical study. To begin with, processes relating to the review are discussed, such as: the motivation behind the review, barriers and challenges encountered, and the implications of the review. Next, the appraisal describes the process of conducting the empirical study, in particular focussing on: the motivation for conducting the study, experiences of working with participants, measurement challenges, experiences of joint working, and the clinical and scientific implications. Finally, conclusions and recommendations for future research are made.

Impact Statement

The overarching aim of this research was to contribute to current understanding of cognitive processes and behaviours relevant to young people with Autism Spectrum Disorder (ASD). It is widely known that those who are affected by ASD struggle with a range of behavioural and cognitive difficulties. Epidemiological studies have reported increased rates of ASD diagnosis, and ASD services are under pressure to cope with growing demands. Therefore, research into these difficulties is needed to provide evidence to support intervention development, service delivery and increase awareness of ASD.

A literature review investigated self- and parent-reported Emotion Regulation (ER) in children and young people with ASD. Overall, ER appeared to be impaired in young people with ASD compared to typically developing (TD) children. This finding suggests that ER could be a fruitful target for future ASD interventions and strategies focussing on ER should be offered to families, carers and professionals supporting children with ASD. A potential trend between age and ER impairments also emerged, with older children appearing to have more ER difficulties than younger children. Professionals and families may find this useful to consider when planning for future placements or care provision. Finally, young people with ASD reported fewer ER difficulties than parents. Clinicians may want to consider the potential impact of this on assessment and could involve third parties to compensate for these discrepancies.

The review was the first to systematically compare informant-reported ER in young people with ASD to TD controls. It raised the need for both a more widely shared understanding of the nature of ER and a consensus about ER measurement. This development would aid comparison of research and support the investigation of

relationships between specific ER domains and resulting behavioural difficulties, which would provide information for more targeted interventions.

The empirical study was the first to investigate the relationship between parent-reported Executive Function (EF) and Pathological Demand Avoidance (PDA) behaviours in children with ASD. Overall, children with ASD and PDA traits appeared to have more EF deficits than non-ASD children, suggesting that difficulties with EF should be considered when offering support to children and families. More specifically, EF deficits predicted non-compliance behaviours, with differential involvement of EF indices. For example, difficulties with transitioning from pleasant to less pleasant tasks were predicted by difficulties with planning, organising, initiating and holding information in mind. Conversely, non-compliance in situations involving uncertainty was predicted by difficulties with inhibition, shift behaviours and emotional control. Therefore, when assessing non-compliance behaviour, it could be useful for clinicians to consider firstly what the demand is, and then which EF skills are needed.

This study raised awareness of some of the difficulties that children with ASD and PDA traits face, and the need for the development of more helpful support strategies. We have laid the ground work for further, more rigorous methods, such as cognitive assessments and multi-method designs, to both replicate and progress these findings. Particularly, methods designed to investigate causal relationships are needed, to better inform interventions.

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Part 1: Literature Review

**Emotion Regulation in Children and Young People with
Autism Spectrum Disorder: A Systematic Review**

1.1 Abstract

Background: It has been suggested that Emotion Regulation (ER) difficulties may contribute to some of the challenges faced by individuals with Autism Spectrum Disorders (ASD). However, the extent or nature of these difficulties still remains unclear. This review aims to evaluate ER in children and young people with ASD in comparison to typically developing (TD) children.

Method: PsycINFO, MEDLINE, PubMed and Google Scholar were searched using terms related to ER and ASD. The search yielded 524 papers, 256 of which were retrieved on the basis of specific inclusion criteria. Seventeen of these were selected for the final analysis. The review was limited to studies using self- or parent-report questionnaires as a method of assessing ER in children with ASD and included a TD comparison group. Standardised effect sizes were calculated to enable comparisons to be made across studies. The review examined the impact of age and informant-type on the extent of ER difficulties, and then investigated ER difficulties in relation to Gross and Thompson's modal model.

Results: Children and young people with ASD were found to have more ER difficulties than TD comparison groups. Age and informant-type appeared to be related to the size of the differences found. It was not possible to explore differences between ER domains, and these barriers were discussed.

Conclusions: Overall, ER appears to be a difficulty for children and young people with ASD and this was apparent across all domains. Further work is needed to explore interacting factors such as age and measurement, and to elucidate the ER construct.

1.2 Introduction

1.2.1 Autism Spectrum Disorder (ASD)

‘Autism Spectrum Disorder’ (ASD) is an umbrella term used to describe a group of pervasive developmental disorders characterised by impairments in social interaction and communication, in the presence of repetitive and stereotyped behaviour, interests and activities (World Health Organization., 1993). ASD is one of the most commonly diagnosed child neurodevelopmental disorders (American Psychiatric Association, 2013) and previously included four subgroups: childhood autism, atypical autism, Asperger’s syndrome and pervasive developmental disorders-not otherwise specified (PDD-NOS). Clinical accounts of Autism were first reported in the forties (Asperger, 1944; Kanner, 1943), but were not introduced into the Diagnostic Statistical Manual of Mental Disorder (DSM) until the DSM-III was published (American Psychiatric Association, 1980). Since this, much debate has ensued around the diagnostic criteria for Autism. However, the DSM-V brought in new revised diagnostic criteria, including a move from triadic to dyadic symptom grouping, and merging previous subcategories (mentioned above) into a single ASD diagnostic category. Between 2000 to 2009 prevalence estimates of ASD have increased from 30.8 per 10,000 (Baron-Cohen et al., 2000), to around 100 per 10,000 (Baron-Cohen et al., 2009), suggesting the need for continued research and development of interventions and services for individuals with ASD.

A diagnosis of ASD has been found to be a significant risk factor for emotional problems (Totsika, Hastings, Emerson, Lancaster, & Berridge, 2011) and in general high rates of emotional problems have been recorded in children with ASD (Salomone et al., 2014). Difficulties with emotional functioning are commonly associated with problems such as irritability, aggression, self-injury, anxiety, and impulsivity

(Lecavalier, 2006). From their cohort sample of children with ASD, Totsika et al. (2011) reported clinically significant levels of hyperactivity, conduct problems emotional problems. There are also large rates of co-morbid psychiatric conditions found in children with ASD. Leyfer et al. (2006) reported rates of about 72% meeting criteria for at least one DSM-IV Axis 1 condition. Later, Simonoff et al. (2008) supported this finding, estimating that 70% of children with ASD met the criteria for at least one comorbid condition, and a recent meta-analysis found that 39.6% of individuals with ASD have a comorbid anxiety disorder (van Steensel, Bögels, & Perrin, 2011). These emotional and behavioural problems can negatively impact not only the individual but also their family and carers, and consequently their quality of life.

Correlations of internalising and externalising symptoms have also been reported in the ASD population (Lecavalier, Gadow, DeVinent, & Edwards, 2009; Leyfer et al., 2006). For instance, it has been found that individuals with ASD who have higher levels of depression or anxiety also show raised levels of noncompliance, aggressive behaviour, and irritability (J. Kim, Szatmari, Bryson, Streiner, & Wilson, 2000; Mayes, Calhoun, Murray, Ahuja, & Smith, 2011). Due to the high rates of co-occurring depression, anxiety and anger problems, the need for a more transdiagnostic way of working with individuals with ASD has been raised (Weiss, 2014). Emotion Regulation (ER) often occurs as the core focus of other transdiagnostic models (McLaughlin, Hatzenbuehler, Mennin, & Nolen-Hoeksema, 2011) and ER difficulties have been proposed as a potential shared factor to explain the high rates of emotional and behavioural problems in children and young people with ASD (Mazefsky et al., 2013; Samson, Hardan, Podell, Phillips, & Gross, 2014; Samson, Wells, Phillips, Hardan, & Gross, 2015; Weiss, 2014).

1.2.2 Emotion Regulation (ER)

Emotion Regulation can be defined as the process of effectively managing emotions in response to environmental demands (Aldao, Nolen-Hoeksema, & Schweizer, 2010). Poor ER has been implicated in a range of emotional problems in children with ASD, such as anxiety, depression, and anger (Mazefsky et al., 2013). Interestingly, interventions targeting ER specifically have been shown to be effective in children and young people with ASD (Cai, Richdale, Dissanayake, & Uljarević, 2017). Thomson, Burnham Riosa and Weiss (2015) found that by targeting ER in children with ASD, parents reported significant improvements in child emotional lability, internalising symptoms, behavioural dysregulation and adaptive behaviour. Others have also found that by delivering modified Cognitive Behavioural Therapy (CBT) to children with ASD, with a specific focus on teaching ER strategies, parents report less negativity and lability, improved ER and shorter outbursts (Scarpa & Reyes, 2011). However, although there is growing acknowledgement of the potential role of ER in the co-morbid presenting difficulties of those with ASD and its fruitfulness as a target for intervention, there is still little known about the extent and nature of ER difficulties in children and young people with ASD (Mazefsky et al., 2013). Further understanding of ER as a construct and its impact in this population seems important in order to develop more targeted and successful interventions.

1.2.3 The modal model of ER

There have been many conceptualisations of ER, and it has been suggested that a clear consensus of the definition is needed in order to identify the boundaries of what is and is not understood to be an ER construct (Compas et al., 2017). However, a common way of understanding ER is as a process through which individuals regulate their emotions, either consciously (effortful and control) or unconsciously (effortless

and automatic) to appropriately respond to emotion-eliciting stimuli (Aldao et al., 2010). Gross and Thompson's (2007) modal model of ER is one of the most frequently employed frameworks used to guide understanding about ER for research and clinical purposes. This ER model includes responses that are directed towards other processes as well as emotions, such as efforts directed at the context in which emotions occur, the modulation of behavioural responses and the cognitive processes that may shape and influence emotions.

Weiss, Thomson and Chan (2014) conducted a systematic review investigating the types of measurements used to assess ER in children with ASD. They were the first to use Gross and Thompson's model to evaluate which ER domains different measures assess. The modal model of ER suggests five temporarily linked domains of ER processes:

- (1) Situation Selection: the ability to understand a given situation, predict the likely outcomes, and evaluate the consequences of entering into that situation adaptively (e.g. avoid potentially dangerous situations) or maladaptively (e.g. consistently avoid reasonably safe situations).
- (2) Situation Modification: modifying a situation in order to regulate emotional responses.
- (3) Attentional Deployment: controlling the allocation of attention to or away from the emotion eliciting features of a given situation.
- (4) Cognitive Change: regulating emotional reactions through appraisals of a situation and one's capacity to cope.
- (5) Response Modulation: physiological and behavioural ways of regulating and expressing emotions after they are experienced.

Gross and Thompson (2007) also describe various types of strategies, (situation, attention, appraisal and response) that may be employed at each stage of the regulatory process and how they correspond to the five ER domains (see Figure 1.1). They suggest that antecedent-focused processes, including situation selection and modification, attentional deployment and cognitive change, occur before appraisals give rise to emotional response (Webb, Miles, & Sheeran, 2012). This concept is supported by research that indicates that adult caregivers will often alter the situations of their children in order to modify their children's emotional experiences (Eisenberg, Cumberland, & Spinrad, 1998). Also, correlational studies suggest that people will alter or avoid situations as a way of coping (D'zurilla, Chang, & Sanna, 2003; Jaffee, & D'zurilla, 2003). Finally, experimental evidence also supports the use of attentional deployment or cognitive change in order to regulate emotions. For example, studies have found that asking participants to think of something else other than an impending pain or danger reduces anxiety (Kalisch, Wiech, Hermann, & Dolan, 2006; Gross, 1998).

The response-focussed processes are conceptualised as occurring after the emotional responses are generated. These involve strategies such as suppression of expressed emotion, which Gross (1998) provided evidence for when asking participants to behave so that others would not know what they were feeling. A recent meta-analysis investigated strategies derived from the modal model of ER (Webb, Miles, & Sheeran, 2012). They found evidence for conceptual distinctions between processes and support that these distinct ER processes were effective in regulating emotions. However, they did find that some, for example cognitive change and response modulation, had larger effects than others, such as attentional deployment.

Emotion Regulation

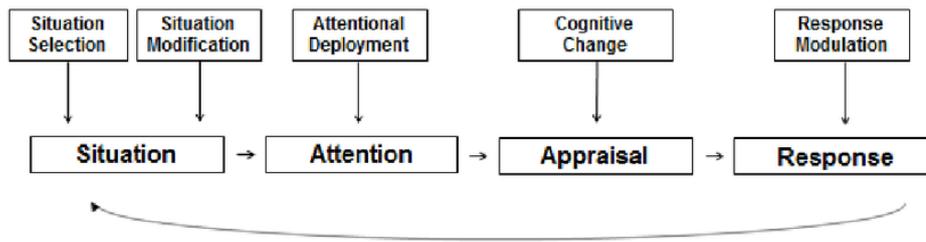


Figure 1.1 The process model of emotion regulation (Gross & Thompson, 2007).

Weiss et al. (2014) thoroughly reviewed the types of ER measurements for children with ASD and indicated which domains these measures specifically assess. However, a comprehensive review of the extent of ER difficulties in children with ASD in relation to TD controls, with a focus on understanding where these difficulties fall in relation to Gross and Thomson's 2007 modal model, has not been conducted.

1.2.4 Research aims

In light of growing interest in the role of ER in co-occurring emotional and behavioural difficulties shown by children and young people with ASD, the main aim of this systematic review is to assess whether children and young people with ASD are impaired in ER in comparison to TD children and young people. This review will be limited to studies that use either self- or parent-report measures to assess ER in children and young people with and without ASD. In the interest of brevity, the terms "children" and "young people" will be used interchangeably to refer to both "children and young people". The measures included in this review will assess a range of ER domains, specifically those described by Gross and Thompson (2007).

This review aims to address three main questions:

1. To what extent do young people with ASD suffer with difficulties in ER in comparison to TD young people?
2. To what extent do other factors, such as age and respondent type, impact reported ER difficulties?

3. By using Gross and Thompson's (2007) model of five ER domains, can any patterns, for example of weaknesses and strengths in ER abilities, be identified in children with ASD?

Thus, the present systematic review aims to synthesise the evidence found around ER difficulties in young people with ASD in comparison to TD young people. The review will build upon conceptual and psychometric work that has previously been developed in this area (Mazefsky et al., 2013; Weiss et al., 2014). The overarching goal is to contribute to current understanding of ER and to provide theoretical and clinical insight into a potentially fruitful target for intervention.

1.3 Method

1.3.1 Data sources and study inclusion

A systematic literature search was performed using three electronic databases (PsycINFO, MEDLINE and PubMed) and this was supplemented by searches in Google Scholar and the reference sections of published articles. Titles, Abstracts and Keywords were searched using the following search terms relating to ASD and ER: (1) "ASD" or "autis*" or "ASC" or "asperger*" or "PDD-NOS" or "pervasive developmental disorder" and (2) "emotion regulation" or "emotional regulation" or "emotional dysregulation" or "emotional control" or "emotion management" or "effortful control" or "maladaptive coping" or "adaptive coping" or "affect regulation." The titles and abstracts of identified articles were then screened for the following inclusion criteria: (1) The study was published between 1985 and September 2017, because before the mid-80s little work on what is now known as "emotion regulation" had been published (Aldao et al., 2010); (2) The study was published in a

peer-reviewed journal; (3) The study was written in English; (4) ER was investigated using a cross-sectional questionnaire methodology; (5) ER was measured using either a parent- or self-report measure that had been identified by Weiss et al. (2014) as measuring ER and using their method had been categorised as assessing one or several of Gross and Thompson's (2007) modal model domains; (6) Participants were children and young people (<20 years) who met diagnostic criteria according to either the Diagnostic and Statistical Manual of Mental Disorders 3rd edition revised (DSM-III-R; American Psychiatric Association, 1980), the DSM-IV (American Psychiatric Association, 1994), the DSM-V (American Psychiatric Association, 2013), or the International Statistical Classification of Diseases and Related Health problems 10th revision (ICD-10; World Health Organization, 1993); (7) A TD comparison group was available.

The initial literature search resulted in a total of 682 findings (307 from PsycINFO, 193 from PubMed, 181 from Medline, and one from Google Scholar). Excluding duplicates produced a total of 524 findings. From a surface scan a further 268 articles were automatically excluded as they did not meet basic inclusion criteria (e.g. date of publication, language of publication, peer-reviewed journal, population) resulting in a total of 256 article findings. Finally, 256 articles were reviewed in more depth and a further 239 articles were excluded due to: ER was not being investigated (131); ER was measured via observation or experimentation (34); the article was a review or narrative piece (27); no TD comparison group was included (20); participants were adults (>20 year; 11); participants were not children or young people with ASD (10); the article did not provide data necessary to calculate an effect size and authors failed to respond to emails requesting information (5); the article used an

unpublished scale to assess ER (1). This resulted in 17 articles that met the criteria and were included in the current review. This search is summarised in Figure 1.2.

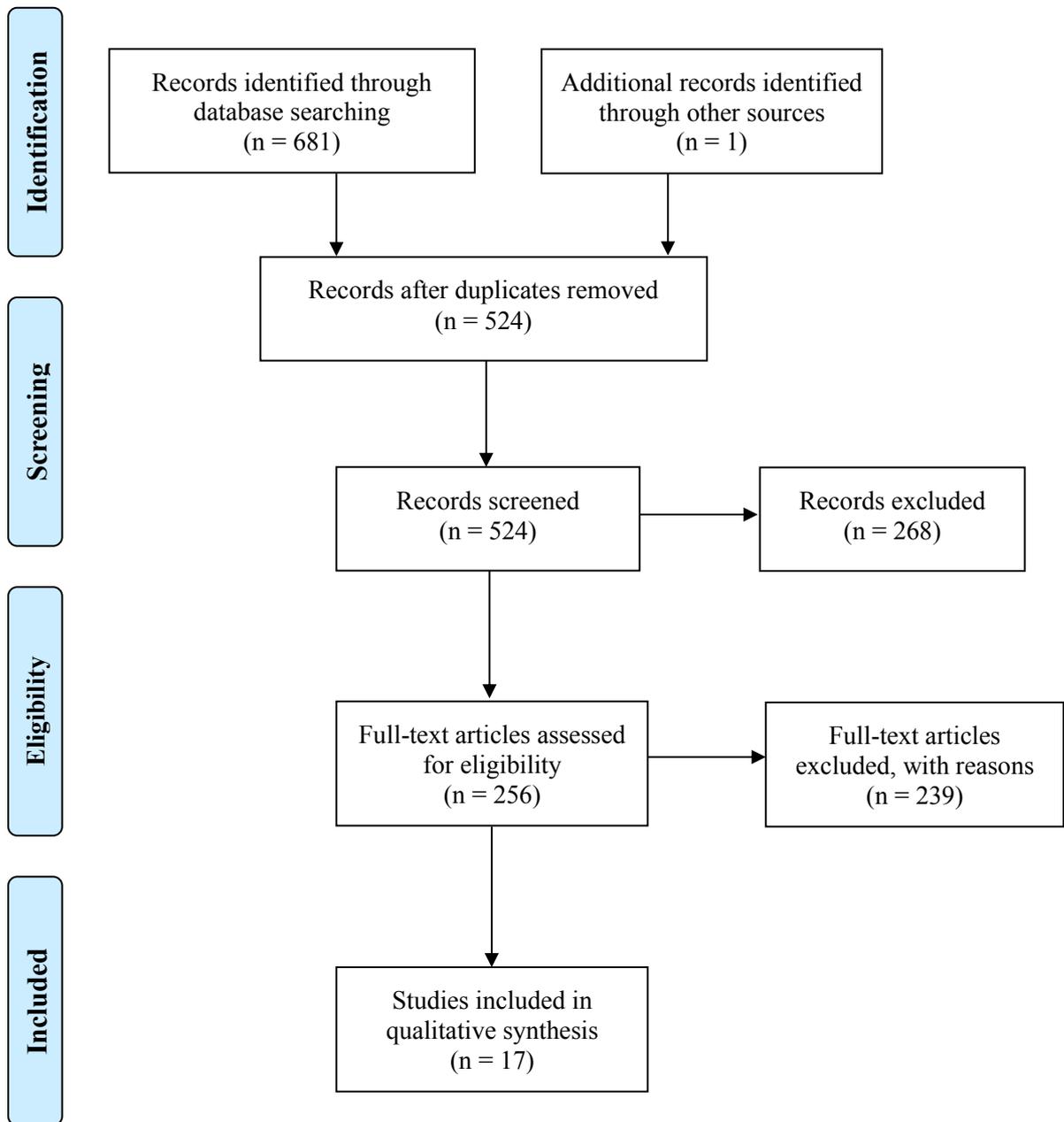


Figure 1.2 PRISMA flow diagram.

1.3.2 Emotion Regulation measures

Weiss et al. (2014) conducted a large systematic review of the types of methods used to measure ER in children with ASD. They used the modal model of ER (Gross & Thompson, 2007) to determine which domain each measure assessed. It was beyond

the scope of the current review to re-assess all questionnaire measures used with ASD populations. Therefore, in order to build on previous work and to further evaluate ER in young people with ASD, only those measures cited by Weiss et al. (2014), that had been rigorously established as mapping onto at least one of Gross and Thompson's ER domains, were included in this review. Only one study was identified that used a measure that was not cited by Weiss et al. (2014). This was the Emotion Regulation Index for Children and Adolescents (ERICA; MacDermott, Gullone, Allen, King, & Tonge, 2010). All data from this measure was not included in this review.

Most studies selected for this review used more than one measure to assess ER. To ensure that each study was neither over- nor under-represented we analysed data from one ER measure per study. An a priori algorithm was developed for selecting which measure to include from studies where more than one measure of ER was used. Firstly, measures were discarded if they did not provide data from the whole measure, including all subscales. Next, if there were still multiple measures remaining, the measure that tapped into the largest number of Gross and Thomson's (2007) ER domains was selected, based on the classification of Weiss et al. (2014). This second step reflects the fact that measures that tap into the full range of ER domains will have greater content validity than those which cover only part of the ER construct. This review separated out parent- and self-report studies, as it would not be reasonable to combine data from these measures. Therefore, if a study used both a parent- and self-report measure, they were permitted to be included twice, as these different types of data were treated separately in the review.

1.3.3 Effect sizes

Cohen's *d* (Cohen, 1977) was calculated as a measure of standardised effect size for ER assessed by a validated self- or parent-report questionnaire. Differences

between means for both the ASD and TD groups were calculated and then divided by the pooled standard deviation for both groups. This is summarised by the following equation (taken from Wykes, Huddy, Cellard, McGurk, & Czobor, 2011):

$$\text{Effect Size (d)} = (M_{\text{ASD}} - M_{\text{TD}}) / SD_{\text{pooled}}$$

Where M_{ASD} represents the mean for the ASD group, M_{TD} represents the mean for the TD group, and SD_{pooled} represents the pooled standard deviation for both the ASD and TD groups. Effect sizes were interpreted as: small = 0.2; medium = 0.5; large = 0.8 (Cohen, 1977). A negative effect size was always indicative of poorer ER ability in the ASD group, implying a bigger impairment in young people with ASD, and a positive effect size indicated that the ASD participants had fewer impairments. In some studies, a total ER mean was not available; instead subscale scores were presented. In these cases, an overall ER effect size was calculated by averaging across subscale effect sizes. Similar methods have been used in other literature reviews (Dong, Maynard, & Perez-Johnson, 2008; Wykes et al., 2011).

It should be noted that the initial intention of this review was to conduct a meta-analysis. However, it was decided that the diversity across ER measures was too great to justify this type of statistical comparison. For example, one measure may assess affect, whilst another may examine coping strategies, but each fall under the ER umbrella, this is often referred to as “mixing apples and oranges” (Borenstein, Hedges, Higgins, & Rothstein, 2009). Therefore, in order to avoid this problem a systematic review was conducted. Nevertheless, standardised effect sizes have been included to add to the rigour of this review and enable more quantifiable comparisons to be made.

1.3.4 Quality and relevance assessment

Many tools are available to assess the quality of intervention studies and randomised designs. However, few appear to specifically assess cross-sectional studies that are similar to those reviewed here. The Newcastle-Ottawa Scale (NOS; Wells et al., 2013) was developed for assessing the quality of non-randomised studies for the purposes of systematic reviews and meta-analyses. Therefore, the NOS was chosen as the most appropriate tool to adapt for this review. The NOS uses a star rating system whereby a study is judged on three broad perspectives: the selection of study groups; the comparability of the groups; and outcome measurement. This scale was adapted to assess the cross-sectional studies included in this review (see Appendix II). An overall rating system of quality for the current review was developed based on the NOS star ratings. Studies scoring five stars or more were rated as “High Quality”, studies scoring four stars were rated as “Medium Quality”, and studies scoring three stars or less were rated as “Low Quality”

1.4 Results

1.4.1 Corpus of studies

The literature search produced 17 studies that fulfilled all inclusion criteria. The studies included in the review are summarised in Table 1.1 and Table 1.2. From the 17 studies, ER was assessed using a combination of 20 different measures, and 15 of these were selected for inclusion in this review. Of the 17 studies, 12 reported data using self-report measures of ER and 12 reported data using parent-report measures of ER.

