Developing and Piloting a New Parent-Report Measure of Executive Function for Children with Autism Spectrum Disorder (ASD)

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D.Clin.Psy. thesis (Volume 1), 2017

University College London

UCL Doctorate in Clinical Psychology

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.

Signature: 

Name: Laurie Ledger-Hardy

Date: 07/07/2017

Overview

This thesis focuses on executive functioning in children with Autism Spectrum Disorder (ASD) and is presented in three parts. Part 1 presents a systematic literature review examining the degree of executive function difficulties in children with ASD. The review focused on studies which used the Behavior Rating Inventory of Executive Function (BRIEF), an informant-report rating scale, with children with ASD and a typically developing (TD) control group. Using a meta-analysis, the review found large effect sizes on the BRIEF, demonstrating that children with ASD are reported to have substantial executive function difficulties when compared to TD children.

Part 2, the empirical research paper, describes the development and piloting of a new parent-report measure of executive function in children with ASD. Qualitative data generated through interviews were analysed and used to develop the measure. The measure was then piloted on the internet with parents of children with ASD and parents of TD children. The psychometric properties of the measure were examined, revealing promising indications of reliability and validity. Limitations of the measure and its relationship to the BRIEF are discussed.

Part 3 is a critical appraisal of the research process. Limitations of both the study and measure are presented, before discussing methodological challenges that arose throughout the project. Consideration is given to broader conceptual issues, before finally offering reflections on the research process and on my own personal development.

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Acknowledgements

I would like to thank everyone who has assisted me in completing this project. Firstly, I would like to thank my supervisor Dr Will Mandy for his continued support and guidance. I have really benefited from Will's knowledge and research expertise and learnt a great deal throughout this process.

I would like to extend a huge thank you to all the parents who took part and were happy to give up their time, without them this project would not have been possible. Thank you also to the professionals involved in the interviews who offered their time and expertise.

Several organisations and support groups deserve particular mention as they supported with recruitment by advertising the project. In particular, I would like to thank Hope GB, Parents Talking Asperger's and Research Autism.

Finally, thank you to my family, friends and fellow trainees for your continued support and encouragement.

Part 1: Literature Review

Parent-Reported Executive Function Deficits in Children with Autism: A Systematic Review and Meta-Analysis of Studies Using the BRIEF

Abstract

Aims: The aim of this meta-analysis was to assess whether children with Autism Spectrum Disorder (ASD) show deficits in executive functioning on the Behavior Rating Inventory of Executive Function (BRIEF), a widely used informant-report questionnaire measure, when compared to typically developing (TD) controls.

Methods: PsycINFO, Web of Science and Medline were searched to identify peer-reviewed studies which used the BRIEF (child version) with an ASD and TD group. A random effects meta-analysis was carried out, looking at the global composite (GEC) and both indices on the BRIEF (BRI and MI). Heterogeneity between studies was explored using sub-group analyses.

Results: After screening, sixteen studies were included in the final analysis. Very large effects were found on the GEC (*d* = 2.38, 95% CI 2.06, 2.71), and both indices of the BRIEF (BRI *d* = 2.19, 95% CI 1.91, 2.46; MI *d* = 2.04, 95% CI 1.78, 2.3), demonstrating that children with ASD are reported to have substantial difficulties with executive function when compared to TD controls. A high level of heterogeneity was observed between studies. Exploratory analyses were carried out to investigate this but moderators were not found.

Conclusion: Children with ASD have significant deficits in executive function compared to TD children as measured by the BRIEF, an informant-report questionnaire measure. This review provides further evidence of a discrepancy between findings on performance-based neuropsychological tests and rating scales, with the latter demonstrating much larger effect sizes.

Introduction

Autism Spectrum Disorder

Autism Spectrum Disorder (ASD) is a neurodevelopmental condition affecting around 1% of the population (Baird et al., 2006; Brugha et al., 2012). The Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition (DSM-5; American Psychiatric Association, 2013) groups the core deficits of ASD into two areas. The first area is "social communication and social interaction" which manifests as deficits in social-emotional reciprocity, non-verbal communication and forming, maintaining and understanding relationships. The second area is "restricted, repetitive patterns of behavior, interests, or activities", and the presence of at least two of the following is necessary to meet criteria for a diagnosis: stereotyped or repetitive behaviours, movements or speech; inflexible or ritualised behaviour; restricted and fixated interests; and sensory abnormalities. ASD is therefore characterised by difficulties in both social and non-social domains. The symptoms of ASD must be present in the early developmental period, although they may not be recognised until later in development when they are likely to cause more impairment. ASD varies in level of severity and may be accompanied by intellectual disability and/or language impairment. It is estimated that around 30 to 50% of people with ASD have an intellectual disability (Baird et al., 2006; Chakrabarti & Fombonne, 2005). ASD is more commonly diagnosed in males, however, the under-recognition of ASD in females is likely to play a role in this (Loomes, Hull, & Mandy, 2017).

Numerous theories have been proposed in an attempt to account for the broad range of cognitive, behavioural and emotional difficulties seen in ASD. Cognitive models seek to explain the diverse signs and symptoms of ASD in terms of underlying patterns of cognitive deficit and strength. Three of the most influential cognitive theories, each focused on different characteristics of ASD, are: Theory of Mind (e.g. Baron-Cohen, Leslie, & Frith, 1985), Weak Central Coherence (e.g.