Table 1.1

Summary of self-report studies included in the review

Study	N (ASD, TD)	Age (mean years, range)	Male: Female	Diagnosis	IQ matching	Emotion Regulation Measure	Effect Size (<i>d</i>)	Construct Assessed (Weiss et al., 2014)	Gross and Thompson (2007) Emotion Regulation Domain (Weiss et al., 2014)				
									Situation Selection	Situation Modification	Attentional Deployment	Cognitive Change	Response Modulation
Costa et al. (2017)	78 (37, 41)	ASD = 9.07 TD = 8.42 Range = 3-13	ASD = 32:5 TD = 32:9	ASD	None reported	Emotion Regulation Questionnaire (ERQ; Gross & John, 2003)	-0.48	Self-report measure of frequency of reappraisal and suppression	-	-	-	Y	Y
Lordo et al. (2017)	29 (16, 13)	ASD = 15.07 TD = 15.57 Range = 12-17	ASD = 12:4 TD = 8:5	ASD	FSIQ>70	Positive and Negative Affect Schedule for Children (PANAS-C; Watson, Clark, & Tellegen, 1988)	-0.59	Self-report measure of positive and negative affect	-	-	Y	-	-
Mazefsky et al. (2014)	48 (25, 23)	ASD = 15.22 TD = 15.56 Range = 12-19	ASD = 24:1 TD = 22:1	ASD	WASI ≥80	Response to Stress Questionnaire (RSQ; Connor-Smith, Compas, Wadsworth, Harding Thomsen, & Saltzman, 2000)	-0.56	Self-report measure of voluntary/involuntary cognitive and behavioural emotion regulation processes	Y	Y	Y	Y	Y

Pouw et al. (2013a)	133 (67, 66)	ASD = 11.58 TD = 11.58 Range = 9-15	ASD = 59:8 TD = 57:9	ASD	IQ>80	Self-Report Instrument for Reactive and Proactive Aggression (IRPA; Rieffe et al., 2016)	-0.04	Self-report of aggressive behaviour	-	-	Y	-	Y
Pouw et al. (2013b)	120 (63, 57)	ASD = 11.7 TD = 11.5	All boys	ASD	IQ>80	Children's Coping Scale (Wright, Banerjee, Hoek, Rieffe, & Novin, 2010)	-0.13	Self-report of coping strategies	-	Y	Y	Y	Y
Rieffe et al. (2011)	184 (66, 118)	ASD = 11.42 TD = 11.42 Range = 10-13	ASD = 58:8 TD = 104:14	Autism	WISC-R>80	Emotion Awareness Questionnaire (EAQ; Rieffe et al., 2007)	-0.13	Self-report to measure key aspects of emotional awareness	-	-	Y	-	Y
Rieffe et al. (2012)	130 (64, 66)	ASD = 11.75 TD = 11.5 Range = 9-15	ASD = 57:7 TD = 66:8	High Functioning ASD	Matched	The Mood Questionnaire (Rieffe, Terwogt, & Bosch, 2004)	-0.38	Self-report measure of affective states for basic emotions in children	-	-	Y	-	-
Rieffe et al. (2014)	212 (81, 131)	ASD = 11.76 TD = 11.68 Range = 8-15	ASD = 72:9 TD = 59:72	High Functioning ASD	IQ Matched WISC	Children's Coping Scale (Wright et al., 2010)	-0.22	Self-report of coping strategies	-	Y	Y	Y	Y
Samson et al. (2015)	59 (31, 28)	ASD = 13.26 TD = 12.43 Range = 8-20	ASD = 27:4 TD = 21:7	ASD	FSIQ: 64-133	Emotion Regulation Questionnaire (ERQ; Gross & John, 2003)	-0.23	Self-report and parent-report measure of frequency of reappraisal and suppression	-	-	-	Y	Y
Samyn et al. (2011)	54 (27, 27)	ASD = 12.73 TD = 12.91	All boys	ASD	FSIQ≥80	The Effortful Control Scale (ECS;	-0.81	Self-report to assess behavioural and	Y	Y	Y	Y	Y

		Range = 10-15				Lonigan & Phillips, 2001)		attention components of ER					
Samyn et al. (2014)	41 (20, 21)	ASD = 12.61 TD = 13.58 Range = 10-15	ASD = 15:5 TD = 14:7	ASD	FSIQ \geq 80	The Effortful Control Scale (ECS; Lonigan & Phillips, 2001)	-1.05	Self-report to assess behavioural and attention components of ER	Y	Y	Y	Y	Y
Samyn et al. (2015)	179 (31, 148)	ASD = 12.83 TD = 12.73 Range = 10-15	All boys	ASD	FISQ \geq 80	The Effortful Control Scale (ECS; Lonigan & Phillips, 2001)	-0.60	Self-report to assess behavioural and attention components of ER	Y	Y	Y	Y	Y

Table 1.2

Summary of parent-report studies included in the review

Study	N (ASD, TD)	Age (mean years, range)	Male: Female	Diagnosis	IQ matching	Emotion Regulation Measure	Effect Size (<i>d</i>)	Construct Assessed (Weiss et al., 2014)	Gross and Thompson (2007) Emotion Regulation Domain (Weiss et al., 2014)				
									Situation Selection	Situation Modification	Situation Deployment	Attentional Change	Cognitive Modulation
Costa et al. (2017)	78 (37, 41)	ASD = 9.07 TD = 8.42 Range = 3-13	ASD = 32:5 TD = 32:9	ASD	None reported	Emotion Regulation Checklist (ERC; Shields & Cicchetti, 1997)	-1.07	Parent-report of perceptions of their child's typical methods of managing emotional experience	-	Y	Y	-	Y
Jahromi et al. (2013)	40 (20, 20)	ASD = 4.91 TD = 4.18 Range = unknown	Total = 36:4 (gender matched)	Autism	Matched on expressive language	Emotion Regulation Checklist (ERC; Shields & Cicchetti, 1997)	-0.87	Parent-report of perceptions of their child's typical methods of managing emotional experience	-	Y	Y	-	Y
Konstanta reas & Stewart, (2006)	42 (19, 23)	ASD = 6.16 TD = 6.37 Range = 3-10	ASD = 12:7 (gender matched)	PDD-NOS = 9 Autistic Disorder = 10	None reported	Children's Behaviour Questionnaire (CBQ; Rothbart, Ahadi, Hershey, & Fisher, 2001)	-0.87	Parent-report of temperament in children ages 3-7	Y	Y	Y	-	Y

Lordo et al. (2017)	29 (16, 13)	ASD = 15.07 TD = 15.57 Range = 12-17	ASD = 12:4 TD = 8:5	ASD	FSIQ>70	Positive and Negative Affect Schedule for Children (PANAS-C; Watson et al., 1988)	-1.45	Parent-report measure of positive and negative affect	-	-	Y	-	-
Rieffe et al. (2011)	184 (66, 118)	ASD = 11.42 TD = 11.42 Range = 10-13	ASD = 58:8 TD = 104:14	Autism	WISC-R>80	Strengths and Difficulties Questionnaire (SDQ; Goodman, 2001)	-1.05	Parent-report to measure adjustment and psychopathology of children and adolescents	Y	Y	Y	-	Y
Samson et al. (2015)	59 (31, 28)	ASD = 13.26 TD = 12.43 Range = 8-20	ASD = 27:4 TD = 21:7	ASD	FSIQ: 64-133	Emotion Regulation Questionnaire (ERQ; Gross & John, 2003)	-1.08	Self-report and parent-report measure of frequency of reappraisal and suppression	-	-	-	Y	Y
Samyn et al. (2011)	54 (27, 27)	ASD = 12.73 TD = 12.91 Range = 10-15	All boys	ASD	FSIQ≥80	The Early Adolescent Temperament Questionnaire - Revised (EATQ-R; Ellis & Rothbart, 2001)	-1.95	Parent-report to assess inhibitory control, attentional control and activation control	Y	-	Y	-	Y
Samyn et al. (2014)	41 (20, 21)	ASD = 12.61 TD = 13.58 Range = 10-15	ASD = 15:5 TD = 14:7	ASD	FSIQ≥80	The Early Adolescent Temperament Questionnaire - Revised (EATQ-R; Ellis & Rothbart, 2001)	-1.66	Parent-report to assess inhibitory control, attentional control and activation control	Y	-	Y	-	Y

Samyn et al. (2015)	179 (31, 148)	ASD = 12.83 TD = 12.73 Range = 10-15	All boys	ASD	FSIQ \geq 80	The Early Adolescent Temperament Questionnaire - Revised (EATQ-R; Ellis & Rothbart, 2001)	-1.49	Parent-report to assess inhibitory control, attentional control and activation control	Y	-	Y	-	Y
South et al. (2012)	59 (30, 29)	ASD = 14.31 TD = 14.21 Range = 11-16	ASD = 26:4 TD = 21:8	ASD	WASI \geq 75	Spence Children's Anxiety Scale-Parents (SCAS-P; Nauta et al., 2004)	-0.99	Parent-report of Anxiety	-	Y	Y	-	Y
Van Hecke et al. (2009)	33 (19, 14)	ASD = 9.95 TD = 9.93 Range = 8-12	ASD = 18:1 TD = 10:4	High Functioning ASD	IQ \geq 75	Social Skills Rating System - Elementary Parent Form (SSRS; Gresham & Elliot, 1990)	-1.81	Parent-report of social skills and problem behaviours	-	-	-	-	Y
Yager & Iarocci (2013)	183 (135, 48)	ASD = 14.17 TD = 14.12 Range = 11-18	Matched on age	High Functioning ASD	ABIQ \geq 80	Multidimensional Social Competence Scale (MSCS; Yager & Iarocci, 2013)	-2.91	Parent-report of social competence including emotion regulation	-	-	-	-	Y

1.4.2 Quality appraisal

Table 1.3 shows the results of the quality appraisal conducted on the 17 studies in this review. Overall, for the studies reporting self-report outcomes, three were rated as “Medium” quality and nine were rated as “High”. For the parent-report studies, six were found to be of “Medium” quality, and six were rated as “High”. No studies were found to be “Low” in quality or relevance. Table 1.3 shows that most studies lost stars either from the “Control” criterion, due to not reporting IQ scores for controls or not controlling stringently enough, or from the “Outcome” criterion, indicating that some studies used less applicable or valid measures than others.

1.4.3 Self-reported ER difficulties

Table 1.1 shows the effect sizes found for self-report measures of ER. An examination of the effect sizes, represented by Cohen’s d , showed that out of the twelve self-report studies, 50% ($n = 6$) found a small effect size, 33% ($n = 4$) found a medium effect size, and 17% ($n = 2$) found a large effect size. Overall, out of the 12 self-report assessments of ER selected for this study, 100% ($n = 12$) were negative, suggesting that the ASD group reported more difficulties with ER than the TD group.

The largest effect size ($d = -1.05$) was found by Samyn et al. (2015), a study assessed as “High” on the quality tool, using The Effortful Control Scale (ECS; Lonigan & Phillips, 2001) which assesses behavioural and attention components of ER. The smallest effect size ($d = -.04$) was found by Pouw et al. (2013a), a study also assessed as “High” on the quality appraisal tool, who used the Instrument for Reactive and proactive Aggression (IRPA; Rieffe et al., 2016) which assesses self-reported aggressive behaviour.

Table 1.3*Table showing quality and relevance ratings of studies included in the review*

Study		Selection	Control	Outcome	Overall Rating
<i>Self-report studies</i>					
Costa et al.	2017	***	-	*	Medium
Lordo et al.	2017	**	*	*	Medium
Mazefsky et al.	2017	**	*	**	High
Pouw et al.	2013a	***	*	*	High
Pouw et al.	2013b	***	*	**	High
Rieffe et al.	2011	***	*	*	High
Rieffe et al.	2012	***	*	*	High
Rieffe et al.	2014	***	-	**	High
Samson et al.	2015	***	-	*	Medium
Samyn et al.	2011	**	*	**	High
Samyn et al.	2014	**	*	**	High
Samyn et al.	2015	***	*	**	High
<i>Parent-report studies</i>					
Costa et al.	2017	***	-	*	Medium
Jahromi et al.	2013	**	-	**	Medium
Konstantareas & Stewart	2006	**	-	**	Medium
Lordo et al.	2017	**	*	*	Medium
Rieffe et al.	2011	***	*	*	High
Samson et al.	2015	***	-	*	Medium
Samyn et al.	2011	**	*	**	High
Samyn et al.	2014	**	*	**	High
Samyn et al.	2015	***	*	**	High
South et al.	2012	***	*	**	High
Van Hecke et al.	2009	**	*	*	Medium
Yager & Iarocci	2013	***	*	*	High

1.4.4 Parent-reported ER difficulties

Table 1.2 shows the effect sizes found for parent-report measures of ER. An examination of the effect sizes, represented by Cohen's d , showed that out of the 12 parent-report studies, 100% ($n = 12$) found a large negative effect size. It was also found that 75% ($n = 9$) of these effect sizes were greater than one. This consistent finding suggests that parents of the ASD groups reported more difficulties with ER than parents of the TD groups.

Yager and Iarocci (2013) found the largest effect size ($d = -2.91$). This study was rated as "High" by the quality appraisal method, and they used the Multidimensional Social Competence Scale (MSCS) which assesses social competence and ER. The smallest effect size ($d = -.87$) was reported from Jahromi et al. (2013), a study assessed as "Medium". They used the parent-report Emotion Regulation Checklist (ERC; Shields & Cicchetti, 1997) which assesses parent's perceptions of their child's typical emotional experience.

1.4.5 The impact of parent- versus self-report

It was found that both self-report and parent-report assessments of ER all resulted in negative effect sizes, suggesting a consistent finding of more ER difficulties in young people with ASD compared to TD young people. However, the main difference found between the self-report assessments of ER and the parent-report assessments were the proportion of large effect sizes. Young people reported much smaller differences between them and the TD control groups, with a larger proportion of the effect sizes being small. Whereas parents consistently reported differences resulting in large effect sizes, which shows a discrepancy between the way parents report their child's ER abilities versus the way the child or young person report their ER abilities.

1.4.6 Age and ER difficulties

The overall age range of participants included in this review was 3-20 years old. Table 1.4 shows the effect sizes, categorised as large, medium and small, across three age categories, for self-report. For the self-report studies, 50% ($n = 6$) assessed ER in young people aged 6-11 years old. Within this age range, the majority of effect sizes (83%) were found to be small, and no effect sizes were found to be large. However, the spread of effect sizes found for the self-report assessments made by young people aged 12-17 years differed from this, with 83% ($n = 5$) of effect sizes in this group being medium or above.

Table 1.4

Frequencies of effect sizes found in each mean age category, for self-report and parent-report studies

Effect Sizes	Mean Age Categories		
	0-5 years	6-11 years	12-17 years
<i>Self-report studies</i>			
Small	0	5	1
Medium	0	1	3
Large	0	0	2
Total	0	6	6
<i>Parent-report studies</i>			
d = .8 – 1	1	3	2
d = 1.1-1.5	0	0	2
d = >1.5	0	1	3
Total	1	4	7

All effect sizes were found to be large for parent-report studies. Therefore, separate Cohen's d categories were created for parent-report studies to assess the distribution of the large effect sizes (see Table 1.4). When looking at the spread of results from parent-reports, 58% ($n = 7$) of these studies assessed ER in young people

aged 12-17. This age category also reported the highest frequency of largest effect sizes ($d > 1.5$), with 75% ($n = 3$) of the largest effect sizes from all parent-report studies falling in this age category. Parent-report studies were the only studies that assessed children of less than 6 years old. Jahromi et al. (2013) used the ERC (Shields & Cicchetti, 1997) to assess children with ASD and TD children with a mean age of 4.5 years. The ERC assesses parent's perceptions of their child's typical methods of managing emotional experiences. Although the effect size fell within the lower end of the categories, it was still large ($d = .87$), suggesting that parents of young children with ASD reported more difficulties on this scale than parents of TD young children. Overall, 33% ($n = 4$) of the parent-report studies assessed children and young people who fell within the age band of 6-11 years. Table 1.4 shows that 75% ($n = 3$) of the effect sizes calculated from these studies fell within the $d = .8-1$ effect size category.

1.4.7 The ER domains

Table 1.1 shows for each self-report study which ER domains were assessed by the measure used, based on the classification used by Weiss et al. (2014). No particular relationship was found between the number or combination of domains included in a measure and the resulting effect size. However, only when self-report measures assessed all five of Gross and Thompson's (2007) ER domains were large effect sizes found. Although, it should be noted that this was only found in studies reaching a "Medium" rating on the quality appraisal tool.

Table 1.2 reports the same information but for studies using parent-report measures. Similarly, no particular relationship was found between the number or combination of domains measured and the resulting effect size reported. For parent-report studies, the largest effect sizes were found using measures assessing either a single ER domain, or a combination of three of the domains.

This review found little consistent differentiation between the measures used in terms of which domains they assessed. When comparing results across self-report studies, the same combinations of ER domains were only ever assessed at most by three studies. This was also found for parent-report studies. Therefore, due to the lack of consistency, great variability in measures, and the complexity of the ER construct, further analysis of the nature of ER difficulties in children with ASD was not able to be conducted. The frequency of domains assessed and findings from measures that assessed only one domain have been summarised below.

1.4.7.1 Situation Selection

Situation selection was the least assessed self-report domain, with only 33% ($n = 4$) of studies using a measure that encompassed this domain. No self-report measures selected for this review assessed Situation Selection alone, and interestingly it was the only ER domain that was solely assessed using measures that tapped into all five ER domains.

When reviewing parent-report studies, Situation Selection was assessed by 42% ($n = 5$) of the measures selected for this review. Similarly to self-report, this domain was not assessed alone by any of the parent-report measures included in this study.

1.4.7.2 Situation Modification

Situation Modification was assessed by 50% ($n = 6$) of the self-report studies. There were no self-report measures that assessed Situation Modification alone in this review, and it was only assessed by measures where at least three other domains of ER were being tapped into.

This review found that Situation Modification was assessed by 42% ($n = 5$) of the parent-report studies. None of the parent-report measures in this review assessed Situation Modification on its own.

1.4.7.3 Attentional Deployment

Attentional Deployment was one of the most commonly self-report assessed ER domains, with 83% ($n = 10$) of studies using a measure that tapped into this domain. Attentional Deployment was the only ER domain assessed on its own by self-report measures. Lordo et al. (2017) used the Positive and Negative Affect Schedule for Children (PANAS-C; Watson et al., 1988) which is a self-report measure of positive and negative affect, and was described as assessing Attentional Deployment by Weiss et al. (2014). Using the PANAS-C they found a medium effect size ($d = -.59$). The other study to do this was Rieffe et al. (2012) using The Mood Questionnaire (Rieffe et al., 2004); a self-report measure of affect states for basic emotions. They found a smaller effect size ($d = -.38$) to Lordo et al. (2017), but it must be recognised that Rieffe et al. (2012) was rated as a “High” quality study, whereas Lordo et al. (2017) received a “Medium” rating.

Attentional Deployment was the second most assessed ER domain for parent-report studies, with 75% ($n = 9$) of studies using a measure that tapped into this domain. Attentional Deployment was assessed alone by Lordo et al. (2017), a study assessed as “Medium” on the quality appraisal tool. Using the PANAS-C parent-report measure they found a large effect size ($d = -1.45$), which is substantially larger than the effect size recorded from the self-report data on this measure as previously mentioned above.

1.4.7.4 Cognitive Change

It was found that Cognitive Change was assessed by 67% ($n = 8$) of the self-report studies included in this review. No self-report measure in this review assessed Cognitive Change alone.

For parent-report studies, Cognitive Change was the least assessed domain, with only one study (8%) assessing this domain (Samson et al, 2015). This was done using the Emotion Regulation Questionnaire (ERQ; Gross & John, 2003) which also assesses Response Modulation.

1.4.7.5 Response Modulation

Alongside Attentional Deployment, Response Modulation was assessed by 83% ($n = 10$) of the self-report studies included in this review. This domain was not assessed on its own by any self-report measure in this review.

Response Modulation was also the most assessed ER domain for parent-report studies, with 92% ($n = 11$) of studies using a measure that tapped into this construct. This ER domain was assessed on its own by two parent-report measures. Van Hecke et al. (2009) used the Social Skills Rating System (SSRS; Gresham & Elliot, 1990) which is a parent-report tool that assesses a child's social skills and problem behaviours. Their study was rated as "Medium" by the quality assessment tool and found an effect size of $d = -1.81$, which was the third largest found in this review. Yager and Iarocci (2013) developed the Multidimensional Social Competence Scale (MSCS) which is a parent-report measure that primarily assesses social competence. The largest negative effect size ($d = -2.91$) in this review came from this study, and the study was rated as "High" by the quality assessment tool.

1.5 Discussion

Central to this systematic review was the aim to describe the extent and nature of ER difficulties in children and young people with ASD and compare this to TD children and young people. There are a wide range of methods and techniques that can be used to assess ER in individuals with ASD (Weiss et al., 2014), so in order to reduce heterogeneity between studies, this review focused on ER assessed via self- and parent-report questionnaire method. This review also aimed to explore ER in young people with and without ASD in relation to Gross and Thompson's (2007) modal model, and only measures that had been assessed and categorised using the modal model framework by Weiss et al. (2014) were included in this study. However, this did not appear to impact the range of studies included as only one study was found (Lordo et al., 2017) that included a measure not categorised by Weiss et al. (2014). This was the Emotion Regulation Index for Children and Adolescents (ERICA; MacDermott et al., 2010) and was excluded from all analysis.

Overall, this review established that young people with ASD show global impairments in ER, as assessed via self- and parent-report, in comparison to TD young people, with large effects found. This finding is supported by the observation that all effect sizes calculated, for both self- and parent-report studies, were in the direction of the ASD groups having greater ER difficulties than the TD groups. For the parent-report studies, all effects sizes were large, and for the self-report studies half were medium or above. This confirms findings from previous non-systematic reviews (Mazefsky et al., 2013; Mazefsky & White, 2014).

1.5.1 Informant type

This review highlighted factors that may influence the reporting of ER. More specifically it revealed that overall there were differences in the effect sizes found

when comparing self-report to parent-report, with parent-report studies finding consistently larger effects. This review also provided further opportunities to explore these differences by comparing results from the same measures in the same samples. Lordo et al. (2017) used the PANAS-C in both the parent- and self-report forms, and Samson et al. (2015) used the ERQ in both parent- and self-report forms. For both of these studies, considerably larger effect sizes were found in the parent-reported data compared to the child-reported. It should however be noted that both of these studies were rated as “Medium” on the critical appraisal tool. These findings raise questions about the validity of parent-report and/or self-report methods of investigating ER.

Evidence suggests an established tendency for children with ASD to report fewer or less severe emotional problems, autistic traits and empathy deficits than their parents (Johnson, Filliter, & Murphy, 2015; Mandy et al., 2016). Mandy et al. (2016) found that children with ASD tended to self-report lower levels of psychopathology than their parents, to the extent where the proportion of children self-reporting scores in the clinical range was no higher than the general population. However, studies have also reported evidence suggesting there can be good inter-rater reliability between children with ASD and their parents when assessing constructs such as anxiety and depression (Kaat & Lecavalier, 2015; Ozsivadjian, Hibberd, & Hollocks, 2014; Stratis & Lecavalier, 2015). Although, certain factors, such as IQ, ASD symptom severity, or social cognitive skills, may impact reliability (Kaat & Lecavalier, 2015).

Johnson et al. (2015) explored the discrepancies between self- and parent-reporting in ASD and suggested that there could be two issues at play. Firstly, they proposed that there could be a lack of insight for the individual with autism, and secondly, they acknowledged that years of experience of learning about and living with a child with ASD is likely to lead to parents being more knowledgeable, observant

and sensitive to autism-related traits. Therefore, they suggested that an appraisal of self-awareness could be conducted before considering self-report tools, parental experiences should be considered, and obtaining ratings from a third source, such as a teacher or peer, could be used to gain a more accurate measurement of difficulties.

This review found that overall, more self-report studies were rated as “High” in quality than parent-report studies. The main reason for this was a lack of rigorous controlling for IQ in the parent-report studies. The poorer IQ controlling found across the parent-report studies may have contributed to the inflated effect sizes and could be one explanation for the large differences found between the self- and parent-report studies overall. However, in general, these findings support the observation that children with ASD tend to report fewer difficulties than their parents (Johnson et al., 2015; Mandy et al., 2016). This supports the need for more research to understand these differences and the use of methodological strategies, such as those described above (Johnson et al., 2015), in order to reconcile these differences. This may be an important avenue for future research in order to better and more reliably understand and assess ER.

1.5.2 Participant age

As well as informant type, the impact of age was also explored. Across both self- and parent-report studies the overall trend suggested that more studies focused on assessing ER in those aged between 12-17 years old. Although, it should be noted that a meta-analysis was not performed, and therefore this trend was not tested formally. However, our evidence suggests that this could be a good topic for future meta-analyses to consider.

Overall, there was a distinct lack of research using questionnaire methods for children below the age of six years old across both parent- and self-report studies. It

was beyond the scope of this review to include studies that use other methodologies to assess ER, such as observation or experimentation. Weiss et al. (2014) identified 20 studies that used naturalistic observation/behaviour coding methodologies to assess ER in young people with ASD. The majority of these studies used samples with age ranges of 2-7 years old, which could explain the lack of younger age samples in the current review. Developmental theory suggests that an essential component of any child's successful development is learning how to regulate emotional responses and related behaviours in socially appropriate and adaptive ways (Denham et al., 2003; Eisenberg, Spinrad, & Morris, 2002; Halberstadt, Denham, & Dunsmore, 2001). This seems particularly important for those with ASD who may naturally struggle more with these skills. Therefore, it appears that future research aimed at understanding the longitudinal development of ER could be needed which would be aided by more appropriate tools which capture ER difficulties in younger children.

Perhaps one of the most useful findings from this review was that smaller effect sizes tended to be found in younger children, and larger effect sizes in those who are older. However, it should be recognised that this finding was not based on any formal analyses or significance testing. The reason for the contrast in the size of ER differences across different age groups is unknown and there appears to be little research to help us understand this. Previous research has also highlighted this lack of understanding, and the potential gains that could come from more clarity around how ER skills develop and change across lifespan in the ASD population (Mazefsky & White, 2014). This finding also raises questions around the developmental nature of ER abilities and whether ER difficulties continue to develop as children age, or whether the contextual and developmental tasks that occur during adolescence mean that ER difficulties become more prominent and challenging. For instance, perhaps

more and more is expected from young people with ASD as they get older and this could particularly be reflected in parent- and teacher-report trends.

1.5.3 Emotion Regulation domains

This review used the framework developed by Weiss et al. (2014) to categorise which ER domains were assessed by which measure of ER. The aim of this review was to build on this framework by developing a better understanding of the extent of ER difficulties in children with ASD and by comparing results between and across ER domains. However, little consistency in measurement was found which meant that making direct comparisons of single domains or combinations of domains became impossible. This was hindered by the diversity of measures used, with a total of 15 measures being included in the review, across both self- and parent-report studies, and by the different combinations of ER domains assessed by different measures. This meant that the impact of any one domain or specific combinations of domains could not be isolated and assessed.

In general, there were few consistent relationships found between the number of ER domains assessed and the resulting effect sizes. However, for self-report studies, large effect sizes were only found when all five domains were assessed by a measure of ER, but this was not the case for parent-report studies. The complexities and difficulties of assessing ER in children have been highlighted before (Cole, Martin, & Dennis, 2004). The construct is broad and how best to define and assess it is still an issue of contention (Compas et al., 2017). However, these findings could suggest that when trying to assess ER difficulties directly from children or young people, a broader measurement of ER which taps into the whole spectrum and also includes subscales for different domains of ER, could produce a more useful measurement for investigating the extent and type of ER difficulties in ASD.

1.5.4 Limitations

The limitations of this review should be kept in mind when considering the implications of the findings. Firstly, the decision not to conduct a meta-analysis meant that no formal statistical testing could be performed on the data. Consequently, this review lacks any formal testing of variables that could be associated with the effect sizes reported. The decision not to conduct a meta-analysis was mainly based on the variability among measures of ER, as well as a lack of consistently used measures. The range of measures was considered too broad to be able to compare accurately and as previously explained, we wanted to avoid the pitfalls of “mixing apples and oranges” (Borenstein et al., 2009). This highlights the breadth of ER as a construct and the potential need for more consistency in measurement and definition.

Following this, the second limitation related to the inclusion of multiple different measures in this review. This was necessary in order to capture the breadth of the construct. However, this ultimately hinders the process of directly comparing across findings, as each measure has a slightly different emphasis. Next, this review was also limited to participants under the age of 20, and as a result included participants aged 3-20 years old. This limits the generalisability of the findings to those who fall within this age range. Finally, the impact of the size of this age range on the results must also be considered. The range includes many developmental milestones and changes, and it cannot be known what impact these factors have had on result.

1.5.5 Implications

1.5.5.1 Clinical implications

There are several clinical implications that can be drawn from the findings of this review. Firstly, it was found that ER, as assessed by parent- and self-report, appeared to be impaired in children and young people with ASD compared to TD

control groups. This suggested that ER could potentially be a fruitful target for clinical intervention and strategies focused around supporting difficulties related to ER impairments may be useful for implementation at home and in educational environments. Secondly, the results of this review suggest that impairments in ER may become more pronounced as children get older. Whether this is related to an actual increase in ER deficit or contextual changes making the original difficulties more pronounced we do not know. However, it may be useful for clinicians and carers to be aware of this potential increased impairment over time, in order to use more strategies as children age or to consider environmental challenges that could be adapted to reduce the impact of ER difficulties. It could also be useful to intervene with ER impairments earlier, in order to prevent them from having more of an impact later on. Thirdly, the differences found between parent- and self-report raise questions around which offers the most valid assessment of ER difficulties. This should be considered when assessing difficulties in young people with ASD. It highlights the importance of clinicians not relying too heavily on one source of information and gaining a third-party perspective in order to help identify issues and difficulties.

1.5.5.2 Scientific implications

The results of this review have several implications for future research and development in this field. Firstly, the need for a better definition and more broadly accepted understanding of what ER is and is not, is needed. This would aid assessment development and comparison between studies, to allow for a richer base of evidence to be built. Additionally, this review highlights the need for a broader measure of ER, which encompasses the wide spectrum of ER processes but also allows for more specific ER domain analysis. If a consistent method of questionnaire measurement could be developed, it would help comparisons across studies and also aid further

investigations into the relationships between ER processes and behavioural challenges in ASD. Related to measurement is the finding that young people reported fewer challenges than parents. This should be considered in future research using questionnaire methodology to investigate ER impairments. Findings may differ depending on informant-type and this could have consequences for interpretations. Future research should focus on trying to explain the differences found between parents and children in their ratings of ER difficulties. Finally, the difference found between age group reports, particularly from self-report data, suggests that more research is needed to understand the developmental nature of ER and related difficulties.

1.5.6 Conclusion

In conclusion, this review found evidence for impairments of ER in children and young people with ASD compared to TD control groups. Differences were found across age and informant-type, with older participants appearing to have more ER difficulties, and parents reporting greater ER difficulties. This review also highlighted the complexity of ER as a construct and the challenges of measuring and evaluating it. Overall, this review contributes to our understanding of ER difficulties in children and young people with ASD and provides evidence of impairment as assessed via questionnaire methodology. However, it is not possible to draw definitive conclusions about the extent of ER difficulties or the type of difficulties in relation to ER domains (Gross, & Thompson, 2007). This review highlights the need for future work to investigate ER difficulties in this group. In particular, areas such as the impact of age, the development of ER and ER difficulties, and the inconsistencies between self- and parent-report on ER outcomes needs further exploration. However, perhaps more pressing, the inconsistencies in measurement and the conceptual boundaries of ER

need further elucidation. Research has already indicated that interventions targeting ER in individuals with ASD can be successful (Cai et al., 2017; Scarpa & Reyes, 2011; Thomson et al., 2015). Therefore, there is a great need for more useful measurement and better understanding of the processes involved in this construct in order to produce more targeted interventions and more accurate methods of outcome measurement.

1.6 References

- Aldao, A., Nolen-Hoeksema, S., & Schweizer, S. (2010). Emotion-regulation strategies across psychopathology: A meta-analytic review. *Clinical Psychology Review, 30*(2), 217–237.
- American Psychiatric Association. (1980). *Diagnostic and statistical manual of mental disorders* (3rd ed.). Arlington, VA: Author.
- American Psychiatric Association. (1994). *Diagnostic and statistical manual of mental disorders* (4th ed.). Washington, DC: Author.
- American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders* (5th ed.). Washington, DC: Author.
- Asperger, H. (1944). Die "autistischen psychopathen" im kindesalter. *Archiv Für Psychiatrie Und Nervenkrankheiten, 117*(1), 76–136.
- Baron-Cohen, S., Scott, F. J., Allison, C., Williams, J., Bolton, P., Matthews, F. E., & Brayne, C. (2009). Prevalence of autism-spectrum conditions: UK school-based population study. *The British Journal of Psychiatry: The Journal of Mental Science, 194*(6), 500–509.
- Baron-Cohen, S., Wheelwright, S., Cox, A., Baird, G., Charman, T., Swettenham, J., ... Doehring, P. (2000). Early identification of autism by the checklist for autism in toddlers (CHAT). *Journal of the Royal Society of Medicine, 93*(10), 521–525.
- Borenstein, M., Hedges, L. V., Higgins, J. P. T., & Rothstein, H. R. (2009). Introduction to meta-analysis. *Clinical Rheumatology, 20*(Supplement 1), 1.
- Cai, R. Y., Richdale, A. L., Dissanayake, C., & Uljarević, M. (2017). Brief report: Inter-relationship between emotion regulation, intolerance of uncertainty, anxiety, and depression in youth with autism spectrum disorder. *Journal of Autism and Developmental Disorders, 1*–10.

- Cohen, J. (1977). Statistical power analysis for the behavioral sciences. *Statistical Power Analysis for the Behavioral Sciences*.
- Cole, P. M., Martin, S. E., & Dennis, T. A. (2004). Emotion regulation as a scientific construct: Methodological challenges and directions for child development research. *Child Development, 75*(2), 317–333.
- Compas, B. E., Jaser, S. S., Bettis, A. H., Watson, K. H., Gruhn, M. A., Dunbar, J. P., ... Thigpen, J. C. (2017). Coping, emotion regulation, and psychopathology in childhood and adolescence: A meta-analysis and narrative review. *Psychological Bulletin, 143*(9), 939–991.
- Connor-Smith, J. K., Compas, B. E., Wadsworth, M. E., Harding Thomsen, A., & Saltzman, H. (2000). Responses to stress in adolescence: Measurement of coping and involuntary stress responses. *Journal of Consulting and Clinical Psychology, 68*(6), 976–992.
- ¹*Costa, A., Steffgen, G., & Ferring, D. (2017). Contributors to well-being and stress in parents of children with autism spectrum disorder. *Research in Autism Spectrum Disorders, 37*, 61–72.
- Denham, S. A., Blair, K. A., DeMulder, E., Levitas, J., Sawyer, K., Auerbach-Major, S., & Queenan, P. (2003). Preschool emotional competence: Pathway to social competence? *Child Development, 74*(1), 238–256.
- Dong, N., Maynard, R. A., & Perez-Johnson, I. (2008). Averaging effect sizes within and across studies of interventions aimed at improving child outcomes. *Child Development Perspectives, 2*(3), 187–197.

¹ References marked with an asterisk (*) were included in the systematic review.

- D'zurilla, T. J., Chang, E. C., & Sanna, L. J. (2003). Self-esteem and social problem solving as predictors of aggression in college students. *Journal of Social and Clinical Psychology, 22*(4), 424-440.
- Eisenberg, N., Cumberland, A., & Spinrad, T. L. (1998). Parental socialization of emotion. *Psychological inquiry, 9*(4), 241-273.
- Eisenberg, N., Spinrad, T. L., & Morris, A. S. (2002). Regulation, resiliency, and quality of social functioning. *Self and Identity, 1*(2), 121–128.
- Ellis, L., & Rothbart, M. (2001). Revision of the early adolescent temperament questionnaire. *Society for Research in Child Development, 81*, 1–5.
- Goodman, R. (2001). Psychometric properties of the strengths and difficulties questionnaire. *Journal of the American Academy of Child and Adolescent Psychiatry, 40*(11), 1337–1345.
- Gresham, F., & Elliot, S. (1990). *The social skills rating system*. Bloomington, MN: Pearson Assessments.
- Gross, J. J. (1998). Antecedent-and response-focused emotion regulation: Divergent consequences for experience, expression, and physiology. *Journal of personality and social psychology, 74*(1), 224.
- Gross, J. J., & Thompson, R. A. (2007). Emotion regulation: Conceptual foundations. In J. J. Gross (Ed.), *Handbook of emotion regulation* (pp. 3–24). New York: Guildford Press.
- Gross, J. J., & John, O. P. (2003). Individual differences in two emotion regulation processes: Implications for affect, relationships, and well-being. *Journal of Personality and Social Psychology, 85*(2), 348–362.
- Halberstadt, A. G., Denham, S. A., & Dunsmore, J. C. (2001). Affective social competence. *Social Development, 10*(1), 79–119.

- Jaffee, W. B., & D'Zurilla, T. J. (2003). Adolescent problem solving, parent problem solving, and externalizing behavior in adolescents. *Behavior Therapy, 34*(3), 295-311.
- *Jahromi, L. B., Bryce, C. I., & Swanson, J. (2013). The importance of self-regulation for the school and peer engagement of children with high-functioning autism. *Research in Autism Spectrum Disorders, 7*(2), 235–246.
- Johnson, S. A., Filliter, J. H., & Murphy, R. R. (2015). Discrepancies between self- and parent-perceptions of autistic traits and empathy in high functioning children and adolescents on the autism spectrum.
- Kaat, A. J., & Lecavalier, L. (2015). Reliability and validity of parent- and child-rated anxiety measures in autism spectrum disorder. *Journal of Autism and Developmental Disorders, 45*(10), 3219–3231.
- Kalisch, R., Wiech, K., Herrmann, K., & Dolan, R. J. (2006). Neural correlates of self-distraction from anxiety and a process model of cognitive emotion regulation. *Journal of cognitive neuroscience, 18*(8), 1266-1276.
- Kanner, L. (1943). Autistic disturbances of affective contact. *Nervous Child, 2*(3), 217–250.
- Kim, J., Szatmari, P., Bryson, S., Streiner, D., & Wilson, F. (2000). The prevalence of anxiety and mood problems among children with autism and Asperger syndrome. *Autism, 4*(2), 117–132.
- *Konstantareas, M. M., & Stewart, K. (2006). Affect regulation and temperament in children with autism spectrum disorder. *Journal of Autism and Developmental Disorders, 36*(2), 143–154.
- Lecavalier, L. (2006). Behavioral and emotional problems in young people with pervasive developmental disorders: Relative prevalence, effects of subject

- characteristics, and empirical classification. *Journal of Autism and Developmental Disorders*, 36(8), 1101–1114.
- Lecavalier, L., Gadow, K. D., DeVincent, C. J., & Edwards, M. C. (2009). Validation of DSM-IV model of psychiatric syndromes in children with autism spectrum disorders. *Journal of Autism and Developmental Disorders*, 39(2), 278–289.
- Leyfer, O. T., Folstein, S. E., Bacalman, S., Davis, N. O., Dinh, E., Morgan, J., ... Lainhart, J. E. (2006). Comorbid psychiatric disorders in children with autism: Interview development and rates of disorders. *Journal of Autism and Developmental Disorders*, 36(7), 849–861.
- Lonigan, C. J., & Phillips, B. M. (2001). Temperamental influences on the development of anxiety disorders. *The Developmental Psychopathology of Anxiety*.
- *Lordo, D., Bertolin, M., Sudikoff, E., Keith, C., Braddock, B., & Kaufman, D. A. S. (2017). Parents perceive improvements in socio-emotional functioning in adolescents with ASD following social skills treatment. *Journal of Autism and Developmental Disorders*, 47(1), 203–214.
- MacDermott, S. T., Gullone, E., Allen, J. S., King, N. J., & Tonge, B. (2010). The emotion regulation index for children and adolescents (ERICA): A Psychometric investigation. *Journal of Psychopathology and Behavioral Assessment*, 32(3), 301–314.
- Mandy, W., Murin, M., Baykaner, O., Staunton, S., Hellriegel, J., Anderson, S., & Skuse, D. (2016). The transition from primary to secondary school in mainstream education for children with autism spectrum disorder. *Autism*, 20(1), 5–13.
- Mayes, S. D., Calhoun, S. L., Murray, M. J., Ahuja, M., & Smith, L. A. (2011). Anxiety, depression, and irritability in children with autism relative to other

- neuropsychiatric disorders and typical development. *Research in Autism Spectrum Disorders*, 5(1), 474–485.
- Mazefsky, C. A., Herrington, J., Siegel, M., Scarpa, A., Maddox, B. B., Scahill, L., & White, S. W. (2013). The role of emotion regulation in autism spectrum disorder. *Journal of the American Academy of Child and Adolescent Psychiatry*, 52(7), 679–688.
- *Mazefsky, C. A., & White, S. W. (2014). Emotion regulation: concepts and practice in autism spectrum disorder. *Child and Adolescent Psychiatric Clinics of North America*, 23(1), 15–24.
- McLaughlin, K. A., Hatzenbuehler, M. L., Mennin, D. S., & Nolen-Hoeksema, S. (2011). Emotion dysregulation and adolescent psychopathology: A prospective study. *Behaviour Research and Therapy*, 49(9), 544–554.
- Nauta, M. H., Scholing, A., Rapee, R. M., Abbott, M., Spence, S. H., & Waters, A. (2004). A parent-report measure of children's anxiety: Psychometric properties and comparison with child-report in a clinic and normal sample. *Behaviour Research and Therapy*, 42(7), 813–839.
- Ozsvadjian, A., Hibberd, C., & Hollocks, M. J. (2014). Brief report: The use of self-report measures in young people with autism spectrum disorder to access symptoms of anxiety, depression and negative thoughts. *Journal of Autism and Developmental Disorders*, 44(4), 969–974.
- *Pouw, L. B., Rieffe, C., Oosterveld, P., Huskens, B., & Stockmann, L. (2013a). Reactive/proactive aggression and affective/cognitive empathy in children with ASD. *Research in developmental disabilities*, 34(4), 1256–1266.
- *Pouw, L. B., Rieffe, C., Stockmann, L., & Gadwo, K. D. (2013b). The link between emotion regulation, social functioning, and depression in boys with

ASD. *Research in Autism Spectrum Disorders*, 7(4), 549-556.

Rieffe, C., Broekhof, E., Kouwenberg, M., Faber, J., Tsutsui, M. M., & Güroğlu, B. (2016). Disentangling proactive and reactive aggression in children using self-report. *European Journal of Developmental Psychology*, 13(4), 439–451.

*Rieffe, C., Camodeca, M., Pouw, L. B. C., Lange, A. M. C., & Stockmann, L. (2012). Don't anger me! Bullying, victimization, and emotion dysregulation in young adolescents with ASD. *European Journal of Developmental Psychology*, 9(3), 351–370.

*Rieffe, C., De Bruine, M., De Rooij, M., & Stockmann, L. (2014). Approach and avoidant emotion regulation prevent depressive symptoms in children with an autism spectrum disorder. *International Journal of Developmental Neuroscience*, 39, 37-43.

*Rieffe, C., Oosterveld, P., Terwogt, M. M., Mootz, S., Van Leeuwen, E., & Stockmann, L. (2011). Emotion regulation and internalizing symptoms in children with autism spectrum disorders. *Autism*, 15(6), 655-670.

Rieffe, C., Terwogt, Mark, M., Petrides, K, V., Cowan, R., Miers, Anne, C., & Tolland, A. (2007). Psychometric properties of the emotion awareness questionnaire for children. *Personality and Individual Differences*, 43(1), 95–105.

Rieffe, C., Terwogt, M. M., & Bosch, J. D. (2004). Emotion understanding in children with frequent somatic complaints. *European Journal of Developmental Psychology*, 1(1), 31–47.

Rothbart, M. K., Ahadi, S. A., Hershey, K. L., & Fisher, P. (2001). Investigations of temperament at three to seven years: The children's behavior questionnaire. *Child Development*.

Salomone, E., Kutlu, B., Derbyshire, K., McCloy, C., Hastings, R. P., Howlin, P., &

- Charman, T. (2014). Emotional and behavioural problems in children and young people with autism spectrum disorder in specialist autism schools. *Research in Autism Spectrum Disorders*, 8(6), 661–668.
- Samson, A. C., Hardan, A. Y., Podell, R. W., Phillips, J. M., & Gross, J. J. (2014). Emotion regulation in children and adolescents with autism spectrum disorder. *Autism Research*, 8(1), 9–18.
- *Samson, A. C., Wells, W. M., Phillips, J. M., Hardan, A. Y., & Gross, J. J. (2015). Emotion regulation in autism spectrum disorder: Evidence from parent interviews and children’s daily diaries. *Journal of Child Psychology and Psychiatry*, 56(8), 903–913.
- *Samyn, V., Roeyers, H., & Bijttebier, P. (2011). Effortful control in typically developing boys and in boys with ADHD or autism spectrum disorder. *Research in Developmental Disabilities*, 32(2), 483-490.
- *Samyn, V., Roeyers, H., Bijttebier, P., Rosseel, Y., & Wiersema, J. R. (2015). Assessing effortful control in typical and atypical development: Are questionnaires and neuropsychological measures interchangeable? A latent-variable analysis. *Research in developmental disabilities*, 36, 587-599.
- *Samyn, V., Wiersema, J. R., Bijttebier, P., & Roeyers, H. (2014). Effortful control and executive attention in typical and atypical development: An event-related potential study. *Biological psychology*, 99, 160-171.
- Scarpa, A., & Reyes, N. M. (2011). Improving emotion regulation with CBT in young children with high functioning autism spectrum disorders: a pilot study. *Behavioural and Cognitive Psychotherapy*, 39(04), 495–500.
- Shields, A., & Cicchetti, D. (1997). Emotion regulation among school-age children: The development and validation of a new criterion Q-sort scale. *Developmental*

Psychology, 33(6), 906–916.

Simonoff, E., Pickles, A., Charman, T., Chandler, S., Loucas, T., & Baird, G. (2008).

Psychiatric disorders in children with autism spectrum disorders: Prevalence, comorbidity, and associated factors in a population-derived sample. *Journal of the American Academy of Child & Adolescent Psychiatry*, 47(8), 921–929.

*South, M., Newton, T., & Chamberlain, P. D. (2012). Delayed reversal learning and association with repetitive behaviour in autism spectrum disorders. *Autism Research*, 5(6), 398-406.

Stratis, E. A., & Lecavalier, L. (2015). Informant agreement for youth with autism spectrum disorder or intellectual disability: A meta-analysis. *Journal of Autism and Developmental Disorders*, 45(4), 1026–1041.

Thomson, K., Burnham Riosa, P., & Weiss, J. A. (2015). Brief report of preliminary outcomes of an emotion regulation intervention for children with autism spectrum disorder. *Journal of Autism and Developmental Disorders*, 45(11), 3487–3495.

Totsika, V., Hastings, R. P., Emerson, E., Lancaster, G. A., & Berridge, D. M. (2011). A population-based investigation of behavioural and emotional problems and maternal mental health: associations with autism spectrum disorder and intellectual disability. *Journal of Child Psychology and Psychiatry*, 52(1), 91–99.

*Van Hecke, A. V., Lebow, J., Bal, E., Lamb, D., Harden, E., Kramer, A., ... & Porges, S. W. (2009). Electroencephalogram and heart rate regulation to familiar and unfamiliar people in children with autism spectrum disorders. *Child development*, 80(4), 1118-1133.

Van Steensel, F. J. A., Bögels, S. M., & Perrin, S. (2011). Anxiety Disorders in children and adolescents with autistic spectrum disorders: A meta-analysis. *Clinical Child and Family Psychology Review*, 14(3), 302–317.

- Watson, D., Clark, L. A., & Tellegen, A. (1988). Development and validation of brief measures of positive and negative affect: The PANAS scales. *Journal of Personality and Social Psychology*, 54(6), 1063–1070.
- Webb, T. L., Miles, E., & Sheeran, P. (2012). Dealing with feeling: a meta-analysis of the effectiveness of strategies derived from the process model of emotion regulation. *Psychological bulletin*, 138(4), 775.
- Weiss, J. A. (2014). Transdiagnostic Case conceptualization of emotional problems in youth with ASD: An emotion regulation approach. *Clinical Psychology: Science and Practice*, 21(4), 331–350.
- Weiss, J. A., Thomson, K., & Chan, L. (2014). A systematic literature review of emotion regulation measurement in individuals with autism spectrum disorder. *Autism Research*, 7(6), 629–648.
- Wells, G. A., Shea, B., O'Connell, D., Peterson, J., Welch, V., Losos, M., & Tugwell, P. (2013). The Newcastle-Ottawa Scale (NOS) for assessing the quality of nonrandomized studies in meta-analyses. *The Ottawa Hospital Research Institute*, (3), 1–4.
- World Health Organization. (1993). *The ICD-10 classification of mental and behavioural disorders: diagnostic criteria for research*. World Health Organization.
- Wright, M., Banerjee, R., Hoek, W., Rieffe, C., & Novin, S. (2010). Depression and social anxiety in children: Differential links with coping strategies. *Journal of Abnormal Child Psychology*, 38(3), 405–419.
- Wykes, T., Huddy, V., Cellard, C., McGurk, S. R., & Czobor, P. (2011). A meta-analysis of cognitive remediation for schizophrenia: Methodology and effect sizes. *American Journal of Psychiatry*, 168(5), 472–485.

*Yager, J., & Iarocci, G. (2013). The development of the multidimensional social competence scale: A standardized measure of social competence in autism spectrum disorders. *Autism Research*, 6(6), 631–641.

Part 2: Empirical Research Paper

**Exploring the Role of Parent-Reported Executive Function in
Pathological Demand Avoidance Behaviours**

2.1 Abstract

Aims: Pathological Demand Avoidance (PDA) is an increasingly used label that describes a subset of children diagnosed with Autism Spectrum Disorder (ASD) who display extreme behaviours in response to everyday demands. To date, little is known about the cognitive profiles associated with these behaviours. The aim of this study was to investigate the potential role of parent-reported Executive Function (EF) in PDA traits in the context of ASD, with a particular focus on emotion dysregulation and non-compliance behaviour.

Methods: Sixty-four parents of children with ASD and 31 parents of children without ASD, aged between 6-11 years old, were recruited to complete questionnaires at a single time point. Questionnaires assessed ASD traits, PDA traits, EF, non-compliance behaviours, and emotion dysregulation.

Results: Group differences in parent-reported EF were found, with children with ASD showing greater deficits than children without ASD. EF was significantly related to global PDA traits in the context of ASD, as well as to dimensional measures of specific associated behaviours such as emotional dysregulation and behavioural non-compliance. Executive Function accounted for a significant proportion of the variance ($r^2 = 34\%$) in PDA traits. Indices of EF were found to differentially predict non-compliance behaviours and ASD traits confounded some of these relationships.

Conclusion: These findings suggest that a range of EF deficits may contribute to PDA behaviours in children with ASD. Evidence for differential involvement of EF in PDA traits suggests that further exploration using more direct measures of EF is warranted to gain a clearer understanding of this relationship, which could benefit interventions and behavioural strategy development.

2.2 Introduction

2.2.1 Autism Spectrum Disorder (ASD)

‘Autism Spectrum Disorder’ (ASD) is a pervasive developmental disorder that is defined along two continuums: (1) persistent difficulties with social communication and social interaction; (2) difficulties with restricted and repetitive patterns of behaviours or interests (American Psychiatric Association, 2013). The first continuum includes problems with socio-emotional reciprocity, deficits in verbal and non-verbal communication used for social interaction, and difficulties understanding, forming and maintaining relationships. The second continuum comprises problems with stereotyped behaviours and speech, insistence on sameness, routines or rituals, highly fixated or restricted interests, and sensory abnormalities. These behavioural difficulties must be present from early childhood, to the extent that they limit and impair the individual’s everyday functioning. Most individuals with ASD show reduced social motivation, and appear socially naïve or peculiar (Wing, 1991). In general, difficulties with developing rapport, maintaining conversation, or responding in socially appropriate ways form a key component of our concept of ASD (O’Nions, Viding, Greven, Ronald, & Happé, 2014).

Population-based epidemiological studies have reported a large increase in the estimated prevalence of ASD, from around 30 per 10,000 in 2000 (Baron-Cohen et al., 2000) to approximately 100 per 10,000 in 2009 (Baron-Cohen et al., 2009). This increase may reflect several factors, including a growing awareness of ASD and a widening of the diagnostic criteria (King & Bearman, 2009; Wing & Potter, 2002). It is recognised that ASD represents a broad spectrum of individuals who show diverse combinations of difficulties and symptoms and who may vary substantially in both their social and cognitive presentations (Nydén, Hagberg, Goussé, & Rastam, 2011;

Wing, 1991; Towgood, Meuwese, Gilbert, Turner, & Burgess, 2009). However, those who have atypical presentations which do not neatly fit diagnostic or conceptual boundaries still produce a major challenge in clinical practice (O’Nions, Viding, et al., 2014).

2.2.2 Pathological Demand Avoidance (PDA)

‘Pathological Demand Avoidance’ (PDA; Newson, Le Maréchal, & David, 2003) is a pattern of difficulties, conceptualised as capturing important elements of variability seen amongst children with ASD. Elizabeth Newson first used the phrase to describe a subset of children diagnosed with ‘atypical autism’ yet who displayed very similar characteristics to each other. Newson reported that many of the observed behaviours were not necessarily what one would expect to see in a child who had autism. There was also an unusually high number of females, with rates of females equalling that of males, whereas gender ratios found in ASD itself are closer to three males to one female (Loomes, Hull, & Mandy, 2017).

Newson et al. (2003) described eight key areas of PDA (see Figure 2.1). Importantly, within this description there were several characteristics where those with ASD who resembled Newson’s descriptions were reported to differ from those who met the criteria for ASD but for whom Newson did not apply the term PDA. One of the main differences described by Newson was the ability to use socially manipulative strategies to avoid demands. Although it is well documented that children with ASD can also display difficulties with compliance (Arbelle, Sigman, & Kasari, 1994; Bryce & Jahromi, 2013; Lemanek, Stone, & Fishel, 1993; Ostfeld-Etzion, Feldman, Hirschler-Guttenberg, Laor, & Golan, 2016), the responses from the children Newson identified as having PDA differed from those with ASD only. For example, when prompted to comply, children with ASD and PDA traits reacted with socially shocking

or manipulative behaviour, or avoided demands by distraction, diversion, or threats (Newson et al., 2003).

- PDA Features described by Newson et al., (2003)**
1. A passive early history where children continue to resist or avoid ordinary life demands
 2. Lability of mood
 3. Impulsivity
 4. Behaviour led by a need for control
 5. At ease in role play
 6. A language delay, which appears to be due to passivity
 7. Obsessive behavior
 8. Neurological signs, such as clumsiness or physical awkwardness

Figure 2.1 Description of the key PDA characteristics.

Newson et al. (2003) also included ‘extreme emotional lability’ as a key PDA trait. This observation has been supported by O’Nions and Viding et al. (2014) who reported evidence that children with ASD and PDA traits displayed emotional symptoms exceeding those found in children with ASD only. More recently, further evidence of difficulties with emotion regulation were highlighted by O’Nions et al. (2016). They found that individuals with higher PDA traits endorsed more items on the Diagnostic Interview for Social and Communication Disorders (DISCO; Wing, Leekam, Libby, Gould, & Larcombe, 2002) that were related to emotion regulation difficulties than individuals with lower or no PDA traits, including inappropriate behavioural responses and frequent fluctuations in emotional expression. Interestingly,

Newson et al. (2003) considered that problem behaviour shown by children with PDA may in fact be a manifestation of emotional symptoms in ASD. For example, they argued that demands may be perceived as threatening and therefore outbursts could be viewed as a panic attack.

Reports suggest that children with ASD who fit the PDA profile suffer with extreme behavioural impairment and challenges (Eaton & Banting, 2012; Newson et al., 2003) which can ultimately result in high levels of educational placement breakdown (Gore Langton & Frederickson, 2016). In order to support the development of interventions and strategies to help with these challenges, the need for a tool to identify PDA features in individuals with ASD was raised (O’Nions et al., 2016; O’Nions, Christie, Gould, Viding, & Happé, 2014). O’Nions and Christie et al. (2014) developed the Extreme Demand Avoidance Questionnaire (EDA-Q), a parent-rated measure that enables the key traits of PDA to be quantified. Although children who were reported to have PDA scored significantly higher on the EDA-Q, a large proportion of children with ASD plus behaviour problems also scored highly. This could suggest that a number of the traits that are characteristic of PDA may not be very specific to the PDA phenotype and may in fact be relatively common across the autism spectrum and beyond, for example in children with disruptive behaviour who do not have ASD.

Although, there still remains debate around how to conceptualise the profile described as ‘PDA’, at the behavioural level it may represent an extreme of a non-compliance trait in ASD, coupled with extreme lability of mood and need for control. A recent review of the evidence describing these behaviours revealed that although evidence has not emerged in support of PDA as a discrete syndrome, it represents an important range of co-occurring difficulties for many children with ASD that can

substantially impact families (Green et al., 2018). Therefore, it seems important to explore the potential underpinnings of these behaviours with a broader aim of further elucidating this specific phenotype. Explorations of the cognitive profile may be able to help settle some of the debate surrounding this potential diagnosis and explain the behavioural profile (O’Nions, Viding, et al., 2014).

2.2.3 Executive Function (EF)

Executive Function is an umbrella term that incorporates a collection of high-level, inter-related cognitive processes responsible for purposeful, goal-directed behaviour, thought to be mediated primarily by the frontal lobes (Anderson, 2002; Kuhn, 2015; Ye, AuCoin-Power, Taylor, & Doesburg, 2015). Although a widely used term, there remains a lack of clarity and some controversy surrounding the formal definition of EF (Jurado & Rosselli, 2007). However, processes such as, response inhibition, initiation, working memory, attention, switching, and planning, that control thought and behaviour (Anderson, 2002; Gioia, Isquith, Guy, & Kenworthy, 2001; Goldstein, Naglieri, Princiotta, & Otero, 2014; Stuss & Knight, 2002), along with behaviours related to emotional responses, self-regulation and self-monitoring (Gioia, Isquith, Guy, & Kenworthy, 2000), are often cited as falling under this umbrella.

Executive Function impairments are common to many developmental disorders and have been considered one of the crucial mechanisms underlying behavioural problems (Johnson, 2012). In particular, EF has been extensively researched and implicated in ASD (Hill, 2004b; Kenworthy, Yerys, Anthony, & Wallace, 2008). Previous reviews provided evidence of overall impairments in EF in individuals with ASD but also highlight the often fairly modest effect sizes and numerous disparities between findings across ages and EF components (Geurts, Corbett, & Solomon, 2009; Hill, 2004a). Historically, others have failed to find any

EF deficits in individuals with ASD (Baron-cohen, Wheelwright, Stone, & Rutherford, 1999; Hill & Russell, 2002; Minshew, Muenz, Goldstein, & Payton, 1992; Russell & Hill, 2001). However, the most recent meta-analyses exploring this relationship have found evidence to support the executive dysfunction hypothesis of ASD (Demetriou et al., 2018; Lai et al., 2017).

One explanation for previous inconsistent findings of EF impairments in ASD is the methodological difficulties associated with investigating EF in ASD (Burgess et al., 2006). The challenges associated with EF measurement have long been recognised (Denckla, 1994). Many performance tests exist for assessing EF in explicit ways, but their ecological validity and generalisability has been questioned as cognitive assessments often do not support observational information from everyday tasks (Wilson, 1993). Evidence has not yet been found for a ‘perfect’ way in which to quantify everyday cognitive ability due to all assessments carrying a certain amount of error (Chaytor, Schmitter-Edgecombe, & Burr, 2006). Therefore, informant-based questionnaires and clinician rating scales have been heavily relied upon to investigate the ecological validity of EF assessment.

Although evidence suggests that the severity of EF deficits may predict ASD symptom severity (Eylen, Boets, Steyaert, Wagemans, & Noens, 2015), there are still limited studies investigating specific relationships between EF and ASD symptoms (Leung, Vogan, Powell, Anagnostou, & Taylor, 2016). Therefore, it is unclear as to which ASD symptoms are most related to EF deficits. The overarching aim of this study is to investigate the potential relationship between EF difficulties and PDA traits in children with ASD. Although recent meta-analyses confirm EF deficits in individuals with ASD, this difference has not yet been investigated in a non-clinic selected ASD group that may be enriched for PDA traits. Therefore, the first aim of

this study is to compare parent-reported EF difficulties between children with and without ASD.

2.2.4 Executive Function and PDA

Lability of mood, a key PDA trait, falls under the umbrella of emotion regulation (Thompson, 2008), which has been shown to be a difficulty for some children and adolescents with ASD (Mazefsky et al., 2013; Weiss et al., 2014). Interestingly, evidence suggests that EF may be involved in the ability to regulate emotions in typically developing (TD) children and adolescents (Carlson & Wang, 2007; Gyurak, Goodkind, Kramer, Miller, & Levenson, 2012; Zelazo & Cunningham, 2007). There are many aspects of EF that could be related to processes considered necessary for emotion regulation, such as the ability to make evaluations and predictions about a situation, to modify one's emotional reactions through appraisals of a situation, and to behaviourally regulate how emotions are expressed (Gross & Thompson, 2007). The role of EF in emotion dysregulation seems an important avenue for investigation; it could be that EF deficits in children with ASD and PDA traits contribute to the observed difficulties with emotional lability.

In TD children, relationships have been found between EF deficits and problem behaviours, including non-compliance (Espy, Sheffield, Wiebe, Clark, & Moehr, 2011; Hughes & Ensor, 2008). Evidence suggests that children with ASD have difficulties with EF skills such as inhibition, stopping and changing behaviour in response to a stimulus, and cognitive flexibility (Mostert-Kerckhoffs, Staal, Houben, & de Jonge, 2015), all of which could impact their ability to comply with demands. Interestingly, it has also been found that variables related to demands, such as open ended rather than structured tasks, may lead to more pronounced performance deficits associated with EF impairments (Eylen et al., 2015). As it is known that EF deficits

can impact behavioural control, and links between EF deficits and non-compliance have been reported in the TD population, it seems sensible to consider that EF may also play a role in non-compliance behaviour.

Non-compliance behaviour may be ‘Demand Specific’, for instance, occur when a child is in a situation where a direct demand requiring effort, such as washing, dressing or chores, is placed on them (Chowdhury et al., 2016). This could be seen as an example of a ‘rich to lean’ transition (Brewer, Strickland-Cohen, Dotson, & Williams, 2014), where a child is asked to move from a preferred activity to a less preferred activity, and where ‘Demand Specific’ problem behaviours may have been negatively reinforced due to the removal of the expected transition when problem behaviour occurs. The ability to inhibit and initiate behaviour, shift to changing demands and modulate emotional responses, all fall under the EF umbrella. These processes seem necessary in order to cope with transitioning from one task to another, especially when the demand involves moving to a less favourable task.

It has also been suggested that children with developmental difficulties may struggle with ‘Socially Inflexible’ behaviours that occur in response to demands that deviate from an expected schedule or, require adjustment to people or places outside of the child’s comfort zone (Chowdhury et al., 2016). The ability to cope with transitions from routine or expected schedules involves a capacity to tolerate a certain level of uncertainty. Poor tolerance of uncertainty is one explanation for why children with ASD may routinely resist demands requiring participation in potentially unpredictable situations (Brewer et al., 2014). A strong conceptual overlap has been found between poor tolerance of uncertainty and EF (Mushtaq, Bland, & Schaefer, 2011), suggesting that the ability to use EF skills is an important part of being able to cope with uncertainty. Therefore, it seems important to consider the potential role of

EF in individuals with ASD and PDA traits when considering their non-compliance in response to unexpected demands.

Overall, it appears that EF deficits may contribute to behaviours described in accounts of PDA in the context of ASD, and specific EF components could relate to specific PDA traits. One might expect children with ASD who resemble descriptions of PDA to show deficits in EF domains above those with lower level features, related to these behaviours, since EF deficits has been found to contribute to these behaviours in ASD more generally. To date, no other studies have investigated the EF profile of children with ASD and PDA traits or how it relates to behaviours associated with PDA. Given that there is still little known about PDA, it is important to continue to investigate this presentation with the broader aim of providing better informed interventions and educational support strategies. In order to better understand the neurocognitive underpinnings of PDA, this study focused on assessing the parent-reported EF profile of children with ASD and PDA traits, with a particular focus on investigating the potential involvement of EF in non-compliance behaviours and emotion dysregulation.

2.2.5 Research aims and predictions

Firstly, this study aimed to establish whether children with ASD and PDA traits show parent-reported deficits in EF above that of children without ASD. Although recent findings suggest that individuals with ASD have difficulties with EF above those seen in TD groups (Demetriou et al., 2018; Lai et al., 2017), this has not been investigated in a sample of children with ASD and PDA traits. This study further aimed to further investigate the relationship between any deficits in EF and PDA features in general, emotional dysregulation, and specific PDA-related difficulties with non-compliance.

Overall it was expected that there would be differences between the ASD group and the non-ASD group in their EF abilities. More specifically, we predicted that the ASD group would have greater difficulties in all areas of EF. We hypothesised that difficulties in EF would be associated with symptoms of PDA. More specifically we expected that EF deficits would predict global symptoms of PDA, difficulties with emotion regulation, and difficulties with non-compliance behaviour. At this time, more specific hypotheses regarding the involvement of EF indices were not made due to a lack of supporting literature on which to base these. However, this study explored whether different indices of EF were differentially related to symptoms of emotion dysregulation and non-compliance.

2.3 Method

2.3.1 Design

This study used a cross-sectional parent-report questionnaire methodology to investigate the relationship between EF deficits and PDA traits in children with ASD. This study was undertaken as part of a joint project with Ellie Bishop who investigated the relationship between Theory of Mind and PDA traits. A description of the individual trainee contributions to the project can be found in Appendix I.

2.3.1.1 Ethical approval

Ethical approval for the study was given by University College London (UCL) Ethics Committee (reference 10193/001; Appendix III). Participants volunteered to take part and informed consent was obtained from all participants prior to any data collection. The information sheet and consent form for parents can be found in Appendix IV-V.

2.3.1.2 Sample size and power analysis

Power calculations were performed to estimate an appropriate sample size for this study. This was done using the “G*Power 3” computer programme (Faul, Erdfelder, Lang, & Buchner, 2007), specifying alpha = 5% and desired power = 80%. Based on assumptions of obtaining a medium to large effect size (Cohen, 1992), which were formed from previous research in this field (O’Nions, Viding, et al., 2014), a minimum sample size of 60 was required.

2.3.1.3 Participants

In total, 95 parents of children (50 male; 45 female), aged 6-11 years old ($M = 8$ years, 8 months; $SD = 1$ year, 6 months), participated in this study. From this, 64 participants reported their child to have previously received a diagnosis of ASD. The remaining 31 participants were parents of children who had not previously received a diagnosis of ASD and so were included in the non-ASD control group. The overall sample consisted of 96% White British, 2% Asian British and 2% Other. For the non-ASD sample, the ages ranged from 6.42-11.58 years, and for the ASD sample the ages ranged between 6-11.92 years. Further participant characteristics are described in Table 2.1.

Table 2.1*Demographic and clinical characteristics of participants*

Measure	Group				Analysis		
	Non-ASD (N=31)		ASD (N = 64)		χ^2	p-value	Phi (Φ)
	N	%	N	%			
<i>Gender</i>					5.43	p<.05	-.24*
Male	11	35.5	39	60.9			
Female	20	64.5	25	39.1			
<i>Additional Diagnoses</i>							
None	29	93.5	42	65.6	8.62	p<.01	.30**
ADHD	0	0	7	10.9			
Mild LD	0	0	4	6.3			
Learning Difficulties	2	6.5	4	6.3			
Other	0	0	7	10.9			
					FET	p-value	Phi (Φ)
<i>Education</i>							
Mainstream	31	100	37	57.8	18.27	p<.001	.44***
Home School	0	0	11	17.2			
Specialised Unit/School	0	0	10	15.6			
Not in Education	0	0	2	3.1			
Other	0	0	4	6.3			
	M	SD	M	SD	t	p-value	d
Age (months)	100.97	19.04	108.7	19.29	-1.84	.069	.40
SDQ (Total)	7.13	3.80	23.94	5.24	-15.93	1<2***	3.67
Emotional Problems	2.23	1.82	6.98	2.31	-10.06	1<2***	2.28
Conduct problems	1.19	1.38	4.47	2.07	-7.99	1<2***	1.86
Hyperactivity	3.06	2.25	7.11	2.41	-7.83	1<2***	1.74
Peer Problems	0.65	1.02	5.38	1.96	-15.49	1<2***	3.03
Prosocial Behaviours	7.68	1.01	4.50	1.75	11.18	1>2***	2.23
Internalising Problems	2.87	2.05	12.36	3.26	-17.30	1<2***	3.49
Externalising Problems	4.26	3.01	11.58	3.45	-10.08	1<2***	2.26
	Median		Median		U	p-value	r
Cast Total	2		20		5.50	1<2***	-.80

*p<.05 **p<.01 ***p<.001

2.3.2 Procedure

2.3.2.1 Participant recruitment

Participants were recruited through social media advertising and convenience sampling through the research team's networks, schools and snowballing methods. An advertising poster was created and distributed on social media outlets such as Facebook and Twitter, as well as being sent to local and national organisations such as The National Autistic Society and The PDA Society (see Appendix VI). Recruitment methods were designed to specifically access individuals with ASD who displayed PDA traits. Importantly, to ensure the final ASD sample was representative of those who struggle with PDA traits, recruitment was monitored to ensure around half of the children in the group received parent-reported scores on the EDA-Q above the cut-off of 50 (O'Nions, Christie, et al., 2014).

In total, 208 participants were screened for inclusion in this study. To establish parent eligibility the following inclusion criteria were applied: (i) over the age of 18 years old; (ii) parent of a child aged 6-11 years old; (iii) sufficient levels of English language necessary to complete questionnaires. Parents were excluded from this study if they reported their child to have a moderate or severe learning disability.

Once participants were identified as meeting basic inclusion and exclusion criteria their responses to the following criteria identified whether they could be included in the ASD group or the non-ASD group. To be included in the non-ASD group participants had to report that their child had: (i) no formal ASD diagnosis; (ii) a score of less than the clinical cut-off of 14 on the Child Autism Spectrum Test (CAST; Scott et al., 2002); (iii) a score below the clinical cut-off of 16 on the Strengths and Difficulties Questionnaire (SDQ; Goodman, 1997); a score below the cut-off of 50 on the Extreme Demand Avoidance Questionnaire (EDA-Q; O'Nions et al., 2014).

To be included in the ASD group participants had to report that their child had received a clinical diagnosis of ASD, autism, high-functioning autism (HFA) or Asperger's syndrome from a qualified clinician.

Overall, 110 participants were identified as being eligible to participate in this study, and a total of 95 parents completed the study. This resulted in a total of 64 participants in the ASD group and 31 in the non-ASD group.

2.3.2.2 Questionnaire administration

Participants volunteered anonymously to take part in the study and were then given the option of completing questionnaires online or via post; all participants chose to complete questionnaires online. After expressing interest in the study, participants were then contacted by email to thank them for their interest and given the necessary instructions in order to access 'Phase One' of the online questionnaires (see Appendix VII). Questionnaires were accessed via an online link, which required the participant to enter their unique identification number and a password, which were all provided in the email. All online questionnaires were set-up using Qualtrics.

After following the link participants were first instructed to read an information sheet about the study which detailed the aims, procedure, and informed participants of their right to withdraw at any time (see Appendix IV). After reading this, participants were asked to provide informed consent (see Appendix V) and could only continue with the study once consent had been obtained. Participants were not paid for their time but were entered into a prize draw to win Amazon vouchers.

Participants were then taken through a set of demographic questions (see Appendix VIII), followed by the SDQ (Goodman, 1997), the CAST (Scott, Baron-Cohen, Bolton, & Brayne, 2002) and the EDA-Q (O'Nions, et al., 2014), examples of which can be found in Appendix IX-XIV. Instructions for all questionnaires were

provided online at the top of each questionnaire. There were also free text spaces available after each questionnaire for participants to leave any specific feedback if they wished to.

After completing phase one, any participants who were identified as not being eligible to continue with the study were contacted with a follow-up email thanking them for their time. Participants who were identified as being eligible were contacted by email again and asked to complete ‘Phase Two’ of the study (see Appendix VII). Phase Two contained a further set of online questionnaires (see Appendix IX-XIV), which included the Behaviour Rating Inventory of Executive Function (BRIEF; Gioia et al., 2000), the Home Situations Questionnaire-ASD version (HSQ-ASD; Chowdhury et al., 2016), and the Affective Reactivity Index (ARI; Stringaris et al., 2012). Participants were sent regular reminder emails to ask them to complete all of the questionnaires and also reminding them of their right to withdraw from the study at any time. Raw data were stored on Qualtrics and downloaded for analysis in SPSS once all data had been collected. All data were kept anonymously and stored in accordance with the UK Data Protection Act.

2.3.3 Parent-report measures

2.3.3.1 Pathological Demand Avoidance

Overall traits relating to the PDA profile were assessed using the Extreme Demand Avoidance - Questionnaire (EDA-Q; O’Nions, Christie, et al., 2014). The EDA-Q (see Appendix IX) is a parent-report questionnaire which presents 26 statements that describe key PDA behaviours described by Newson et al. (2003). It asks parents to rate on a 4-point Likert scale (e.g. 0 = “Not True” to 3 = “Very True”) how true each statement is in relation to their child. Statements ask parents to consider a range of behaviours related to the PDA profile. For example, the measure includes,

but is not limited to, questions referring to demand avoidance behaviours, “Obsessively resists and avoids ordinary demands and requests”, others their emotional lability, “Mood changes very rapidly”, and some their developmental difficulties, “S/he was passive and difficult to engage as an infant”. The EDA-Q is the only specific measure of PDA traits and has been shown to have high internal consistency (.87) and high split-half reliability (.91). Preliminary evidence also suggests good validity and discriminatory power in children (5-17 years), with the EDA-Q distinguishing PDA groups from non-ASD groups and ASD groups, with good sensitivity (.88) and specificity (.78).

2.3.3.2 Executive Function

The Behaviour Rating Inventory of Executive Function (BRIEF; Gioia et al., 2000) was used to assess EF. The BRIEF (see Appendix X) is a parent- or teacher-rated questionnaire for children aged 5-18 years consisting of 86-items and takes about 10 minutes to complete. Each item asks the parent to comment using a 3-point Likert scale (e.g. 1 = “Never” to 3 = “Often”), on how frequently over the past six months certain behaviours such as, “Has a short attention span”, or “Interrupts others”, have been a problem. It is designed to assess EF in everyday life that may not be captured by performance measures and therefore may have more ecological validity than other tests of EF (Denckla, 1994).

This measure has been demonstrated to have adequate test-retest reliability for teachers (.88) and for parents (.82; Gioia et al., 2000). The BRIEF also shows good convergent and discriminant validity when compared to tests of behaviour (Gioia, Isquith, & Guy, 1998). Associations have been found between the BRIEF and performance-based measures of EF (Toplak, Bucciarelli, Jain, & Tannock, 2008; Vriezen & Pigott, 2002) and between BRIEF scores and neural substrate of EF

(Anderson, Northam, Jacobs, Mikiewicz, & Anderson, 2002; Anderson, Jacobs, & Harvey, 2005). A recent meta-analysis (Demetriou et al., 2018) suggests that the BRIEF may have clinical utility above and beyond that of more performance based measures, particularly for differentiating between individuals with ASD and TD controls. The BRIEF has been validated in a range of clinical samples, including ASD (Gioia, Isquith, Retzlaff, & Espy, 2002) and *T*-scores of 65 or more are indicative of clinically significant symptoms.

There are eight clinical scales that can be combined to form two separate indices. The Behaviour Regulation Index (BRI), which includes the Inhibit, Shift and Emotional Control scales, represents the ability to shift cognitive set and modulate behaviour and emotions. The Metacognition Index (MI), which includes the Initiate, Working Memory, Plan/Organise, Organisation of Materials, and Monitor scales, represents the ability to get started on an activity, plan, organise and hold information in mind for future-orientated problem solving (Roth, Isquith, & Gioia, 2014).

2.3.3.3 Behavioural non-compliance

The Home Situations Questionnaire-ASD (HSQ-ASD) is a caregiver questionnaire designed to assess behavioural non-compliance by children with ASD in everyday situations. The HSQ-ASD (see Appendix XI) was originally developed in TD children (Barkley & Edelbrock, 1987) and was modified by Chowdury et al. (2016) to a 24-item version specifically for use with children with ASD. It asks parents to consider whether their children have had difficulties with following instructions, commands or rules in specific situations and to rate how severe these problems are on a scale (e.g. 1 = Mild, 9 = Severe). It has been found to have satisfactory construct validity, internal validity and test-retest reliability, as well as good convergent and divergent validity.

In this study it was used as a dimensional measure of non-compliance and demand avoidance. The HSQ-ASD has two subscales which assess non-compliance. The first, labelled the Demand Specific subscale (HSQ-DS), assesses difficulties with compliance in response to direct demands such as, “Getting up in the morning”, or “When moving from one activity to another”. The second, labelled the Social Inflexibility subscale (HSQ-SI), measures difficulties with compliance in response to unexpected or non-routine demands, for instance demands given “In public places”, or “When there is an unexpected change in daily routine”.

2.3.3.4 Emotion dysregulation

The Affective Reactivity Index (ARI; Stringaris et al., 2012), is a 6-item scale that is both parent- and self-rated. It assesses irritability in children between the ages of 5-17 years and has been validated in a UK sample with a range of disorders, including ASD. The ARI (see Appendix XII) was used as a measure of emotional lability, which taps into one of the key behavioural difficulties found to be a difficulty for children who appear to struggle with traits related to the PDA profile. It uses a 3-point Likert scale (e.g. 0 = “Not True” to 2 = “Certainly True”) and asks parents to comment on how accurate each statement is of their child based on the last six months. The scale uses statements such as “Is easily annoyed by others” and “Often loses his/her temper”. Overall, the ARI showed excellent internal consistencies in TD children, with Cronbach’s alphas 0.89 (parent-report) and 0.90 (self-rated). Scores on the scale have been shown to discriminate between healthy children, children with bipolar and children with severe mood dysregulation (Stringaris et al., 2012).

2.3.3.5 Autism Spectrum Disorder

Traits of ASD were assessed using the Child Autism Spectrum Test (CAST; Scott et al., 2002) which is a 37-item parent report questionnaire. The CAST (see

Appendix XIII) asks parents to respond with either “Yes” or “No” to questions about their child such as “Was s/he speaking by 2 years old?” and “Are people important to him/her?”. The cut-off point for concerns of possible ASD is 15 or higher, with sensitivity being found to be 100%, and specificity 97%, and positive predictive value of 50% using research diagnostic assessments (Williams et al., 2005). The CAST shows moderate to good test–retest reliability (Allison et al., 2007).

2.3.3.6 Behaviour

The Strengths and Difficulties Questionnaire (SDQ; Goodman, 1997) was used as a measure of child behaviour. The SDQ (see Appendix XIV) has a parent-report version for use with children between the ages of 4-17 years that includes 25-items which screen for emotional, peer, social and behavioural problems. The SDQ asks parents to respond to statements about their child’s behaviour, such as “Considerate of other people’s feelings” and “Often unhappy, down-hearted or tearful”, on a 3-point Likert scale (e.g. 0 = “Not True” to 3 = “Certainly True”). It has been found to have satisfactory internal consistency and test-retest stability (Goodman, 2001). It has also been found to have good construct validity (Goodman & Scott, 1999) and discriminates well between children with and without psychopathological symptoms (Goodman, 1999).

2.3.4 Data analysis

Statistical analysis was conducted using SPSS version 25.0. Before running parametric tests, heterogeneity and normality checks were performed, details of which are outlined below.

Overall, there were 64 participants in the ASD group. For the ASD group, 57 out of the 64 recruited participants completed the ARI questionnaire, therefore all analysis involving data from this measure in the ASD group was performed on a

sample size of $n = 57$. For the HSQ-ASD, in order to receive a valid score, the respondent needs to answer nine or more of the questions from each subscale. One participant in the ASD group failed to answer enough questions on the Demand Specific subscale, and so for this subscale there was a final sample size of $n = 63$.

2.3.4.1 Participant characteristics

For all participant characteristic analyses, normality checks were performed to ensure distributions were appropriate for parametric tests. Distributions were visibly checked, and levels of skew and kurtosis were assessed by dividing the statistics by their standard error to produce a z -score. The cut-off of 3.29 was applied so that any value greater than this would indicate significant deviation from normality (Kim, 2013; Field, 2009). Age and SDQ data were found to meet assumptions of normality and equality of variance and therefore an independent samples t -test was performed. Significant levels of skew and were found within the CAST for the non-ASD group, so a non-parametric Mann-Whitney U test was used to assess group differences. An effect size of r was calculated for the Mann-Whitney U using the below equation where Z indicates the z -score produced from the Mann-Whitney analysis and N represents the sample size (Field, 2009):

$$r = Z/\sqrt{N}$$

Standardised effect sizes (Cohen's d ; Cohen, 1977) were also calculated for all t -tests using the difference between means for the ASD and non-ASD group, divided by the pooled standard deviation for the two groups. This is summarised in the following equations (Wykes et al., 2011):

$$\text{Effect Size } (d) = (M_{\text{ASD}} - M_{\text{NASD}}) / SD_{\text{pooled}}$$

Where M_{ASD} indicates the mean for the ASD group, M_{NASD} indicates the mean for the non-ASD comparison group, and SD_{pooled} indicates the pooled standard deviation for the ASD and non-ASD groups. Effect sizes were interpreted as: small = 0.2; medium = 0.5; large = 0.8 (Cohen, 1977).

Chi-square analyses were conducted to investigate relationships between group and gender, and group and additional diagnosis. A Fisher's Exact Test was conducted to assess the relationship between group and mainstream school education instead of a chi-squared analysis due to the count of children from the non-ASD group not in mainstream education equalling zero. For all chi-square analyses and Fisher's Exact Tests, Phi was calculated as a size of effect, where: small = 0.1; medium = 0.3; large = 0.5 (Cohen, 1988).

2.3.4.2 Group differences in EF

To examine the first hypothesis that there are differences in EF between ASD and non-ASD children, group differences between the ASD group and the non-ASD group on the BRIEF Global Executive Composite (GEC) and the two Indices, Behavioural Regulation Index (BRI) and Metacognition Index (MI), were examined. Normality checks were conducted by assessing levels of skew and kurtosis as described in the previous section. The GEC and MI were found to fall within the acceptable range for the assumption of normality. Therefore, for group comparisons, *t*-tests were performed, and Cohen's *d* effect sizes were calculated. For the ASD group, the BRI was found to show acceptable levels of kurtosis but slightly high levels of skew. Transformations were attempted on the BRI scale, but these did not lead to more acceptable levels of skew. Therefore, a non-parametric Mann-Whitney U test was conducted, and an effect size of *r* was calculated.

2.3.4.3 Correlations

Pearson's correlation analyses were conducted to investigate relationships between measures of EF and PDA traits within the ASD group. All assumptions of linearity and homoscedasticity were met. This analysis was only conducted within the ASD group to investigate the second hypothesis that PDA traits are associated with EF difficulties in children with ASD and PDA traits. Levels of skew and kurtosis were found to fall within the acceptable boundaries, as described earlier, for all variables assessing ASD and PDA traits.

2.3.4.4 Regression Analyses

Multiple and hierarchical regression analyses using a forced entry method were conducted to determine how much of the variance in PDA traits can be explained by indices of EF. For all final models, assumptions of independent errors between residuals, lack of multicollinearity, accuracy and lack of heteroscedasticity were fulfilled (see Appendix XV).

2.4 Results

2.4.1 Group characteristics

Table 2.1 shows demographic and clinical characteristics of children with ASD compared to non-ASD children. A chi-squared test of independence was performed to examine the relationship between group and gender. This relationship was significant, with children with ASD being more likely to be male (64%) than children without ASD (35%). A small effect size was found for this relationship.

A further chi-squared test of independence was performed to examine the relationship between group and additional diagnosis. This relationship was also significant, with children with ASD being more likely have an additional diagnosis

(34%) than children without ASD (6%). A medium effect size was found for this relationship.

A Fisher's Exact Test was conducted to assess the relationship between group and mainstream school education and a significant relationship was found. Therefore, children without ASD were more likely to be in mainstream education (100%) than children with ASD (54%). A medium effect size was found for this relationship.

When assessing dimensions of child behaviour problems, *t*-test results (see Table 2.1) showed that the ASD group had significantly higher scores on the SDQ Total and all subscales, indicating more difficulties. This was the case for all subscales except the Prosocial Behaviours subscale where the ASD group had significantly lower scores indicating more difficulties in this area. All of these differences were found to have very large effect sizes, with the smallest effect size being $d = 1.74$. A Mann-Whitney U test (see Table 2.1) indicated that the non-ASD group scored significantly lower on the CAST than the ASD group, which also resulted in a large effect size ($r = -.80$).

2.4.2 Executive Function deficits in children with ASD compared to children without ASD

Independent samples *t*-tests were conducted to assess the differences in EF between the non-ASD ($n = 31$) and ASD groups ($n = 64$). On average, the ASD group scored higher ($M = 78.98$, $SD = 7.77$) on GEC (with higher scores indicating more problems) than the non-ASD group ($M = 50.16$, $SD = 9.73$). This difference was significant, $t(93) = -15.58$, $p < .001$, and a very large effect size was found ($d = 3.27$). For the Meta-Cognition Index (MI), the ASD group on average scored higher ($M = 75.17$, $SD = 7.19$) than the non-ASD group ($M = 49.61$, $SD = 9.52$) and again this difference was significant, $t(93) = -14.57$, $p < .001$, and very large effect size was found

($d = 3.03$). A Mann Whitney U test revealed that the ASD scores on the BRI ($Mdn = 82.5$) were significantly higher than the non-ASD group's scores ($Mdn = 48$), $U = 27$, $z = -7.66$, $p < .001$, and a large effect size was found ($r = -.79$).

2.4.3 The relationship between EF deficits and PDA traits

Before further analysis within the ASD group was performed, exploration of the relationships between EF and PDA traits using Pearson's correlations was conducted to assess inter-variable relationships. Table 2.2 shows that the GEC, the BRI and the MI all correlated significantly with all measures of PDA traits. The weakest significance levels were found between measures of EF and emotion dysregulation. Age and gender were not found to correlate significantly with any variables and so were not considered to be confounding variables. The CAST was found to significantly correlate with all measures of EF and PDA traits, apart from emotion dysregulation. Therefore, it was decided that the potential confounding impact of the CAST on the relationship between EF and PDA traits should be considered in further analysis.

Table 2.2*Pearson's correlation coefficients for EF, PDA traits, demographic variables and ASD traits*

	Executive Function			PDA Traits					Demographics		ASD Traits
	1	2	3	4	5	6	7	8	9	10	12
<i>Executive Function</i>											
1. Global Executive Composite		.778**	.877**	.636**	.328*	.598**	.624**	.599**	-.161	-.019	.465**
2. Behaviour Regulation Index			.546**	.544**	.269*	.506**	.577**	.374**	-.165	-.093	.436**
3. Metacognition index				.479**	.268*	.486**	.467**	.498**	-.159	-.010	.405**
<i>PDA-relevant Traits</i>											
4. EDA-Q Total					.298*	.511**	.626**	.531**	.002	-.064	.450**
5. Affect Regulation						.336*	.401**	.378**	-.046	-.082	.170
6. HSQ-ASD Total							.862**	.816**	-.036	-.094	.456**
7. Social Inflexibility								.753**	-.050	-.142	.525**
8. Demand Specific									.016	-.005	.299*
<i>Demographics</i>											
9. Age										.001	-.069
10. Gender											-.261*
<i>ASD Traits</i>											
12. CAST											

* $p < .05$ ** $p < .01$ *** $p < .001$

2.4.4 Do deficits in EF predict symptoms of PDA in children with ASD?

A regression analysis was performed to assess whether the BRI and the MI predict symptoms of PDA and to test whether they are independent predictors. The overall model was significant, $F(2,61) = 15.93, p < .001$, Adjusted R Square = .32 (see Table 2.3). The model shows that both BRI and MI were significant predictors of the EDA-Q, with BRI explaining more of the variance and having a smaller confidence interval. The regression suggests that for every one-point increase in the BRI there is an associated .65 increase in the EDA-Q. Also, for every one-point increase in the MI there is an associated .53 increase in the EDA-Q. The final model explained 34% of the variance in the EDA-Q.

Table 2.3

Summary of regression assessing the contribution of the BRI and MI to the EDA-Q

		B	SEB	β	<i>t</i>	<i>p</i>	95% Confidence Intervals for B	
							Lower Bound	Upper Bound
Model 1	Constant	-41.83	16.99		-2.46	.02	-75.79	-7.86
	BRI	.65	.20	.40**	3.25	.00	.25	1.04
	MI	.53	.25	.26*	2.09	.04	.02	1.04

Note: B, Beta; SEB, Standardised Error Beta; β , Standardised Betas.

* $p < .05$ ** $p < .01$ *** $p < .001$

Next, the potential role of ASD traits as a confounder in the relationship between EF deficits and PDA traits was investigated. A regression was performed, and the CAST was entered into the model first to control for the variance accounted for by ASD traits severity, followed by the BRI and the MI. The overall model was significant, $F(3,60) = 12.72, p < .001$, Adjusted R Square = .35 (see Table 2.4). The model shows that, after controlling for the CAST, the BRI was the only significant predictor of the EDA-Q. After controlling for the variance explained by ASD traits,

for every one-point increase on the BRI there is an associated .54 increase in the EDA-Q. The final model explained 38% of the variance.

Table 2.4

Summary of regression assessing the contribution of the BRI and MI to the variance in the EDA-Q, controlling for the CAST

		B	SEB	β	<i>t</i>	<i>p</i>	95% Confidence Intervals for B	
							Lower Bound	Upper Bound
Model 2	Constant	-38.28	16.73		-2.29	.03	-71.76	-4.80
	CAST	.65	.34	.22	1.90	.06	-.04	1.33
	BRI	.54	.20	.34*	2.65	.01	.13	.94
	MI	.42	.26	.21	1.66	.10	-.09	.93

Note: B, Beta; SEB, Standardised Error Beta; β , Standardised Betas.

* $p < .05$ ** $p < .01$ *** $p < .001$

2.4.5 Do deficits in EF predict difficulties with emotion dysregulation in children with ASD?

A regression analysis was performed to assess whether the BRI and the MI predict emotion dysregulation and to test whether they are independent predictors. The overall model was not significant, $F(2,54) = 2.80$, $p = .070$, Adjusted R Square = .06 (see Table 2.5). The model shows that neither BRI nor MI were significant predictors of the ARI. The overall model explained 9% of the variance. Further analysis with ASD traits was not performed as the CAST was not found to correlate significantly with the ARI.

Table 2.5

Summary of regression assessing the contribution of the BRI and MI to the variance in the ARI

		95% Confidence Intervals for B						
		B	SEB	β	t	p	Lower Bound	Upper Bound
Model 1	Constant	-4.68	5.23		-.90	.38	-15.17	5.80
	BRI	.07	.06	.18	1.14	.26	-.05	.19
	MI	.09	.08	.17	1.13	.26	-.07	.24

Note: B, Beta; SEB, Standardised Error Beta; β , Standardised Betas.

* $p < .05$ ** $p < .01$ *** $p < .001$

2.4.6 Do deficits in ER predict difficulties with non-compliance in children with ASD?

2.4.6.1 Regression of the HSQ-DS and EF indices

A regression analysis was performed to assess whether the BRI and the MI predict demand specific non-compliance and to test whether they are independent predictors. The overall model was significant, $F(2,60) = 10.61$, $p < .001$, Adjusted R square = .24 (see Table 2.6). The model shows that only the MI was a significant predictor of the HSQ-DS. Therefore, the regression suggests that for every one-point increase in the MI there is an associated .13 increase in the HSQ-DS. The final model explained 26% of the variance.

Table 2.6

Summary of regression assessing the contribution of the BRI and MI to the variance in the HSQ-DS

		95% Confidence Intervals for B						
		B	SEB	β	t	p	Lower Bound	Upper Bound
Model 1	Constant	-6.86	2.64		-2.60	.01	-12.14	-1.58
	BRI	.03	.03	.14	1.04	.30	-.03	.10
	MI	.13	.04	.42**	3.14	.00	.05	.21

Note: B, Beta; SEB, Standardised Error Beta; β , Standardised Betas.

* $p < .05$ ** $p < .01$ *** $p < .001$

Next, the potential role of ASD traits as a confounder in the relationship between EF deficits and demand specific non-compliance was investigated. A regression was performed, and the CAST was entered into the model first to control for the variance accounted for by ASD trait severity, followed by the BRI and the MI. The overall model was significant, $F(3,59) = 7.19, p < .001$, Adjusted R Square = .23 (see Table 2.7). The model shows that, after controlling for the CAST, the MI was still the only significant predictor of the HSQ-DS. Therefore, the regression suggests that, after controlling for the variance explained by ASD traits, for every one-point increase on the MI there is an associated .12 increase in the HSQ-DS. The final model explained 27% of the variance.

Table 2.7

Summary of regression assessing the contribution of the BRI and MI to the variance in HSQ-DS, controlling for the CAST

		B	SEB	β	<i>t</i>	<i>p</i>	95% Confidence Intervals for B	
							Lower Bound	Upper Bound
Model 2	Constant	-6.65	2.67		-2.49	.02	-11.99	-1.31
	CAST	.04	.06	.09	.70	.49	-.07	.15
	BRI	.03	.03	.11	.78	.44	-.04	.09
	MI	.12	.04	.40**	2.92	.01	.04	.20

Note: B, Beta; SEB, Standardised Error Beta; β , Standardised Betas.

* $p < .05$ ** $p < .01$ *** $p < .001$

2.4.6.2 Regression of the HSQ-SI and EF indices

A regression analysis was performed in order to assess whether the BRI or the MI predict non-compliance associated with social inflexibility, and to test whether they are independent predictors. The overall model was significant, $F(2,61) = 17.59$, $p < .001$, Adjusted R square = .35 (see Table 2.8). The BRI was the only index of EF dysfunction that was found to be a significant predictor of the HSQ-SI, with a one-point increase in the BRI being associated with a .10 increase in the HSQ-SI. The overall model explained 37% of the variance.

Table 2.8

Summary of regression assessing the contribution of the BRI and MI to the variance in the HSQ-SI

		B	SEB	β	<i>t</i>	<i>p</i>	95% Confidence Intervals for B	
							Lower Bound	Upper Bound
Model 1	Constant	-6.86	2.19		-3.13	.00	-11.24	-2.48
	BRI	.10	.03	.46***	3.77	.00	.05	.15
	MI	.06	.03	.22	1.77	.08	-.01	.12

Note: B, Beta; SEB, Standardised Error Beta; β , Standardised Betas.

* $p < .05$ ** $p < .01$ *** $p < .001$

Next, the potential role of ASD traits as a confounder in the relationship between EF deficits and socially-inflexible non-compliance was investigated. A regression was performed, and the CAST was entered into the model first to control for the variance accounted for by ASD traits severity, followed by the BRI and the MI. The overall model was significant, $F(3,60) = 15.66, p < .001$, Adjusted R Square = .41 (see Table 2.9). The model shows that, after controlling for the CAST, the BRI and the CAST are both significant predictors of the HSQ-SI. This suggests that for every one-point increase on the CAST, there is a .12 increase in the HSQ-SI and for every one-point increase in the BRI there is an associated .08 rise in the HSQ-SI. The final model explained 44% of the variance.

Table 2.9

Summary of regression assessing the contribution of the BRI and MI to the variance in HSQ-SI, controlling for the CAST

		B	SEB	β	t	p	95% Confidence Intervals for B	
							Lower Bound	Upper Bound
Model 2	Constant	-6.21	2.10		-2.97	.00	-10.34	-2.03
	CAST	.12	.04	.31**	2.80	.01	.03	.20
	BRI	.08	.03	.37**	3.03	.00	.02	.13
	MI	.04	.03	.14	1.21	.23	-.03	.10

Note: B, Beta; SEB, Standardised Error Beta; β , Standardised Betas.

* $p < .05$ ** $p < .01$ *** $p < .001$

2.5 Discussion

This study is the first to use a standardised measure of behaviours associated with everyday EF impairments to investigate the relationship between EF and features associated with PDA in children with ASD. It provides evidence of a relationship between parent-reported everyday behavioural deficits in EF and PDA traits in children with ASD. The results regarding differences between groups and relationships between parent-reported EF and PDA traits will be discussed. The study's limitations will be described, and their impact on the findings considered. Implications in relation to clinical work and future research will be highlighted and conclusions on the study as a whole will be made.

2.5.1 Executive Function in children with and without ASD

Although recent meta-analyses provide evidence of overall EF impairments in children with ASD, this has not been confirmed in a sample of children with ASD, not recruited from a clinic, who display PDA features. Therefore, analysis was conducted to compare parent-reported EF impairments between children with and without ASD.

Overall, this study found evidence of significant group differences in EF with large effects. The ASD group showed higher levels of deficits in everyday EF than the non-ASD group. These results support previous findings of deficits in EF in ASD populations (Demetriou et al., 2018; Hill, 2004b; Kenworthy et al., 2008; Lai et al., 2017) and highlight the need for support with everyday tasks and situations requiring EF abilities.

A recent study investigated the differences between parent-reported BRIEF scores for children with ASD compared to TD children (Leung et al., 2016). We report higher average scores on both the BRI and MI compared to the ASD group in this study, but similar scores in our non-ASD group compared to the TD group in their study. Given that our study sought to recruit individuals with ASD who displayed PDA traits, this could suggest that a sample of children with ASD and PDA traits may struggle more with EF than a more generally representative sample of ASD children. It is not known what impact having an ASD sample with generally higher EF impairments will have on the results of this study. However, one might expect there to be more general impairment across behaviours associated with EF impairment.

2.5.2 The relationship between EF and PDA traits

The Extreme Demand Avoidance Questionnaire (EDA-Q; O’Nions, Christie, et al., 2014) was used as a global measure of PDA traits in children with ASD. Both indices of EF significantly predicted PDA traits, with the BRI being a larger predictor. However, it must be recognised that this was a cross-sectional correlational finding, therefore no inferences of causality can be made. Overall, these findings indicate that parent-reported EF impairments may contribute to some of the behavioural difficulties seen in children with ASD and PDA traits. This is an important finding as it provides

the ground work for future research to explore the potential cognitive underpinnings of behaviours displayed by children with ASD and PDA.

The role of ASD traits as potential confounders to the relationship between EF deficits and PDA traits was investigated. Interestingly, after controlling for ASD traits, only the BRI was still a significant predictor of PDA traits. This suggests that variance related to MI may impact both ASD and PDA traits, whereas the BRI may differentially impact the EDA-Q. Therefore, when considering PDA traits specifically, deficits in inhibition of action and thought, a lack of flexibility around change or problem solving, and difficulties with emotional response regulation, may be important areas of cognition for intervention strategies (Roth et al., 2014). By including ASD traits the model explained a further 4% of the variance. This could suggest that ASD severity has a small but significant role in the severity of PDA traits.

2.5.3 The relationship between EF and emotion dysregulation

Lability of mood and emotional symptoms are considered key characteristics of the PDA profile (Newson et al., 2003; O’Nions, Viding, et al., 2014). Based on previous evidence of strong relationships between EF and the ability to regulate emotions in TD young people it was hypothesised that EF deficits may play an integral role in emotional lability (Carlson & Wang, 2007; Gyurak et al., 2012; Zelazo & Cunningham, 2007). All indices of EF correlated significantly with the ARI. However, these were the weakest correlations found across the whole study, and neither indices were significant predictors of the ARI. It was expected that indices of EF would contribute to a larger proportion of the variance in the ARI. In particular, the finding of no significant relationship between the BRI and the ARI seems surprising as the BRI includes the ‘Emotional Control’ subscale which measures the ability to modulate

emotional responses, with poor emotional control being described as emotional lability or explosiveness (Roth et al., 2014).

There are a wide range of methods for measuring emotion dysregulation in ASD (Weiss et al., 2014) which makes it a complicated construct to assess. The ARI (Stringaris et al., 2012) was chosen for this study because it has been validated in an ASD population, and it is a brief scale with a parent-report version making it easy to complete and distribute. However, it may be that the questions in the ARI do not tap into the same ‘lability of mood’ that was observed by Newson et al. (2003). The ARI focuses on anger/irritability, which is one manifestation of emotion dysregulation. It could be that emotion dysregulation in ASD is also reflected in other behaviours such as avoidance, freezing or escape behaviour. The definition of what is and is not considered to fall under the umbrella of emotional lability or regulation is still under debate and the need for clearer boundaries of the construct has been raised (Compas et al., 2017). Future studies may want to consider this potential limitation when assessing the role of EF in ‘lability of mood’ in PDA.

2.5.4 The relationship between EF and demand specific non-compliance

The HSQ-DS measures an individual’s ability to cope with direct demands that require effort, such as washing or cleaning. The two indices of the BRIEF explained 26% of the variance in the HSQ-DS. However, the MI was the only significant predictor of the HSQ-DS, even after controlling for ASD traits. This finding suggests that there could be a fairly unique relationship between MI functions, such as organisation and planning, working memory and initiation behaviour, and the ability to cope with demands requiring effort and a switch from pleasant to less pleasant tasks. As previously noted, Brewer et al. (2014) described similar demands in the context of transitions since they often involve transitioning from one activity to another.

Kenworthy et al. (2014) recently conducted a randomised control trial to investigate the effectiveness of an EF intervention for children with ASD. Interestingly, those who received the intervention made greater improvement in their ability to follow rules and make transitions. Part of this intervention focussed specifically on improving planning abilities, an EF skill that falls within the MI of the BRIEF. This could suggest, that planning specifically, is an important cognitive skill necessary for complying with demands involving transitions. Therefore, similar interventions that focus on EF could be useful for children with ASD who suffer with difficulties related to PDA and non-compliance.

Pugliese et al. (2015) recently found that impairments in the MI from the BRIEF also contributed to adaptive impairments in young people with high functioning ASD. They specifically identified that working memory, the capacity to hold information in mind for the purpose of completing a task, encoding information or generating goals, is critical to carrying out multistep activities and following complex instructions. Tasks such as chores or homework, or ‘lean’ tasks (Brewer et al., 2014), often involve multiple steps and complex instructions. Therefore, it is plausible that difficulty sustaining working memory could also have significant impacts on an individual’s ability to complete these tasks or demands therefore supporting the relationship found between the MI and the HSQ-DS. Perhaps children with ASD and PDA traits struggle to cope with compliance during ‘rich to lean’ transitions due to an inability to hold and use information relevant to the more complex ‘lean’ task.

2.5.6 The relationship between EF and socially inflexible non-compliance

The HSQ-SI describes behaviours in response to demands in non-routine and potentially unpredictable contexts where there is a deviation from the expected. For

example, it asks parents whether their child has problems following instructions, commands or rules when: ‘in public places’ or ‘there is an unexpected change in daily routine’. It was hypothesised that to cope with these demands, an individual needs the capacity to tolerate uncertainty (Freeston et al., 1994), which has been shown to be related to EF (Mushtaq et al., 2011). Therefore, it was expected that EF may contribute to variance in the HSQ-SI. Together, the indices of the BRIEF accounted for 37% of the variance, more than that accounted for in the HSQ-DS. The BRI was the only significant predictor of the HSQ-SI, which remained the same after controlling for ASD traits. This indicates that the BRI predicts variance in the HSQ-SI that is unrelated to ASD trait severity. Overall, this finding suggests that skills associated with inhibition of behaviour, the ability to move freely from one situation or activity to another, and emotional control, are implicated in impairments to comply with unexpected demands in children with ASD and PDA traits.

Notably, when ASD traits were entered into the model, it explained 7% more of the variance in the HSQ-SI. This suggests that some of the variance in non-compliance behaviours specific to non-routine and unpredictable demands can be explained by ASD trait severity. Therefore, perhaps those who have more severe levels of ASD traits may struggle more with non-compliance in these situations.

Items from the HSQ-SI often involve non-routine or unexpected demands where others are also present. For instance, it asks parents whether their child has problems following instructions, commands or rules when: ‘playing with other children’ or ‘visitors are in your home’. It has been found that poorer shifting abilities, as assessed via the BRIEF, are related to lower scores on measures of socialisation skills (Pugliese et al., 2015). The shift scale on the BRIEF forms part for the BRI and assesses the ability to transition to a new situation or activity, to tolerate change, and

to problem-solve flexibly (Roth et al., 2014). Therefore, perhaps when a task requires both the ability to manage unexpected demands, and the capacity to cope with socially unexpected factors, deficits in BRI related to shifting may contribute to demand avoidance and non-compliance behaviour seen in children with ASD and PDA. This would suggest that when placing a demand or a task on a child with ASD and PDA traits, one should be aware of not only how unexpected or out of routine the task is, but also what social complexities are involved.

2.5.6 Limitations

The first limitation of this study that should be considered is the use of parent-report questionnaire to assess EF. Traditionally more direct tasks have been used to measure EF and it should be noted that the BRIEF is not a direct measure of EF. However, direct cognitive assessments have been criticised for not supporting observational information from everyday tasks (Wilson, 1993) and failing to capture the impact of EF deficits (Dawson et al., 2009). The relationship between cognitive testing and real-world performance is complex and new tests of EF are continually being developed in an attempt to improve ecological validity. The BRIEF offers data based on the potential impact of EF deficits and therefore it could be argued that impairments identified by the BRIEF may be more representative of the daily difficulties that individuals face. Overall, it has been suggested that the best way to assess EF deficits is using a multi-method approach (Bakar, Taner, Soysal, Karakas, & Turgay, 2011), however this was not possible due to research constraints. Results must be interpreted with this limitation in mind, and future work should be conducted to assess whether results differ when different methods of measurement are used.

Informant-report methodologies are open to error introduced through response-biases such as social desirability, question order, and demand characteristics.

Unfortunately, the impact of these biases cannot be wholly known or controlled for. When asking parents of children who present with challenging behaviours to accurately rate the impact, size or frequency of their child's difficulties, the influence of their rich knowledge and experience of raising their child must be considered. For instance, parents may have a heightened awareness for areas of difficulties and extreme behaviours. This could mean that certain behaviours or difficulties may be over reported, and the impact of this bias should be considered when interpreting the results. However, questionnaire methodologies are often necessary in order to develop hypotheses and pave the way for further multi-method investigations to examine whether findings hold true.

This study used a cross-sectional questionnaire methodology because this allowed for a larger sample size to be recruited, which seemed important when investigating a population about which little is still known. However, there are several limitations to this methodology. Firstly, the findings could to some extent be an artefact of the measures used and be impacted by underlying method variance. For example, we cannot say to what extent systematic error variance may be shared between the variables used in this study. This may have resulted in inflated estimates of the relationships between variables, such as EF and non-compliance behaviours. Secondly, this methodology lacks evidence from experimental or observational tasks which, if used in combination with questionnaires, may provide a more direct assessment of EF. Therefore, this method meant that no causal or developmental relationships could be investigated. Future research should aim to explore the relationships found between EF and PDA behaviours using mixed methodologies in order to reduce the impact of method variance and to explore causal relationships.

The limitations of certain sample characteristics in this study should be considered. Due to financial constraints, the sample size was limited, although it did surpass that required according to power analysis calculations. The sample age range was limited to children aged 6-11 years in an attempt to avoid introducing developmental variation but whilst also ensuring that a large enough sample could be recruited. However, more research should be conducted to explore the role of EF in older children and adults with ASD and PDA traits. It should also be noted that children were included in this study based on parent report of diagnosis. Unfortunately, due to restricted resources, no clinical assessments or data were collected about the basis of these diagnoses.

Although originally a TD comparison group was sought, our comparison group resulted in two children with mild learning difficulties. However, these participants were included as they both fell below threshold on the SDQ (Goodman, 1997) and CAST (Scott et al., 2002). Our ASD sample also included a large proportion of children with additional diagnoses. Unfortunately, we cannot say what impact these confounders may have had. However, it is well known that comorbidity in ASD population is high, especially for intellectual disabilities and ADHD (Gargaro, Rinehart, Bradshaw, Tonge, & Sheppard, 2011; Matson & Shoemaker, 2009; Pinborough-Zimmerman et al., 2007). Therefore, although the impact of comorbidities was not controlled for, the results may be more representative of the general ASD population. The level of educational need was also much higher in the ASD group than the non-ASD group, with just over 42% of the ASD sample not attending mainstream school. This was not controlled for and therefore the impact of educational ability on the results cannot be accounted for.

2.5.7 Implications of this study

2.5.7.1 Future research

This study provides justification for further investigation of the role of EF in PDA traits. It is suggested that firstly, future research should focus on using different designs, such as longitudinal or experimental, to assess EF in children with ASD and PDA traits. This would allow comparison with the results from this study, and investigation of causality and developmental hypotheses. The results of this study also suggest that there may be differential relationships between specific types of EF difficulties and non-compliance behaviours. By using more direct assessments of EF, such as neuropsychological assessment, this may enable more direct associations between specific EF difficulties and behaviours to be made and tested. It could also be beneficial to explore this connection at a strategy level. For instance, could interventions that target particular vulnerabilities, such as increasing the reinforcement value of a non-preferred activity by embedding more rewards, help as it might counter particular vulnerability pathways and compensate for EF deficits.

In agreement with previous research (O’Nions, Viding, et al., 2014), the present study highlights the need for further elucidation of the PDA phenotype and its cognitive underpinnings. For example, overall, the final model of EF and PDA traits only explained 34% of the variance. This suggests that much of the variance in PDA traits is still left unexplained. Therefore, it seems necessary that other cognitive constructs which play a role in ASD, such as Intolerance of Uncertainty (Freeston et al., 1994) and Theory of Mind (Baron-Cohen, 1991), are explored with a view to gaining a better understanding of the contributors to behavioural difficulties seen in those with ASD and PDA traits. Finally, future research should also focus on exploring potential confounds that may have caused problems in the current study, such as

comorbidities and intellectual functioning, to allow clearer hypotheses to be formed and tested.

2.5.7.2 Clinical implications

Overall, our findings suggest that parent-reported EF difficulties are related to and predict PDA traits in children with ASD. More specifically, relationships were found between EF deficits and emotional dysregulation and non-compliance. Real world interventions that target EF deficits have been shown to be successful in those with ASD (de Vries, Prins, Schmand, & Geurts, 2015; Kenworthy et al., 2014). The findings from this study suggest that these types of interventions could help support individuals with ASD and PDA traits to ensure that they can cope in environments such as school, where challenges with emotional lability and demand avoidance may have severe impacts on their ability to learn and integrate. This study has also highlighted the specific difficulty that children with ASD and PDA traits may have with transitions. This may also be useful for professionals and carers to consider when planning transitions, as techniques such as using pre-transitions cues have been found to support children with these difficulties (Brewer et al., 2014).

2.5.8 Conclusion

The general finding of the importance of the role of EF in PDA symptoms may go some way to explain the behavioural overlaps found between ASD and PDA traits and those with only ASD or even other neurodevelopmental difficulties such as conduct disorder (O’Nions, Viding, et al., 2014). For instance, it is already known that deficits in EF are common across a range of developmental disorders (Johnson, 2012). Therefore, it could be postulated that EF represents a transdiagnostic problem that may be a fruitful target for intervention.

This study contributes a novel understanding of the cognitive processes associated with PDA traits and behaviours in children with ASD. To date no other study has investigated the cognitive profile of PDA. Significant differences were found between children with and without ASD on measures of EF. Relationships were found between parent-reported EF deficits and measures of PDA traits. More specifically, deficits in EF predicted non-compliance behaviour. The MI of the BRIEF specifically predicted non-compliance associated with ‘rich to lean’ transitions, and the BRI of the BRIEF predicted non-compliance associated with non-routine and unexpected demands. This study has contributed to our still limited understanding of the behavioural and cognitive profile of PDA and also raised awareness of the difficulties experienced by families with children who resemble the ASD and PDA profile. There is still a need for further understanding of the PDA phenotype and investigation to elucidate the involvement of EF to aid intervention development.

2.6 References

- Allison, C., Williams, J., Scott, F., Stott, C., Bolton, P., Baron-Cohen, S., & Brayne, C. (2007). The Childhood Asperger Syndrome Test (CAST). *Autism, 11*(2), 173–185.
- American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders* (5th ed.). Washington, DC: Author.
- Anderson, P. (2002). Assessment and development of executive function (EF) during childhood. *Child Neuropsychology, 8*(2), 71–82.
- Anderson, P., Northam, E., Jacobs, R., Mikiewicz, O., & Anderson, V. A. (2002). Relationships between cognitive and behavioral measures of executive function in children with brain disease. *Child Neuropsychology (Neuropsychology, Development and Cognition: Section C), 8*(4), 231–240.
- Anderson, V., Jacobs, R., & Harvey, A. S. (2005). Prefrontal lesions and attentional skills in childhood. *Journal of the International Neuropsychological Society, 11*(07), 817–831.
- Arbelle, S., Sigman, M. D., & Kasari, C. (1994). Compliance with parental prohibition in autistic children. *Journal of Autism and Developmental Disorders, 24*(6), 693–702.
- Bakar, E. E., Taner, Y. I., Soysal, A. S., Karakas, S., & Turgay, A. (2011). Behavioral rating inventory and laboratory tests measure different aspects of executive functioning in boys: A validity study. *Klinik Psikofarmakoloji Bülteni-Bulletin of Clinical Psychopharmacology, 21*(4), 302–316.
- Barkley, R., & Edelbrock, C. (1987). Assessing situational variation in children's behavior problems: The home and school situations questionnaires. *Advances in Behavioral Assessment of Children and Families, 3*, 157–176.

- Baron-Cohen, S. (1991). Precursors to a theory of mind: Understanding attention in others. *Natural Theories of Mind: Evolution, Development and Simulation of Everyday Mindreading, 1*, 233–251.
- Baron-Cohen, S., Scott, F. J., Allison, C., Williams, J., Bolton, P., Matthews, F. E., & Brayne, C. (2009). Prevalence of autism-spectrum conditions: UK school-based population study. *The British Journal of Psychiatry: The Journal of Mental Science, 194*(6), 500–509.
- Baron-Cohen, S., Wheelwright, S., Cox, A., Baird, G., Charman, T., Swettenham, J., ... Doehring, P. (2000). Early identification of autism by the checklist for autism in toddlers (CHAT). *Journal of the Royal Society of Medicine, 93*(10), 521–525.
- Baron-cohen, S., Wheelwright, S., Stone, V., & Rutherford, M. (1999). A mathematician, a physicist and a computer scientist with Asperger syndrome: Performance on folk psychology and folk physics tests. *Neurocase, 5*(6), 475–483.
- Brewer, A. T., Strickland-Cohen, K., Dotson, W., & Williams, D. C. (2014). Advance notice for transition-related problem behavior: Practice guidelines. *Behavior Analysis in Practice, 7*(2), 117–125.
- Bryce, C. I., & Jahromi, L. B. (2013). Brief report: Compliance and noncompliance to parental control strategies in children with high-functioning autism and their typical peers. *Journal of Autism and Developmental Disorders, 43*(1), 236–243.
- Burgess, P. W., Alderman, N., Forbes, C., Costello, A., M-A.Coates, L., Dawson, D. R., ... Channon, S. (2006). The case for the development and use of “ecologically valid” measures of executive function in experimental and clinical neuropsychology. *Journal of the International Neuropsychological Society, 12*(02), 194–209.

- Carlson, S. M., & Wang, T. S. (2007). Inhibitory control and emotion regulation in preschool children. *Cognitive Development, 22*(4), 489–510.
- Chaytor, N., Schmitter-Edgecombe, M., & Burr, R. (2006). Improving the ecological validity of executive functioning assessment. *Archives of Clinical Neuropsychology, 21*(3), 217–227.
- Chowdhury, M., Aman, M. G., Lecavalier, L., Smith, T., Johnson, C., Swiezy, N., ... Scahill, L. (2016). Factor structure and psychometric properties of the revised home situations questionnaire for autism spectrum disorder: The home situations questionnaire-autism spectrum disorder. *Autism, 20*(5), 528–537.
- Cohen, J. (1977). Statistical power analysis for the behavioral sciences. *Statistical Power Analysis for the Behavioral Sciences*.
- Cohen, J. (1988). *Statistical Power Analysis for the Behavioral Sciences* (2nd Edition).
- Cohen, J. (1992). A power primer. *Psychological Bulletin, 112*(1), 155.
- Compas, B. E., Jaser, S. S., Bettis, A. H., Watson, K. H., Gruhn, M. A., Dunbar, J. P., ... Thigpen, J. C. (2017). Coping, emotion regulation, and psychopathology in childhood and adolescence: A meta-analysis and narrative review. *Psychological Bulletin, 143*(9), 939–991.
- Dawson, D. R., Anderson, N. D., Burgess, P., Cooper, E., Krpan, K. M., & Stuss, D. T. (2009). Further development of the multiple errands test: Standardized scoring, reliability, and ecological validity for the baycrest version. *Archives of Physical Medicine and Rehabilitation, 90*(11), S41–S51.
- de Vries, M., Prins, P. J. M., Schmand, B. A., & Geurts, H. M. (2015). Working memory and cognitive flexibility-training for children with an autism spectrum disorder: a randomized controlled trial. *Journal of Child Psychology and Psychiatry, 56*(5), 566–576.

- Demetriou, E. A., Lampit, A., Quintana, D. S., Naismith, S. L., Song, Y. J. C., Pye, J. E., ... Guastella, A. J. (2018). Autism spectrum disorders: A meta-analysis of executive function. *Molecular Psychiatry*, 23(5), 1198–1204.
- Denckla, M. (1994). Measurement of executive function.
- Eaton, J., & Banting, R. (2012). Adult diagnosis of pathological demand avoidance – subsequent care planning. *Journal of Learning Disabilities and Offending Behaviour*, 3(3), 150–157.
- Espy, K. A., Sheffield, T. D., Wiebe, S. A., Clark, C. A. C., & Moehr, M. J. (2011). Executive control and dimensions of problem behaviors in preschool children. *Journal of Child Psychology and Psychiatry*, 52(1), 33–46.
- Eylen, L. Van, Boets, B., Steyaert, J., Wagemans, J., & Noens, I. (2015). Executive functioning in autism spectrum disorders: Influence of task and sample characteristics and relation to symptom severity. *European Child & Adolescent Psychiatry*, 24(11), 1399–1417.
- Faul, F., Erdfelder, E., Lang, A.-G., & Buchner, A. (2007). G*Power 3: A flexible statistical power analysis program for the social, behavioral, and biomedical sciences. *Behavior Research Methods*, 39(2), 175–191.
- Field, A. P. (2009). *Discovering statistics using SPSS: (and sex and drugs and rock “n” roll)*. SAGE Publications.
- Freeston, M., Rhéaume, J., Letarte, H., Dugas, M., & Ladouceur, R. (1994). Why do people worry? *Personality and Individual Differences*, 17(6), 791–802.
- Gargaro, B. A., Rinehart, N. J., Bradshaw, J. L., Tonge, B. J., & Sheppard, D. M. (2011). Autism and ADHD: How far have we come in the comorbidity debate? *Neuroscience & Biobehavioral Reviews*, 35(5), 1081–1088.
- Geurts, H. M., Corbett, B., & Solomon, M. (2009). The paradox of cognitive flexibility

- in autism. *Trends in Cognitive Sciences*, 13(2), 74–82.
- Gioia, G. A., Isquith, P. K., & Guy, S. C. (1998). The regulatory role of executive control processes in children's behavioral, social, and emotional functioning. *Journal of Neuropsychiatry and Clinical Neurosciences*, 9(663).
- Gioia, G. A., Isquith, P., Guy, S., & Kenworthy, L. (2001). Brief rating inventory of executive function (BRIEF). *Odessa, FL: Psychological Assessment Resources*.
- Gioia, G. A., Isquith, P. K., Guy, S. C., & Kenworthy, L. (2000). Test review behavior rating inventory of executive function. *Child Neuropsychology (Neuropsychology, Development and Cognition: Section C)*, 6(3), 235–238.
- Gioia, G. A., Isquith, P. K., Retzlaff, P. D., & Espy, K. A. (2002). Confirmatory factor analysis of the behavior rating inventory of executive function (BRIEF) in a Clinical Sample. *Child Neuropsychology (Neuropsychology, Development and Cognition: Section C)*, 8(4), 249–257.
- Goldstein, S., Naglieri, J. A., Princiotta, D., & Otero, T. M. (2014). Introduction: A history of executive functioning as a theoretical and clinical construct. In *Handbook of Executive Functioning* (pp. 3–12). New York, NY: Springer New York.
- Goodman, R. (1997). The strengths and difficulties questionnaire: A research note. *Journal of Child Psychology and Psychiatry*, 38(5), 581–586.
- Goodman, R. (1999). The extended version of the strengths and difficulties questionnaire as a guide to child psychiatric caseness and consequent burden. *Journal of Child Psychology and Psychia*, 40(05), 791–799.
- Goodman, R. (2001). Psychometric properties of the strengths and difficulties questionnaire. *Journal of the American Academy of Child and Adolescent Psychiatry*, 40(11), 1337–1345.

- Goodman, R., & Scott, S. (1999). Comparing the strengths and difficulties questionnaire and the child behavior checklist: Is small beautiful? *Journal of Abnormal Child Psychology*, 27(1), 17–24.
- Gore Langton, E., & Frederickson, N. (2016). Mapping the educational experiences of children with pathological demand avoidance. *Journal of Research in Special Educational Needs*, 16(4), 254–263.
- Green, J., Absoud, M., Grahame, V., Malik, O., Simonoff, E., Le Couteur, A., & Baird, G. (2018). Pathological demand avoidance: Symptoms but not a syndrome. *The Lancet Child & Adolescent Health*, 0(0).
- Gross, J. J., & Thompson, R. A. (2007). Emotion regulation: Conceptual foundations. In J. J. Gross (Ed.), *Handbook of emotion regulation* (pp. 3–24). New York: Guilford Press.
- Gyurak, A., Goodkind, M. S., Kramer, J. H., Miller, B. L., & Levenson, R. W. (2012). Executive functions and the down-regulation and up-regulation of emotion. *Cognition & Emotion*, 26(1), 103–118.
- Hill, E. L. (2004a). Evaluating the theory of executive dysfunction in autism. *Developmental Review*, 24(2), 189–233.
- Hill, E. L. (2004b). Executive dysfunction in autism. *Trends in Cognitive Sciences*, 8(1), 26–32.
- Hill, E. L., & Russell, J. (2002). Action memory and self-monitoring in children with autism: self versus other. *Infant and Child Development*, 11(2), 159–170.
- Hughes, C., & Ensor, R. (2008). Does executive function matter for preschoolers' problem behaviors? *Journal of Abnormal Child Psychology*, 36(1), 1–14.
- Johnson, M. H. (2012). Executive function and developmental disorders: the flip side of the coin. *Trends in Cognitive Sciences*, 16(9), 454–457.

- Jurado, M. B., & Rosselli, M. (2007). The elusive nature of executive functions: A review of our current understanding. *Neuropsychology Review*, *17*(3), 213–233.
- Kenworthy, L., Anthony, L. G., Naiman, D. Q., Cannon, L., Wills, M. C., Luong-Tran, C., ... Wallace, G. L. (2014). Randomized controlled effectiveness trial of executive function intervention for children on the autism spectrum. *Journal of Child Psychology and Psychiatry*, *55*(4), 374–383.
- Kenworthy, L., Yerys, B. E., Anthony, L. G., & Wallace, G. L. (2008). Understanding executive control in autism spectrum disorders in the lab and in the real world. *Neuropsychology Review*, *18*(4), 320–338.
- Kim, H. (2013). Statistical notes for clinical researchers: assessing normal distribution (2) using skewness and kurtosis. *Restorative Dentistry & Endodontics*, *38*(1), 52.
- King, M., & Bearman, P. (2009). Diagnostic change and the increased prevalence of autism. *International Journal of Epidemiology*, *38*(5), 1224–1234.
- Kuhn, C. (2015). Emergence of sex differences in the development of substance use and abuse during adolescence. *Pharmacology & Therapeutics*, *153*, 55–78.
- Lai, C., Lau, Z., Lui, S., Lok, E., Tam, V., Chan, Q., ... & Cheung, E. F. (2017). Meta-analysis of neuropsychological measures of executive functioning in children and adolescents with high-functioning autism spectrum disorder. *Autism Research*, *10*(5), 911-939.
- Lemanek, K. L., Stone, W. L., & Fishel, P. T. (1993). Parent-child interactions in handicapped preschoolers: The relation between parent behaviors and compliance. *Journal of Clinical Child Psychology*, *22*(1), 68–77.
- Leung, R. C., Vogan, V. M., Powell, T. L., Anagnostou, E., & Taylor, M. J. (2016). The role of executive functions in social impairment in autism spectrum disorder. *Child Neuropsychology*, *22*(3), 336–344.

- Loomes, R., Hull, L., & Mandy, W. P. L. (2017). What is the male-to-female ratio in autism spectrum disorder? A systematic review and meta-analysis. *Journal of the American Academy of Child and Adolescent Psychiatry*, *56*(6), 466–474.
- Matson, J. L., & Shoemaker, M. (2009). Intellectual disability and its relationship to autism spectrum disorders. *Research in Developmental Disabilities*, *30*(6), 1107–1114.
- Mazefsky, C. A., Herrington, J., Siegel, M., Scarpa, A., Maddox, B. B., Scahill, L., & White, S. W. (2013). The role of emotion regulation in autism spectrum disorder. *Journal of the American Academy of Child and Adolescent Psychiatry*, *52*(7), 679–688.
- Minshew, N. J., Muenz, L. R., Goldstein, G., & Payton, J. B. (1992). Neuropsychological functioning in nonmentally retarded autistic individuals. *Journal of Clinical and Experimental Neuropsychology*, *14*(5), 749–761.
- Mostert-Kerckhoffs, M. A. L., Staal, W. G., Houben, R. H., & de Jonge, M. V. (2015). Stop and change: Inhibition and flexibility skills are related to repetitive behavior in children and young adults with autism spectrum disorders. *Journal of Autism and Developmental Disorders*, *45*(10), 3148–3158.
- Mushtaq, F., Bland, A. R., & Schaefer, A. (2011). Uncertainty and cognitive control. *Frontiers in Psychology*, *2*, 249.
- Newson, E., Le Maréchal, K., & David, C. (2003). Pathological demand avoidance syndrome: a necessary distinction within the pervasive developmental disorders. *Archives of Disease in Childhood*, *88*(7), 595–600.
- Nydén, A., Hagberg, B., Goussé, V., & Rastam, M. (2011). A cognitive endophenotype of autism in families with multiple incidence. *Research in Autism Spectrum Disorders*, *5*(1), 191–200.

- O’Nions, E., Christie, P., Gould, J., Viding, E., & Happé, F. (2014). Development of the ‘extreme demand avoidance questionnaire’ (EDA-Q): Preliminary observations on a trait measure for Pathological demand avoidance. *Journal of Child Psychology and Psychiatry*, *55*(7), 758–768.
- O’Nions, E., Gould, J., Christie, P., Gillberg, C., Viding, E., & Happé, F. (2016). Identifying features of ‘pathological demand avoidance’ using the diagnostic interview for social and communication disorders (DISCO). *European Child & Adolescent Psychiatry*, *25*(4), 407–419.
- O’Nions, E., Viding, E., Greven, C. U., Ronald, A., & Happé, F. (2014). Pathological demand avoidance: Exploring the behavioural profile. *Autism*, *18*(5), 538–544.
- Ostfeld-Etzion, S., Feldman, R., Hirschler-Guttenberg, Y., Laor, N., & Golan, O. (2016). Self-regulated compliance in preschoolers with autism spectrum disorder: The role of temperament and parental disciplinary style. *Autism*, *20*(7), 868–878.
- Pinborough-Zimmerman, J., Satterfield, R., Miller, J., Bilder, D., Hossain, S., & McMahon, W. (2007). Communication disorders: Prevalence and comorbid intellectual disability, autism, and emotional/behavioral disorders. *American Journal of Speech-Language Pathology*, *16*(4), 359.
- Pugliese, C. E., Anthony, L., Strang, J. F., Dudley, K., Wallace, G. L., & Kenworthy, L. (2015). Increasing adaptive behavior skill deficits from childhood to adolescence in autism spectrum disorder: role of executive function. *Journal of Autism and Developmental Disorders*, *45*(6), 1579–1587.
- Roth, R. M., Isquith, P. K., & Gioia, G. A. (2014). Assessment of executive functioning using the behavior rating inventory of executive function (BRIEF). In *Handbook of Executive Functioning* (pp. 301–331). New York, NY: Springer New York.

- Russell, J., & Hill, E. L. (2001). Action-monitoring and intention reporting in children with autism. *Journal of Child Psychology and Psychiatry and Allied Disciplines*, 42(3), 317-328.
- Scott, F. J., Baron-Cohen, S., Bolton, P., & Brayne, C. (2002). The CAST (Childhood Asperger Syndrome Test). *Autism*, 6(1), 9–31.
- Stringaris, A., Goodman, R., Ferdinando, S., Razdan, V., Muhrer, E., Leibenluft, E., & Brotman, M. A. (2012). The affective reactivity index: A concise irritability scale for clinical and research settings. *Journal of Child Psychology and Psychiatry*, 53(11), 1109–1117.
- Stuss, D. T., & Knight, R. T. (2002). *Principles of frontal lobe function*. Oxford University Press.
- Thompson, R. A. (2008). Emotion regulation: a theme in search of definition. *Monographs of the Society for Research in Child Development*, 59(2–3), 25–52.
- Toplak, M. E., Bucciarelli, S. M., Jain, U., & Tannock, R. (2008). Executive functions: Performance-based measures and the behavior rating inventory of executive function (BRIEF) in adolescents with attention deficit/hyperactivity disorder (ADHD). *Child Neuropsychology*, 15(1), 53–72.
- Towgood, K. J., Meuwese, J. D. I., Gilbert, S. J., Turner, M. S., & Burgess, P. W. (2009). Advantages of the multiple case series approach to the study of cognitive deficits in autism spectrum disorder. *Neuropsychologia*, 47(13), 2981–2988.
- Vriezen, E. R., & Pigott, S. E. (2002). The relationship between parental report on the BRIEF and performance-based measures of executive function in children with moderate to severe traumatic brain injury. *Child Neuropsychology (Neuropsychology, Development and Cognition: Section C)*, 8(4), 296–303.
- Weiss, J. A., Thomson, K., & Chan, L. (2014). A systematic literature review of

- emotion regulation measurement in individuals with autism spectrum disorder. *Autism Research*, 7(6), 629–648.
- Williams, J., Scott, F., Stott, C., Allison, C., Bolton, P., Baron-Cohen, S., & Brayne, C. (2005). The CAST (Childhood Asperger Syndrome Test) test accuracy. *Autism*, 9(1), 45–68.
- Wilson, B. A. (1993). Ecological validity of neuropsychological assessment: Do neuropsychological indexes predict performance in everyday activities? *Applied and Preventive Psychology*, 2(4), 209–215.
- Wing, L. (1991). The relationship between Asperger's syndrome and Kanner's autism. *Autism and Asperger Syndrome*, 93–121.
- Wing, L., Leekam, S. R., Libby, S. J., Gould, J., & Larcombe, M. (2002). The diagnostic interview for social and communication disorders: Background, inter-rater reliability and clinical use. *Journal of Child Psychology and Psychiatry*, 43(3), 307–325.
- Wing, L., & Potter, D. (2002). The epidemiology of autistic spectrum disorders: is the prevalence rising? *Mental Retardation and Developmental Disabilities Research Reviews*, 8(3), 151–161.
- Wykes, T., Huddy, V., Cellard, C., McGurk, S. R., & Czobor, P. (2011). A Meta-Analysis of cognitive remediation for schizophrenia: Methodology and effect sizes. *American Journal of Psychiatry*, 168(5), 472–485.
- Ye, A. X., AuCoin-Power, M., Taylor, M. J., & Doesburg, S. M. (2015). Disconnected neuromagnetic networks in children born very preterm: Disconnected MEG networks in preterm children. *NeuroImage: Clinical*, 9, 376–384.
- Zelazo, P., & Cunningham, W. (2007). Executive function: Mechanisms underlying emotion regulation.

Part 3: Critical Appraisal

3.1 Introduction

The following appraisal reflects the insights gained during the process of undertaking the presented literature review and empirical study. First, I reflect on the process of conducting a literature review and describe the motivation behind selecting the topic. The difficulties inherent in studying psychological constructs, such as Emotion Regulation (ER), are described, as well as the challenges encountered along the way. Next, my observations on the process of conducting the empirical study are discussed, with a particular focus on exploring the practical and personal challenges encountered with recruitment of participants, the use of questionnaires and measurement selection, and my general experience of joint working. Finally, the appraisal closes with a discussion about the clinical and scientific implications of the empirical study, and a conclusion on the appraisal as a whole.

3.2 Literature Review

3.2.1 Why ER in Autism Spectrum Disorders (ASD)?

There has been growing interest in understanding the potential involvement and resulting impact of ER in ASD (Mazefsky et al., 2013; Mazefsky & White, 2014). When searching the literature, it became apparent that attempts had been made to summarise the findings of ER research in ASD, but there appeared to be a lack of systematic syntheses. A recent systematic review had looked specifically at the measurement of ER in ASD (Weiss et al., 2014) but this did not present the findings of these studies with respect to the type and level of ER difficulties in ASD. I felt that a coherent narrative or understanding of the extent of potential ER difficulties in young people with ASD was missing. From my initial reading around ER and ASD I started to learn more about the difficulties that deficits in ER could contribute to, for example

tantrums, distressing outbursts, aggression and self-harm, all of which could mistakenly be interpreted as defiant or deliberate behaviour (Mazefsky et al., 2013; Mazefsky, Pelphrey, & Dahl, 2012).

Before and during my Clinical Psychology training I have been lucky enough to work with both adults and children affected by ASD. Through these experiences I have gained insight into some of the challenges that having ASD can cause for both the individual and their family or support network. Psychiatric comorbidity is high in ASD and it has been suggested that these comorbidities could be underpinned by more fundamental difficulties with ER which may form part of ASD itself (Mazefsky et al., 2013; Mazefsky, Oswald, et al., 2012; Weiss et al., 2018). The idea that ER may underpin some of the difficulties that affect individuals with ASD resonated with me and my experiences of supporting those who struggles with difficulties such as adapting to environments and managing their emotions.

Interestingly, evidence has been found supporting the use of interventions that target ER difficulties in individuals with ASD (Scarpa & Reyes, 2011; Thomson et al., 2015). By specifically targeting ER, a reduction in emotional lability, outbursts, behavioural regulation and an increase in adaptive behaviour has been reported. These findings seemed encouraging and sparked my motivation and interest in exploring this field further. I considered that by contributing to the understanding of ER deficits in ASD I could make a real impact in terms of supporting further intervention developments.

3.2.2 Emotion Regulation: a construct under debate

Controversy regarding construct validity and definition is an issue that is quite common within the field of psychological research. Emotion Regulation has been researched across a diverse range of studies, some comparing individual differences,

others looking at ER specifically as a trait, and some viewing it as a transitory state. It has also been investigated across a range of contexts, from intimate relationships through to public behaviour, and across ages (Cole et al., 2004). However, the broad application of this construct to such a range of phenomena has caused debates over its definition, conceptual boundaries and utility as a construct (Cicchetti, Ackerman, & Izard, 1995; Eisenberg & Spinrad, 2004; Gross, 1998). Therefore, one of the first challenges I experienced when researching ER was the range of models, definitions (Eisenberg & Spinrad, 2004; Gratz & Roemer, 2004; Gross, J & Thompson, R, 2007) and methods of measurement (Weiss et al., 2014).

Initially, I found the controversies, debates and range of theories surrounding ER quite overwhelming and I struggled to decide which model, definition or method of measurement to base my review and searches upon. I later came to realise that these early decisions would ultimately have a large impact on the review process and findings. I concluded, that perhaps the most useful or meaningful way to explore ER is to consider it as an ‘umbrella term’ that actually captures a wide range of cognitive abilities and behaviours. However, when reflecting on this decision, I wondered if I should have paid more attention to the debates and controversy surrounding this construct. For instance, an alternative approach that I could have taken would have been to focus on one area or domain of ER instead of trying to capture the breadth of ER. However, due to the inconsistency in the literature and a lack of evidence supporting specific behaviours or measures related to ER deficits in ASD, I felt that a broad approach was a good starting point, allowing me to include more studies in the review and provide a base for others to build upon.

3.2.3 Barriers and challenges

Throughout the review process I encountered several challenges which threatened the validity of the review. The first obstacle I faced was related to the earlier discussion of the difficulties surrounding ER as a construct and measurement. Meta-analyses are often considered the ‘gold standard’ of literature reviews. However, recent discussions have questioned this due to the need for subjective decisions to be made along the way, which can influence the outcome (Stegenga, 2011). I had initially hoped that I would be able to conduct a meta-analysis as, to the best of my knowledge, there are currently no meta-analyses that specifically address the questions of whether there are ER impairments in young people with ASD. I thought a meta-analysis would be a valuable contribution to the field and could provide more robust evidence for future research to build upon. However, during the process of conducting searches, defining search terms and making decisions on measurement inclusion, it became apparent that I would have to abandon the meta-analysis goal in order to avoid “mixing apples and oranges” (Borenstein, Hedges, Higgins, & Rothstein, 2009). I found that there was little consistent ER measurement within the ASD literature. This resulted in a wide range of measures of ER being included in the review, meaning that comparing them using meta-analyses methods could have resulted in invalid and uninterpretable results. After discussing this dilemma with my supervisor, it was decided that I would present effect sizes from each study in the review, to aid comparison and interpretation of the effects, and to provide the reader with as much evidence as possible to guide future research decisions. This felt like a good compromise between ensuring the risk of making invalid interpretations and comparisons was reduced whilst providing the reader with as much information as possible.

Another initial aim of the review was to build on previous investigations into the measurement of ER domains (Weiss et al., 2014) by exploring whether there were any patterns of deficits within or across these domains. For instance, we aimed to answer questions such as were there more deficits in one domain compared to another. However, with hindsight it may have been naive to expect that I could disentangle the domains of ER from one another. As the review progressed it became clearer that it would not be possible to isolate one particular domain. This was partly due to the fact that nearly all measures included in the review assessed at least two ER domains. There was also little consistency between measures and which combinations of domains they tapped into. I was disappointed that I would not be able to provide more evidence of specific areas of ER deficit in individuals with ASD. However, I think this finding did highlight the real need for more consistent ER measurement in ASD and also greater consistency in the understanding of what is meant by ER.

Throughout this process I have learnt the need for flexibility when conducting reviews. I found the process challenging and frustrating at times. However, when reflecting on the struggles and dilemmas, I noticed some similarities between conducting the review and clinical work. For instance, when working with clients, I have learnt the importance of remaining flexible, continuing to be questioning and curious, and being adaptable to unforeseen challenges. All of these skills have been required at one time or another during the review process.

3.2.4 Implications

Overall, the review is the first to provide systematic evidence of parent- and self-report ER difficulties in children and young people with ASD compared to typically developing (TD) children and young people. This should hopefully continue to motivate research in this field. This review also highlights the complexities of trying

to establish a more precise understanding of ER difficulties in the ASD population. These challenges mainly appear to be due to the wide range of behaviours, and cognitive processes that are considered to fall under the ER umbrella. Future work is needed to help disentangle and define ER as a construct. This would make it more possible to investigate potential relationships between ER deficits and resulting behavioural disturbances, which could ultimately guide intervention development.

3.3 Empirical paper

3.3.1 Why EF in Pathological Demand Avoidance (PDA)?

Pathological Demand Avoidance is a term that is currently used to describe a group of extreme behavioural challenges observed by a subset of children with ASD (Newson et al., 2003; O’Nions, Christie, et al., 2014). Central to these is an obsessive resistance of everyday demands and requests, a need to be in control, and a tendency to go to extreme lengths to either gain control or avoid demands. It was initially observed by Newson et al., (2003) in a subset of children with Atypical Autism. Presently, it continues to remain a controversial term and more recently it has been considered to be an important pattern of behavioural challenges, rather than a distinct syndrome or diagnosis in its own right (Green et al., 2018).

As noted previously, I have spent some time working with individuals with ASD diagnoses. I have always thoroughly enjoyed these experiences and so already had a natural interest in participating in research within the ASD field. When reading more about PDA from a range of sources I was interested by the level of need that children who are considered to display this behavioural profile have and also the level of interest behind this label, particularly from families. I considered that research in this area could potentially offer further insight for families and the ground for

intervention development. This motivated me as it appeared possible to make a real contribution to people's quality of life.

Executive Function is an umbrella term describing cognitive process important for the control and regulation of behaviour (Gioia, Isquith, & Guy, 1998). It seemed an important area to consider due to its more recently confirmed contribution to difficulties seen in ASD (Demetriou et al., 2018; Lai et al., 2017) and also its general involvement in behavioural control. However, one of the difficulties that I have noticed from conducting research in a fairly new field, is the lack of an established base of knowledge from which to draw hypotheses and predictions. Throughout the process I have found it quite challenging not being as able to draw on previous evidence and research. However, I hope that my contribution to the PDA literature will spark further interest in these behaviours and raise awareness of the difficulties that individuals affected by these behaviours face on a daily basis.

3.3.2 Working with parents of children with ASD and PDA traits

The empirical study involved comparing parent reports from a sample of children with ASD to a sample of children without ASD. However, as we were particularly interested in investigating PDA, we sought to recruit a sample of ASD children who also displayed PDA traits. This was challenging because PDA is not recognised as a diagnosable syndrome and, although interest in it has been growing, it is still not yet well known or understood, making it harder to reach participants. This meant that we had to be proactive in our recruitment strategy. This involved much researching of networks and societies where we might be able to access parents of children who presented with PDA behaviours. We found that often, we might be able to contact a charity or local support network but received no response from parents.

These challenges highlighted the difficulties of recruiting participants from a relatively new and under-researched area of interest.

Working with this population brought other challenges. Potentially, due to the high levels of needs of children with ASD and PDA traits (Gore Langton & Frederickson, 2016), parents were often busy and we had to be proactive in prompting and reminding parents to complete questionnaires. I found this process difficult as I was aware that we were placing added burden on families who already were caring for children with behavioural challenges. In an attempt to try to reduce the burden of our research, we ensured that participants were always reminded of their right to withdraw and encouraged them to contact us with any questions or issues they had. Also, when designing the method, we tried to keep the questionnaire battery as short as possible and provided an online and paper option of completing them to increase flexibility.

Many parents contacted us to ask for advice about further support or strategies to help them cope with their children's behaviours. We directed parents to support networks or societies but this at times felt like a tokenistic response to parents' often lengthily descriptions of their difficulties. This feeling of inadequacy resonated with experiences I have had whilst training as a Clinical Psychologist. At times whilst on placements I have felt unable to provide enough support for clients. I wondered whether this feeling of inadequacy was heightened due to the fact that we were recruiting a population that falls under the neurodevelopmental disorder umbrella, where there can sometimes be a sense of helplessness due to a lack of accessible interventions that can effectively ameliorate difficulties.

This study used a questionnaire methodology, which made it open to many informant-biases such as social desirability, demand characteristics, and extreme responding. The impact of these biases must be considered when interpreting results

and critiquing the methodology. As mentioned earlier, individuals with PDA traits may have high levels of need and parents may struggle to cope with difficult behaviours associated with this profile (Gore Langton & Frederickson, 2016). Many of the parents we recruited either asked us directly for advice about support or strategies, or made comments at the end of their questionnaires related to some of the challenging behaviours their children exhibit. It should be considered that parents may have thought that by providing the most extreme description of their child's profile they might help raise awareness of the difficulties and impact of having a child with PDA. It is also possible, that parents of children who display the most challenging behaviour are more motivated to take part. It is hard to estimate the extent of, or to control for these biases. However, we designed information sheets to clearly state the aims and intentions of the research and we provided thorough instructions before each questionnaire. Using observational methods alongside the questionnaires may have helped to assess the reliability of the questionnaire results.

The general struggles that exist among parents who have children with ASD who display PDA traits were apparent throughout the recruitment process. Due to the amount of feedback and personal stories we gathered from parents who wanted to share their opinions and experiences with us. This made me also reflect on the quantitative nature of the study. A quantitative method was chosen so we could statistically assess the relationships between EF and PDA traits. When formulating the research questions and methodology, a qualitative element was considered but not progressed due to concerns about resources and time. However, both parents and researchers may have gained more from the research process had we used a face-to-face qualitative method. This may have provided a richer understanding of the difficulties that families face, and a more supportive space in which to hear their

struggles and to meet their children. Alongside this, integrating their comments and views into the research could have enriched our findings. Future research would benefit from using qualitative methods as well as quantitative to validate findings and produce potentially more ecologically valid conclusions.

3.3.2 Parent-report measure selection

The first difficulty with measurement selection was related to PDA still being a fairly under researched topic and so few measures exist that have been created for use in this population or validated in it. Therefore, many measures were selected on the basis that they had previously been validated in ASD populations. However, the Extreme Demand Avoidance Questionnaire (EDQ-Q; O’Nions, Christie, Gould, Viding, & Happé, 2014) was the obvious choice to assess general levels of PDA traits as it is the only validated measure that attempts to measure PDA traits. However, as a measure it is still in its infancy in terms of research use, and therefore its validity and reliability must be considered when interpreting the results of the study.

When considering EF measurement I was aware of the debate surrounding how best to measure EF deficits so they reflect real world impacts (Burgess et al., 2006). When investigating the most recommended method for assessing EF in ASD, it was often suggested that parent-report measures are incorporated into studies alongside neuropsychological assessments to allow for the assessment of everyday behavioural manifestations of EF deficits (Burgess et al., 2006; Kenworthy et al., 2008). The Behaviour Rating Inventory of Executive Function (BRIEF; Gioia, Isquith, Guy, & Kenworthy, 2001) appeared to be the most validated and used parent-report questionnaire in the ASD population and this was therefore selected as a measure of EF. Although the BRIEF is not a direct measure of EF, other methods of assessing EF do not necessarily assess EF directly either. It is still not possible to account for other

processes that may contribute to EF abilities, such as the necessity of being able to understand a task and expectations (White, 2013). Due to the potential ecological validity benefits of parent-report measures, research constraints, and the wide use of the BRIEF within neurodevelopmental disorders research, it was decided that using the BRIEF was an acceptable method of assessing EF difficulties for the purposes of this study.

Measurement selection for specific behaviours that could be considered to resemble part of the PDA profile was not straight forward due to a lack of validated measures in this population. Demand avoidance is a prominent behavioural difficulty seen in those with ASD and PDA traits (Newson et al., 2003). Therefore, it was decided this should be a focus of part of the study. However, there are few measures for use with individuals with ASD that assess this specific behaviour. The Home Situation Questionnaire, which was developed for use with individuals with ASD (HSQ-ASD; Chowdhury et al., 2016), appeared to be the most suitable, validated measure that quantified this type of behaviour. This measure assessed two specific types of non-compliance behaviours, relating to demand type and to non-routine demands. Although the HSQ-ASD has been tested in children with ASD and behavioural problems, it has not been used directly to investigate PDA traits. Therefore, some items may refer to other types of non-compliance behaviour, rather than those specifically seen in individuals with PDA traits. This problem highlights the difficulties of conducting research in a relatively new field, and the trade-offs that must be made in order to start to develop a research base.

We also aimed to assess emotional lability in PDA (Newson et al., 2003). It seemed logical that there may be a connection between EF deficits and emotional lability due to previous findings of this in TD children (Carlson & Wang, 2007; Gyurak

et al., 2012; Zelazo & Cunningham, 2007). Financial constraints and methodological considerations, such as acceptability for participants, played a role in measurement selection. Emotional lability is a widely researched construct and therefore many questionnaires have been designed that appear to tap into this construct.

I chose the Affective Reactivity Inventory (ARI; Stringaris et al., 2012) as a measure of ER. Firstly, as it is a free to access measure, this allowed finances to be allocated to measures of other constructs where less choice was available. Secondly, we were conscious of keeping the total questionnaire battery as short as possible to increase acceptability of the method for busy parents, therefore the ARI was also chosen based on its conciseness. This measure specifically taps into anger, which seemed to also be appropriate due to the challenging behaviour that individuals with PDA often present with. However, this meant that it was a very narrow measure of emotional lability and therefore perhaps limited our quantification of the range of difficulties that could reflect emotional lability in an ASD population. Overall, the weakest relationships were found between this measure and measures of EF. Therefore, the choice of measure may have led us to draw too narrow conclusions about the relationship between EF and emotional lability in children with ASD and PDA. Importantly, this limitation was explored in the study's discussion, to highlight this problem.

The process of having to select less than optimal measures, and then develop a supporting argument for their use, highlighted to me the very real compromises that have to be made within the research context. It is possible that measurement selection may have influenced the results of this study. Considering the impact of measurement seemed important when interpreting the results and the necessity of being able to justify the decisions made about measurement selection became apparent. Using

questionnaires to capture EF deficits and PDA behaviours also meant that our study lacked the rich detail that more direct measures, such as observational and experimental assessments, would bring. I believe this experience has led me to develop more critical evaluation skills when considering research methodologies and presented results. Future research is needed to build on these initial findings using more robust and varied methodologies and measurements.

3.3.3 My experience of joint working

The empirical study was conducted in collaboration with Ellie Bishop, another Trainee Clinical Psychologist at University College London. I thoroughly enjoyed this experience and valued the opportunity to work closely with another trainee. When working clinically or in research you are often expected to work closely with other members of your team, and this joint project gave me the chance to reflect on the benefit of working with others. The saying, “two heads are better than one”, really felt apt when thinking about my experience. I found that creative thinking was easier when I was able to talk through challenges or barriers with my colleague and problem solve the issues together. By working with another trainee, I also felt more motivated to keep on track and work towards agreed goals. This experience made me think of the parallels between this and the importance of a good therapeutic relationship, when working clinically, to foster engagement and motivation.

3.3.4 Implications

3.3.4.1 Clinical

The overall finding that children with ASD and PDA traits appear to have more EF deficits than children without ASD, and also that these group differences are large, suggests that this should be considered when offering support to young people and their families. Certain strategies that are used to support individuals with EF

difficulties may also be successful for children with ASD and PDA traits. It was also found that certain EF deficits were more related to non-compliance behaviours in different contexts. For example, struggling with transitioning from a pleasant to less pleasant task was predicted by the Metacognition Index of the BRIEF, which involves skills such as planning, organising, initiating and holding information in mind. Conversely, non-compliance in situations involving uncertainty was predicted by the Behavioural Regulation Index of the BRIEF, which describes the ability to inhibit, shift behaviour and exercise emotional control. Therefore, when assessing non-compliance behaviour, it could be useful to consider firstly, what demand is being placed on the child, and then secondly, which EF skills they may rely on to comply with the demand.

3.3.4.2 Scientific

Most importantly, this research has raised awareness of the need for further research into EF in children with ASD and PDA traits. These preliminary findings suggest that there is a relationship between EF and PDA traits. As such, future studies should focus on establishing whether there is a causal relationship between EF and PDA features and continuing to explore other underlying mechanisms within the PDA profile. Related to this, this study has highlighted the need for the development of further measures that assess behaviours related to PDA. This would allow for more valid research to be conducted and aid interpretation of relationships found. Future research should include different methods of assessing EF such as, cognitive assessments, to gain a more direct understanding about the contribution of EF impairments to PDA behaviours. It could also be useful to compare EF profiles with other disorders where similar behavioural challenges are seen, such as ADHD or Conduct Disorder. This would make it possible to assess whether this is a unique

relationship to PDA traits, or whether deficits in EF contribute to similar behaviours in other groups.

3.4 Conclusion

The process of undertaking this research was both challenging and rewarding. My understanding of the problems inherent to broad psychological constructs, such as ER, has improved my ability to critically analyse both my own and others' research. I have learnt the importance of being flexible when conducting research in order to adapt to unforeseen challenges, to be able to problem solve, and to consider the impact of subjective decisions on scientific results. Investigating a fairly new field of research has provided me with a better understanding of the limits of researching an area with little background evidence upon which to build, and the compromises that are often made in order to begin to explore the area.

Overall, the finding that children and young people with ASD appear to have ER deficits across a wide range of domains when compared to TD controls should provide a better foundation for future investigation of these deficits and their potential relationship to symptoms of ASD. Future research is also warranted to further elucidate the cognitive underpinnings of the PDA phenotype and build upon the finding that deficits in EF appear to be related to PDA traits. Importantly, the findings also have highlighted that EF could be a target for intervention to support children with ASD and PDA traits with their difficulties, particularly with non-compliance behaviour.

3.5 References

- Borenstein, M., Hedges, L. V., Higgins, J. P. T., & Rothstein, H. R. (2009). Introduction to meta-analysis. *Clinical Rheumatology*, *20*(Supplement 1), 1.
- Burgess, P. W., Alderman, N., Forbes, C., Costello, A., M-A.Coates, L., Dawson, D. R., ... Channon, S. (2006). The case for the development and use of “ecologically valid” measures of executive function in experimental and clinical neuropsychology. *Journal of the International Neuropsychological Society*, *12*(02), 194–209.
- Carlson, S. M., & Wang, T. S. (2007). Inhibitory control and emotion regulation in preschool children. *Cognitive Development*, *22*(4), 489–510.
- Chowdhury, M., Aman, M. G., Lecavalier, L., Smith, T., Johnson, C., Swiezy, N., ... Scahill, L. (2016). Factor structure and psychometric properties of the revised home situations questionnaire for autism spectrum disorder: The Home situations questionnaire-autism spectrum disorder. *Autism*, *20*(5), 528–537.
- Cicchetti, D., Ackerman, B., & Izard, C. (1995). Emotions and emotion regulation in developmental psychopathology. *Development and Psychopathology*, *7*(1), 1–10.
- Cole, P. M., Martin, S. E., & Dennis, T. A. (2004). Emotion regulation as a scientific construct: Methodological challenges and directions for child development research. *Child Development*, *75*(2), 317–333.
- Demetriou, E. A., Lampit, A., Quintana, D. S., Naismith, S. L., Song, Y. J. C., Pye, J. E., ... Guastella, A. J. (2018). Autism spectrum disorders: a meta-analysis of executive function. *Molecular Psychiatry*, *23*(5), 1198–1204.
- Eisenberg, N., & Spinrad, T. L. (2004). Emotion-related regulation: sharpening the definition. *Child Development*, *75*(2), 334–339.
- Gioia, G. A., Isquith, P. K., & Guy, S. C. (1998). The regulatory role of executive

- control processes in children's behavioral, social, and emotional functioning. *Journal of Neuropsychiatry and Clinical Neurosciences*, 9(663).
- Gioia, G. A., Isquith, P., Guy, S., & Kenworthy, L. (2001). Brief rating inventory of executive function (BRIEF). *Odessa, FL: Psychological Assessment Resources*.
- Gore Langton, E., & Frederickson, N. (2016). Mapping the educational experiences of children with pathological demand avoidance. *Journal of Research in Special Educational Needs*, 16(4), 254–263.
- Gratz, K. L., & Roemer, L. (2004). Multidimensional assessment of emotion regulation and dysregulation: Development, factor structure, and initial validation of the difficulties in emotion regulation scale 1. *Journal of Psychopathology and Behavioral Assessment*, 26(1).
- Green, J., Absoud, M., Grahame, V., Malik, O., Simonoff, E., Le Couteur, A., & Baird, G. (2018). Pathological demand avoidance: Symptoms but not a syndrome. *The Lancet Child & Adolescent Health*, 0(0).
- Gross, J. J., & Thompson, R. A. (2007). Emotion regulation: Conceptual foundations. In J. J. Gross (Ed.), *Handbook of emotion regulation* (pp. 3–24). New York: Guildford Press.
- Gross, J. J. (1998). The emerging field of emotion regulation: An integrative review. *Review of General Psychology*, 2(5), 271–299.
- Gyurak, A., Goodkind, M. S., Kramer, J. H., Miller, B. L., & Levenson, R. W. (2012). Executive functions and the down-regulation and up-regulation of emotion. *Cognition & Emotion*, 26(1), 103–118.
- Kenworthy, L., Yerys, B. E., Anthony, L. G., & Wallace, G. L. (2008). Understanding executive control in autism spectrum disorders in the lab and in the real world. *Neuropsychology Review*, 18(4), 320–338.

- Lai, C., Lau, Z., Lui, S., Lok, E., Tam, V., Chan, Q., ... & Cheung, E. F. (2017). Meta-analysis of neuropsychological measures of executive functioning in children and adolescents with high-functioning autism spectrum disorder. *Autism Research, 10*(5), 911-939.
- Mazefsky, C. A., Herrington, J., Siegel, M., Scarpa, A., Maddox, B. B., Scahill, L., & White, S. W. (2013). The role of emotion regulation in autism spectrum disorder. *Journal of the American Academy of Child and Adolescent Psychiatry, 52*(7), 679–688.
- Mazefsky, C. A., Oswald, D. P., Day, T. N., Eack, S. M., Minshew, N. J., & Lainhart, J. E. (2012). ASD, a psychiatric disorder, or both? Psychiatric diagnoses in adolescents with high-functioning ASD. *Journal of Clinical Child and Adolescent Psychology: The Official Journal for the Society of Clinical Child and Adolescent Psychology, American Psychological Association, Division 53, 41*(4), 516–523.
- Mazefsky, C. A., Pelphrey, K. A., & Dahl, R. E. (2012). The need for a broader approach to emotion regulation research in autism. *Child Development Perspectives, 6*(1), 92–97.
- Mazefsky, C. A., & White, S. W. (2014). Emotion regulation: concepts and practice in autism spectrum disorder. *Child and Adolescent Psychiatric Clinics of North America, 23*(1), 15–24.
- Newson, E., Le Maréchal, K., & David, C. (2003). Pathological demand avoidance syndrome: A necessary distinction within the pervasive developmental disorders. *Archives of Disease in Childhood, 88*(7), 595–600.
- O’Nions, E., Christie, P., Gould, J., Viding, E., & Happé, F. (2014). Development of the ‘extreme demand avoidance questionnaire’ (EDA-Q): Preliminary

- observations on a trait measure for pathological demand avoidance. *Journal of Child Psychology and Psychiatry*, 55(7), 758–768.
- Scarpa, A., & Reyes, N. M. (2011). Improving emotion regulation with CBT in young children with high functioning autism spectrum disorders: A pilot study. *Behavioural and Cognitive Psychotherapy*, 39(04), 495–500.
- Stegenga, J. (2011). Is meta-analysis the platinum standard of evidence? *Studies in History and Philosophy of Science Part C: Studies in History and Philosophy of Biological and Biomedical Sciences*, 42(4), 497–507.
- Stringaris, A., Goodman, R., Ferdinando, S., Razdan, V., Muhrer, E., Leibenluft, E., & Brotman, M. A. (2012). The affective reactivity index: A concise irritability scale for clinical and research settings. *Journal of Child Psychology and Psychiatry*, 53(11), 1109–1117.
- Thomson, K., Burnham Riosa, P., & Weiss, J. A. (2015). Brief report of preliminary outcomes of an emotion regulation intervention for children with autism spectrum disorder. *Journal of Autism and Developmental Disorders*, 45(11), 3487–3495.
- Weiss, J. A., Thomson, K., & Chan, L. (2014). A Systematic literature review of emotion regulation measurement in individuals with autism spectrum disorder. *Autism Research*, 7(6), 629–648.
- Weiss, J. A., Thomson, K., Riosa, P. B., Albaum, C., Chan, V., Maughan, A., ... Black, K. (2018). A randomized waitlist-controlled trial of cognitive behavior therapy to improve emotion regulation in children with autism. *The Journal of Child Psychology and Psychiatry*.
- White, S. J. (2013). The triple I hypothesis: Taking another('s) perspective on executive dysfunction in autism. *Journal of Autism and Developmental Disorders*, 43(1), 114–121.

Zelazo, P., & Cunningham, W. (2007). Executive function: Mechanisms underlying emotion regulation.

Appendix I

Outline of contributions to joint project

Contributions to joint project

This project was part of a joint project with Ellie Bishop, a Trainee Clinical Psychologist from UCL. I will briefly outline what work was undertaken jointly and describe my individual contribution.

The ethics application was constructed and submitted jointly along with the Qualtrics online questionnaire base. The overall methodology was developed collaboratively, and Ellie and I were both responsible for recruiting participants and collecting data. This involved sending emails, communicating regularly about recruitment progress, discussing inclusion and exclusion criteria and answering questions from participants. However, this also involved independently conducting searches to find contacts and relevant networks and groups and contacting them to advertise the study.

Research questions, synthesis of theories and hypotheses development were all done independently, but with the support of our supervisors (Dr Will Mandy and Dr Liz O’Nions). Ellie’s project had a specific focus on the construct, Theory of Mind, and therefore explored different concepts, relationships and behaviours to mine, which investigated Executive Function. Data for each project were collated and analysed independently. The write up of the findings were also done independently.

Appendix II

Quality and relevance assessment

Adapted from Newcastle-Ottawa Quality Assessment Scale (Cohort Studies)

Italics represent changes from original assessment scale

Note: A study can be awarded a maximum of one star for each numbered item within each category.

Selection (Max 3*)

- 1) Representativeness of exposed cohort (ASD Sample)
 - a. Truly representative of the *average young person with ASD* *
 - b. Somewhat representative of the *average young person with ASD* *
 - c. Selected group of users (*e.g. using specialist services or with a particular need*)
 - d. No description of the derivation of the cohort
- 2) *CRITERION 2 (SELECTION OF THE NON-EXPOSED COHORT) REMOVED AS NOT APPLICABLE TO CURRENT REVIEW*
- 3) Ascertainment of ASD diagnosis
 - a. *Diagnosis confirmed with validated measures (E.g. ADI-R and ADOS)* *
 - b. *Evidence of comprehensive assessment by a health professional* *
 - c. *Self- or parent-report of diagnosis*
 - d. No description
- 4) *CRITERION 4 (DEMONSTRATION THAT OUTCOME OF INTEREST WAS NOT PRESENT AT START OF STUDY) REMOVED AS NOT APPLICABLE TO CURRENT REVIEW*
- 5) Sample Size
 - a. *Thirty or more young people with ASD included* *
 - b. *Less than thirty young people with ASD included*

Control (Max 1*)

- 1) Appropriate control for significant confounding factor
 - a. Study controls for *learning disability or IQ<70 in analysis* such that it is possible to draw conclusions about ASD independent of learning disability *
 - b. *Participants with learning disability, IQ<70 or below 'normal range' excluded* *
 - c. *Learning disability or IQ* not controlled for
 - d. *Learning disability or IQ* not reported

Outcome (Max 2*)

- 1) A. Assessment of outcome
 - a. *Appropriate outcome measures (e.g. validated tool)* *
 - b. Inappropriate measures
- 2) B. Number of ER domains assessed by outcome measure
 - a. *Three or more ER domains assessed* *

b. *Less than three domains assessed*

- 3) *CRITERION 2 (WAS FOLLOW-UP LONG ENOUGH OR OUTCOMES TO OCCUR) REMOVED AS NOT APPLICABLE TO CURRENT REVIEW*
- 4) *CRITERION 3 (ADEQUACY OF FOLLOW-UP OF COHORTS) REMOVED AS NOT APPLICABLE TO CURRENT REVIEW*

Appendix III

Research ethical committee approval letter



24th February 2017

Dr William Mandy
UCL Division of Psychology and Language Sciences

Dear Dr Mandy

Notification of Ethical Approval

Re: Ethics Application 10193/001: Exploring demand avoidance in children with and without Autism

I am pleased to confirm in my capacity as Chair of the UCL Research Ethics Committee (REC) that your study has been ethically approved by the REC until **30th September 2018**.

Approval is subject to the following conditions:

Notification of Amendments to the Research

You must seek Chair's approval for proposed amendments (to include extensions to the duration of the project) to the research for which this approval has been given. Ethical approval is specific to this project and must not be treated as applicable to research of a similar nature. Each research project is reviewed separately and if there are significant changes to the research protocol you should seek confirmation of continued ethical approval by completing the 'Amendment Approval Request Form':

<http://ethics.grad.ucl.ac.uk/responsibilities.php>

Adverse Event Reporting – Serious and Non-Serious

It is your responsibility to report to the Committee any unanticipated problems or adverse events involving risks to participants or others. The Ethics Committee should be notified of all serious adverse events via the Ethics Committee Administrator (ethics@ucl.ac.uk) immediately the incident occurs. Where the adverse incident is unexpected and serious, the Chair or Vice-Chair will decide whether the study should be terminated pending the opinion of an independent expert. For non-serious adverse events the Chair or Vice-Chair of the Ethics Committee should again be notified via the Ethics Committee Administrator within ten days of the incident occurring and provide a full written report that should include any amendments to the participant information sheet and study protocol. The Chair or Vice-Chair will confirm that the incident is non-serious and report to the Committee at the next meeting. The final view of the Committee will be communicated to you.

Final Report

At the end of the data collection element of your research we ask that you submit a very brief report (1-2 paragraphs will suffice) which includes in particular issues relating to the ethical implications of the research i.e. issues obtaining consent, participants withdrawing from the research, confidentiality, protection of participants from physical and mental harm etc.

Yours sincerely


Professor Michael Heinrich
Interim Chair, UCL Research Ethics Committee

Cc: Anna Goodson & Ellie Bishop

Academic Services, 1-19 Torrington Place (9th Floor),
University College London
Tel: +44 (0)20 3108 8216
Email: ethics@ucl.ac.uk
<http://ethics.grad.ucl.ac.uk/>

Appendix IV

Participant information sheet

Exploring Demand Avoidance in Children with and without Autism

Thank you for expressing an interest in taking part in our study (UCL Ethical Approval Ref: 10193/001)

You should have already received a copy of this information sheet but please check that you have read it carefully before continuing.

Before you decide whether to take part, it is important for you to understand why the research is being done and what it will involve. Please take time to read the following information carefully and feel free to discuss with others if you wish. If there is anything that is not clear, or if you would like further information, please send us an email (e.bishop@ucl.ac.uk / anna.goodson.15@ucl.ac.uk).

What is the research about?

This research is exploring demand avoidance behaviour in children. Having a child who struggles with demand avoidance behaviours can be stressful and challenging at times. Children with demand avoidance difficulties have some of the highest rates of exclusion from schools and education. By taking part in this study, you are helping contribute to an increased understanding of the cognitive processes in demand avoidance. This could help raise awareness, inform behavioural management strategies for children, develop support for parents, and increase the availability and understanding of these.

What does taking part involve?

There are two stages to participation in this study. Firstly, all participants will be asked to complete a short set of questionnaires. You will be taken to this part of the study once you have finished reading the information on this page and given your consent to participate.

Secondly, some participants will be asked to complete some follow-up questionnaires about their child's behaviour. These can be done online or on paper and should take approximately one hour. You do not need to complete all the questionnaires at one time and will be able to save your progress so you can return to it at a more convenient time.

After completing the questionnaires, to thank you for your participation you will be given the chance to enter a prize draw to win an Amazon voucher, ranging in value from £10 to £50.

What will happen to my information?

All information collected about you during the course of the study will be kept strictly confidential and stored in secure University College London (UCL) premises. Your name and contact details will be stored separately from the data collected. All information will be kept securely according to the requirements of the Data Protection Act 1998.

It is likely that the results of this study will be published, but any published outcomes will remain strictly anonymous. Only group results will be presented and no individual will be discussed. Identifiable information (such as name or date of birth) will not appear on any publications or reports about this research. If any work is to be published, you will be notified of this and able to request a copy.

Do I have you take part?

No. It is up to you to decide whether or not to take part in this study. In other words, this is voluntary. If you do decide to take part you are still free to stop your participation at any time and have any research data withdrawn without giving a reason.

Having read this information, do you wish to continue with this study?

Appendix V

Participant consent form (as displayed on Qualtrics)

By continuing with this study, I agree that:

- I have read the notes written above and the Information Page, and understand what the study involves.
- I understand that if I decide at any time that I no longer wish to take part in this project, I can notify the researchers and withdraw immediately.
- I consent to the processing of my personal information for the purposes of this research study.
- I agree that my data, after it has been fully anonymised, can be shared with other researchers.
- I understand that such information will be treated as strictly confidential and handled in accordance with the provisions of the Data Protection Act 1998.
- I consent to the above and agree to participate in this study.

By continuing with this study, you consent to all of the above.

- I consent to participate in this study
- I do not consent to participate in this study

Appendix VI

Research poster for advertisement

RESEARCH



Exploring Demand Avoidance in Children with and without Autism



Inviting parents of children aged 6-11 years old to take part in research....

WHAT IS IT ABOUT???

This research will explore demand avoidance behaviours in children, and the thoughts and processes associated with these behaviours. Specifically in three separate groups of children:

1. Children with an **autistic spectrum diagnosis**
2. Children who display **demand avoidance** behaviours
3. Children **without autism**

WHAT DOES IT INVOLVE???

The study will ask you as parents to complete some basic questionnaires about your child's behaviour. This should not take more than 1 hour of your time. You will be entered into a prize draw to win amazon vouchers!

YES

NO

If you are interested/for further information please contact:

Anna Goodson or Ellie Bishop

anna.goodson.15@ucl.ac.uk

e.bishop@ucl.ac.uk

THANK YOU



This study complies with the Data Protection Act (1998) and has been approved by the UCL ethics committee 10193/001

Appendix VII

Participant contact email templates

Phase One

Dear X,

Thanks so much for your interest in our study. It would be great if you could participate in our research. All the information you need should be below, but if you have any questions or comments regarding the questionnaires, please just email myself or Ellie (copied in).

This study involves completing an online questionnaire about your child who is aged between 6-11 years old. All the answers you provide must be related to just one of your children. Based on your responses to these questions, you might be asked to complete some further questions. We will get back in touch with you to let you know if this is the case.

Below is a participant code. This is unique to you and needs to be entered into the online questionnaire. There is also a link which will take you directly to the online questionnaire. It is important that you try to complete the questionnaire in one sitting to ensure it saves all your responses. Therefore, it is best to start the questionnaire when you have about 20 minutes of free time.

If you do not wish to complete the questionnaire online and would prefer a paper version please let us know by replying to this email with your postal address and we will send them to you with a stamped, addressed envelope for you to return them.

Participant Code:

Password:

Click below for the study link (you may need to hold ctrl + click the link)
[Phase 1: Exploring Demand Avoidance in Children with and without Autism](#)

We'll send you reminders every week to complete the questionnaires, but if you want to opt out at any point please just let us know and we'll stop! Additionally, if you know anyone else who might be interested in participating, please feel free to pass our details on.

Again, thank you very much for your help and interest in our study. If you have any questions please do not hesitate to contact either Anna Goodson (anna.goodson.15@ucl.ac.uk) or Ellie Bishop (e.bishop@ucl.ac.uk).

Best Wishes,

Ellie & Anna

Phase Two

Dear X,

Thank you very much for completing the first phase of our research study! We appreciate the time and thought taken.

Based on your responses to the first set of questionnaires, we would be very grateful if you could complete the second phase of our study. This is done in **two parts** and it's very important you complete both.

Below is a participant code. This is unique to you and needs to be entered when prompted. There are also **two links** which will take you directly to the online questionnaires. It is important that you try to complete the questionnaires in one sitting to ensure it saves all your responses. Therefore, it is best to start the questionnaires when you have about 15 minutes of free time.

Participant Code:

Password:

PART 1 (you may need to copy and paste the link into your browser window)

<https://www.theoryofmindinventory.com/professionals/caregiver-assessment/>

Click below for PART 2 link

[Phase 2: Exploring Demand Avoidance in Children with and without Autism](#)

Please follow both links separately, as the questionnaires are different!

Again, thank you very much for your help and interest in our study. If you have any questions please do not hesitate to contact either Anna Goodson (anna.goodson.15@ucl.ac.uk) or Ellie Bishop (e.bishop@ucl.ac.uk).

Best Wishes,

Ellie & Anna

Appendix VIII

Demographic questionnaire (as displayed on Qualtrics)

Please answer the following questions about YOURSELF

1. *Date of Birth (year)*

2. *Gender*

- Male
- Female
- Prefer not to say

3. *First Language*

4. *Ethnicity*

- White British / White Other
- Asian British / Asian Indian / Pakistani / Chinese / Other
- Black British / Black African / Black Caribbean / Other
- Mixed (please specify)

- Other (please specify)

- Prefer not to say

5. *Does your child live with you?*

- Yes - all of the time
- Yes - some of the time
- No

Please answer the following questions about YOUR CHILD

1. *Date of Birth (month/year)*

Month

Year

2. *Gender*

- Male
- Female
- Prefer not to say

3. *First Language*

4. *Ethnicity*

- White British / White Other
- Asian British / Asian Indian / Pakistani / Chinese / Other
- Black British / Black African / Black Caribbean / Other
- Mixed (please specify)
- Other (please specify)
- Prefer not to say

5. *Please indicate whether your child has ever received one of the following Autistic Spectrum Disorder diagnoses:*

- Autism Spectrum Disorder (ASD) / Autism Spectrum Condition (ASC)
- Autism/Autistic Disorder
- Asperger's Syndrome/Asperger's Disorder
- High-Functioning Autism
- Pervasive Developmental Disorder - Not Otherwise Specified (PDD-NOS)
- Other (please specify)
- None of the above

6. *If yes to the previous question, where/from whom was the diagnosis received?*

- Child and Adolescent Mental Health Service (CAMHS)
- Child and Adolescent Psychiatrist
- Clinical Psychologist
- Educational Psychologist
- Paediatrician

- Speech and Language Therapist
- Other (please specify)
- × I don't know
- × N/A

7. *Has your child ever been given any of the following:*

- A diagnosis of PDA (Pathological Demand Avoidance)
- A diagnosis of demand avoidant traits/PDA features
- Suspected PDA but not clinically diagnosed
- None of the above
- I don't know

8. *If yes to the previous question, who gave your child the diagnosis?*

- Child and Adolescent Mental Health Service (CAMHS)
- Child and Adolescent Psychiatrist
- Clinical Psychologist
- Educational Psychologist
- Paediatrician
- Speech and Language Therapist
- Other (please specify)
- × I don't know
- × N/A

9. *What type of school does your child attend?*

1. Mainstream school
2. Special school for children with Autism

3. Special school for children with Learning Disabilities
4. Specialised unit within a mainstream school
5. Home school
6. My child is not in education
7. Other (please specify)
8. × I don't know

10. Has your child been diagnosed with any of the following specific learning disabilities/difficulties? Please select one or more of the options below:

- Mild Learning Disability
- Moderate Learning Disability
- Severe Learning Disability
- Profound and Multiple Learning Disability (PMLD)
- Dyslexia
- Dyscalculia
- Dyspraxia
- Dysgraphia
- Attention Deficit Disorder (ADD)
- Attention Deficit Hyperactivity Disorder (ADHD)
- Other (please specify)
- None of the above
- × I don't know

Appendix IX

Extreme Demand Avoidance – Questionnaire (EDA-Q; O’Nions et al., 2014)

To be completed by parent and/or teacher. One box to be ticked per question.

	Not true	Somewhat true	Mostly true	Very true
1. Obsessively resists and avoids ordinary demands and requests.				
2. Complains about illness or physical incapacity when avoiding a request or demand.				
3. Is driven by the need to be in charge.				
4. Finds everyday pressures (e.g. having to go on a school trip/ visit dentist) intolerably stressful.				
5. Tells other children how they should behave, but does not feel these rules apply to him/herself.				
6. Mimics adult mannerisms and styles (e.g. uses phrases adopted from teacher/parent to tell other children off).				
7. Has difficulty complying with demands unless they are carefully presented.				
8. Takes on roles or characters (from TV/real life) and 'acts them out'.				
9. Shows little shame or embarrassment (e.g. might throw a tantrum in public and not be embarrassed).				
10. Invents fantasy worlds or games and acts them out.				
11. Good at getting around others and making them do as s/he wants.				
12. Seems unaware of the differences between him/herself and authority figures (e.g. parents, teachers, police).				
13. If pressurised to do something, s/he may have a 'meltdown' (e.g. scream, tantrum, hit or kick).				
14. Likes to be told s/he has done a good job.				
15. Mood changes very rapidly (e.g. switches from affectionate to angry in an instant).				
16. Knows what to do or say to upset specific people.				

17. Blames or targets a particular person.				
18. Denies behaviour s/he has committed, even when caught red handed.				
19. Seems as if s/he is distracted 'from within'.				
20. Makes an effort to maintain his/her reputation with peers.				
21. Uses outrageous or shocking behaviour to get out of doing something.				
22. Has bouts of extreme emotional responses to small events (e.g. crying/giggling, becoming furious).				
23. Social interaction has to be on his or her own terms.				
24. Prefers to interact with others in an adopted role, or communicate through props/toys.				
25. Attempts to negotiate better terms with adults.				
26. S/he was passive and difficult to engage as an infant.				

Appendix X

The Behaviour Regulation Index of Executive Function (BRIEF; Gioia et al., 2000)

Removed due to copyright.

Appendix XI

Home Situations Questionnaire – ASD (HSQ-ASD; Chowdhury et al., 2016)

Removed due to copyright.

Appendix XII

The Affective Reactivity Index (ARI; Stringaris et al., 2012)

Removed due to copyright.

Appendix XIII

Childhood Autism Spectrum Test (CAST; Scott et al., 2002)

1. Does s/he join in playing games with other children easily?	Yes	No
2. Does s/he come up to you spontaneously for a chat?	Yes	No
3. Was s/he speaking by 2 years old?	Yes	No
4. Does s/he enjoy sports?	Yes	No
5. Is it important to him/her to fit in with the peer group?	Yes	No
6. Does s/he appear to notice unusual details that others miss	Yes	No
7. Does s/he tend to take things literally?	Yes	No
8. When s/he was 3 years old, did s/he spend a lot of time pretending (e.g., play-acting being a superhero, or holding teddy's tea parties)?	Yes	No
9. Does s/he like to do things over and over again, in the same way all the time?	Yes	No
10. Does s/he find it easy to interact with other children?	Yes	No
11. Can s/he keep a two-way conversation going?	Yes	No
12. Can s/he read appropriately for his/her age?	Yes	No
13. Does s/he mostly have the same interests as his/her peers?	Yes	No
14. Does s/he have an interest which takes up so much time that s/he does little else?	Yes	No
15. Does s/he have friends, rather than just acquaintances?	Yes	No
16. Does s/he often bring you things s/he is interested in to show you?	Yes	No
17. Does s/he enjoy joking around?	Yes	No
18. Does s/he have difficulty understanding the rules for polite behavior?	Yes	No
19. Does s/he appear to have an unusual memory for details?	Yes	No
20. Is his/her voice unusual (e.g., overly adult, flat, or very monotonous)?	Yes	No
21. Are people important to him/her?	Yes	No
22. Can s/he dress him/herself?	Yes	No
23. Is s/he good at turn-taking in conversation?	Yes	No
24. Does s/he play imaginatively with other children, and engage in role-play?	Yes	No
25. Does s/he often do or say things that are tactless or socially inappropriate?	Yes	No
26. Can s/he count to 50 without leaving out any numbers?	Yes	No
27. Does s/he make normal eye-contact?	Yes	No
28. Does s/he have any unusual and repetitive movements?	Yes	No
29. Is his/her social behaviour very one-sided and always on his/her own terms?	Yes	No
30. Does s/he sometimes say "you" or "s/he" when s/he means "I"?	Yes	No
31. Does s/he prefer imaginative activities such as play-acting or story-telling, rather than numbers or lists of facts?	Yes	No
32. Does s/he sometimes lose the listener because of not explaining what s/he is talking about?	Yes	No
33. Can s/he ride a bicycle (even if with stabilizers)?	Yes	No
34. Does s/he try to impose routines on him/herself, or on others, in such a way that it causes problems?	Yes	No
35. Does s/he care how s/he is perceived by the rest of the group?	Yes	No

36. Does s/he often turn conversations to his/her favorite subject rather than following what the other person wants to talk about?	Yes	No
37. Does s/he have odd or unusual phrases?	Yes	No
38. Have teachers/health visitors ever expressed any concerns about his/her development?	Yes	No
39. Has s/he ever been diagnosed with any of the following: Language delay, ADHD, hearing or visual difficulties, Autism Spectrum Condition (including Asperger's Syndrome, or a physical disability)?	Yes	No

Appendix XIV

Strengths and Difficulties Questionnaire (SDQ; Goodman, 2001)

For each item, please mark the box for Not True, Somewhat True or Certainly True. It would help us if you answered all items as best you can even if you are not absolutely certain or the item seems daft! Please give your answers on the basis of the child's behaviour over the last six months or this school year.

Child's Name

Male/Female

Date of Birth.....

	Not True	Somewhat True	Certainly True
Considerate of other people's feelings	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Restless, overactive, cannot stay still for long	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Often complains of headaches, stomach-aches or sickness	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Shares readily with other children (treats, toys, pencils etc.)	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Often has temper tantrums or hot tempers	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Rather solitary, tends to play alone	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Generally obedient, usually does what adults request	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Many worries, often seems worried	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Helpful if someone is hurt, upset or feeling ill	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Constantly fidgeting or squirming	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Has at least one good friend	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Often fights with other children or bullies them	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Often unhappy, down-hearted or tearful	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Generally liked by other children	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Easily distracted, concentration wanders	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Nervous or clingy in new situations, easily loses confidence	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Kind to younger children	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Often lies or cheats	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Picked on or bullied by other children	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Often volunteers to help others (parents, teachers, other children)	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Thinks things out before acting	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Steals from home, school or elsewhere	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Gets on better with adults than with other children	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Many fears, easily scared	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Sees tasks through to the end, good attention span	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

Signature

Date

Parent/Teacher/Other (please specify:)

Thank you very much for your help

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Appendix XV

Final regression model adequacies: checking assumptions

1. Regression of the EDA-Q and indices of EF

The Durbin-Watson value was close to 2 (1.98) which indicated that the assumption of independent errors between residuals is almost fulfilled. An assessment of the VIF and Tolerance statistics indicated that the assumptions of no multicollinearity have been met as all Tolerance values are above .20 and all VIF values are not substantially greater than 1. Casewise diagnostics were also examined and in a sample of 64 we would expect no more than three cases (5%) of the standardised residuals to lie outside of +/- 2. None of the standardised residuals were outside this range. Therefore, the sample appears to confirm to what is expected for a fairly accurate model. A visual inspection of the plotted residuals suggests an approximately normal distribution and therefore supports the assumption of no heteroscedasticity.

2. Regression of the EDA-Q, the CAST and indices of EF

The Durbin-Watson value was close to 2 (2.05) which indicated that the assumption of independent errors between residuals is almost fulfilled. An assessment of the VIF and Tolerance statistics indicated that the assumptions of no multicollinearity have been met as all Tolerance values are above .20 and all VIF values are not substantially greater than 1. Casewise diagnostics were also examined and in a sample of 64 we would expect no more than three cases (5%) of the standardised residuals to lie outside of +/- 2. None of the standardised residuals were outside this range. Therefore, the sample appears to confirm to what is expected for a fairly accurate model. A visual inspection of the plotted residuals suggests an approximately normal distribution and therefore supports the assumption of no heteroscedasticity.

3. Regression of the ARI and indices of EF

The Durbin-Watson value was close to 2 (2.01) which indicated that the assumption of independent errors between residuals is almost fulfilled. An assessment of the VIF and Tolerance statistics indicated that the assumptions of no multicollinearity have been met as all Tolerance values are above .20 and all VIF values are not substantially greater than 1. Casewise diagnostics were also examined and in a sample of 64 we would expect no more than three cases (5%) of the standardised residuals to lie outside of +/- 2. Only two of the standardised residuals were outside this range. Therefore, the sample appears to confirm to what is expected for a fairly accurate model. A visual inspection of the plotted residuals suggests an approximately normal distribution and therefore supports the assumption of no heteroscedasticity.

4. Regression of the HSQ-DS and indices of EF

The Durbin-Watson value was close to 2 (2.17) which indicated that the assumption of independent errors between residuals is almost fulfilled. An assessment of the VIF and Tolerance statistics indicated that the assumptions of no multicollinearity have been met as all Tolerance values are above .20 and all VIF values are not substantially greater than 1. Casewise diagnostics were also examined and in a sample of 64 we would expect no more than three cases (5%) of the standardised residuals to lie outside of +/- 2. Only two of the standardised residuals were outside this range. Therefore, the sample appears to confirm to what is expected for a fairly accurate model. A visual

inspection of the plotted residuals suggests an approximately normal distribution and therefore supports the assumption of no heteroscedasticity.

5. *Regression of the HSQ-DS, the CAST and indices of EF*

The Durbin-Watson value was close to 2 (2.20) which indicated that the assumption of independent errors between residuals is almost fulfilled. An assessment of the VIF and Tolerance statistics indicated that the assumptions of no multicollinearity have been met as all Tolerance values are above .20 and all VIF values are not substantially greater than 1. Casewise diagnostics were also examined and in a sample of 64 we would expect no more than three cases (5%) of the standardised residuals to lie outside of +/- 2. Only two of the standardised residuals were outside this range. Therefore, the sample appears to confirm to what is expected for a fairly accurate model. A visual inspection of the plotted residuals suggests an approximately normal distribution and therefore supports the assumption of no heteroscedasticity.

6. *Regression of the HSQ-SI and the indices of EF*

The Durbin-Watson value was close to 2 (2.16) which indicated that the assumption of independent errors between residuals is almost fulfilled. An assessment of the VIF and Tolerance statistics indicated that the assumptions of no multicollinearity have been met as all Tolerance values are above .20 and all VIF values are not substantially greater than 1. Casewise diagnostics were also examined and in a sample of 64 we would expect no more than three cases (5%) of the standardised residuals to lie outside of +/- 2. Only two of the standardised residuals were outside this range. Therefore, the sample appears to confirm to what is expected for a fairly accurate model. A visual inspection of the plotted residuals suggests an approximately normal distribution and therefore supports the assumption of no heteroscedasticity.

7. *Regression of the HSQ-SI and the indices of EF*

The Durbin-Watson value was close to 2 (2.34) which indicated that the assumption of independent errors between residuals is almost fulfilled. An assessment of the VIF and Tolerance statistics indicated that the assumptions of no multicollinearity have been met as all Tolerance values are above .20 and all VIF values are not substantially greater than 1. Casewise diagnostics were also examined and in a sample of 64 we would expect no more than three cases (5%) of the standardised residuals to lie outside of +/- 2. Only one of the standardised residuals were outside this range. Therefore, the sample appears to confirm to what is expected for a fairly accurate model. A visual inspection of the plotted residuals suggests an approximately normal distribution and therefore supports the assumption of no heteroscedasticity.

Appendix XVI

Alphabetised list of abbreviations

ARI: Affective Reactivity Index

ASD: Autism Spectrum Disorder

ASC: Autism Spectrum Condition

BRIEF: Behaviour Rating Inventory of Executive Function

BRI: Behaviour Regulation index

CAST: Child Autism Spectrum Test

CBQ: Children's Behaviour Questionnaire

CBT: Cognitive Behavioural Therapy

DISCO: Diagnostic Interview for Social and Communication Disorders

DSM: Diagnostic Statistical Manual of Mental Disorder

EAQ: Emotion Awareness Questionnaire

EATQ-R: Early Adolescent Temperament Questionnaire-Revised

ECS: Effortful Control Scale

EDA-Q: Extreme Demand Avoidance - Questionnaire

EF: Executive Function

ER: Emotion Regulation

ERC: Emotion Regulation Checklist

ERICA: Emotion Regulation Index for Children and Adolescents

ERQ: Emotion Regulation Questionnaire

GEC: Global Executive Composite

HFA: High-Functioning Autism

HSQ-ASD: Home Situation Questionnaire – Autism Spectrum Disorder

HSQ-DS: Home Situation Questionnaire – Demand Specific

HSQ-SI: Home Situation Questionnaire – Social Inflexibility

ICD-10: International Statistical Classification of Diseases and Related Health problems 10th revision

IRPA: Instrument for Reactive and Proactive Aggression

MI: Metacognition Index

MSCS: Multidimensional Social Competence Scale

NOS: Newcastle-Ottawa Scale

PANAS-C: Positive and Negative Affect Schedule for Children

PDA: Pathological Demand Avoidance

PDD-NOS: Pervasive Developmental Disorders – Not Otherwise Specified

RSQ: Response to Stress Questionnaire

SCAS-P: Spence Children's Anxiety Scale - Parents

SDQ: Strengths and Difficulties Questionnaire

SSRS: Social Skills Rating System – Elementary Parent Form

TD: Typically Developing

UCL: University College London