Happé & Frith, 2006) and Executive Dysfunction (e.g. Hughes, Russell, & Robbins, 1994). This review will focus on the relationship between executive dysfunction and ASD. The executive dysfunction theory proposes that deficits in executive function are a primary or causal feature of ASD and that this is linked to neural abnormalities in the frontal lobes (Damasio & Maurer, 1978; Maurer & Damsaio, 1982). Early evidence for the executive dysfunction account of ASD stemmed from the observation that patients with frontal lobe damage showed similar behaviours and social difficulties to those seen in individuals with ASD (Damsaio & Maurer, 1978). Proponents of the executive dysfunction model argue that it can account for both the social and non-social features of ASD (Hughes, Russell, & Robbins, 1994). It has been proposed that deficits in specific executive functions may account for particular features of ASD, for example, deficits in cognitive flexibility may underlie the observed inflexibility (Turner, 1999). This model has fuelled much research investigating whether executive dysfunction is universal in people with ASD and whether there is an autism-specific pattern of executive dysfunction. Before exploring evidence of executive dysfunction in ASD, background to the concept of executive function will be presented.

Executive Function

Executive function is an umbrella term for a set of high-level cognitive processes which control and regulate behaviour, enabling purposeful, goal-directed behaviour and therefore particularly necessary when performing non-routine tasks (Gilbert & Burgess, 2008). It is important to note that executive function is a broad construct which can be hard to define and as a result multiple definitions exist; a comprehensive review listed 33 distinct definitions (Goldstein, Naglieri, Princiotta, & Otero, 2014). However, there is a general consensus that executive function is an overarching construct comprised of multiple distinct, but related, functions. Naglieri and Goldstein (2013) suggest that executive function is best represented as a single phenomenon which is made up of nine domains: attention, emotion regulation,

flexibility, inhibitory control, initiation, organisation, planning, self-monitoring, and working memory. The pre-frontal cortex has been shown to be instrumental in executive functioning, so acquired frontal damage, or disorders affecting this area of the brain, such as ADHD, will likely affect it (Hill, 2004). Amongst other areas of the brain, many recent imaging studies have demonstrated structural and functional abnormalities in the frontal cortex of individuals with ASD (e.g. Amaral, Schumann, & Nordahl, 2008).

ASD and Executive Functioning

Executive function difficulties are well-documented in ASD (Pennington & Ozonoff, 1996; Hill, 2004), however, the particular profile of deficit is not clearly established. A review of the area concluded that individuals with ASD show impairments in tasks of planning, flexibility and generativity, with relatively intact inhibition skills (Hill, 2004). Other studies have suggested that working memory is also a relative strength (e.g. Lopez, Lincoln, Ozonoff, & Lai, 2005). However, the picture is complex as executive function deficits in ASD are not consistently found in the laboratory and findings can be difficult to replicate. In contrast to Hill's (2004) position that cognitive flexibility is a clear deficit, another review looking specifically at cognitive flexibility in ASD found inconsistent results (Geurts, Corbett, & Solomon, 2009). Only studies using the Wisconsin Card Sorting Test (WCST) clearly reported deficits, whilst the findings of studies using other clinical neuropsychological measures of cognitive inflexibility were mixed and did not generally support a deficit. It has been argued that the WCST may not be a pure measure of cognitive flexibility but is affected by other factors, such as social-motivational ones and other executive functions including generativity and inhibition (Ozonoff, 1995).

Various factors are likely to contribute to the conflicting findings including participant variables such as the IQ and age of participants included in a study. General or verbal IQ is likely to confound performance on some traditional tasks of executive functioning (Hill, 2004). The types of task used and reported dependent

measures will also impact on the results (Geurts, de Vries, & van den Bergh, 2014). In particular, the way the task is administered has been shown to impact outcomes, for example, individuals with ASD often perform better on tasks administered on a computer rather than by a person (Kenworthy, Yerys, Anthony, & Wallace, 2008). Furthermore, it has been found that children with ASD show greater impairment on "open-ended" tasks of executive function; these are tasks that present a less structured situation and may therefore be more representative of real-life scenarios (White, Burgess, & Hill, 2009).

Given the heterogeneity of people meeting a diagnosis of ASD and the aforementioned difficulty identifying a universal cognitive profile, it is possible that there is not a common executive function impairment across people with ASD (White et al., 2009) and the inconsistent findings may reflect a genuine heterogeneity in the ASD population. Significant individual differences have been found in executive function abilities in young children with ASD (Pellicano, 2010). However, it is also possible that some performance-based tests of executive function underestimate the problems that individuals with ASD face in the real-world due to their inherently structured nature. Although Hill (2004) notes that greater perseveration is commonly observed in individuals with ASD in the lab (therefore indicating deficits in mental flexibility), this finding is not always replicated. Despite the mixed findings on tasks of cognitive flexibility, behavioural inflexibility is a core feature of ASD and perseverative behaviour is commonly seen in the daily lives of people with ASD. Geurts et al. (2009) suggest that more ecologically valid measures are required in order to resolve this observed discrepancy. The question that arises is whether traditional performance-based tasks of executive function have sufficient ecological validity to accurately detect executive function problems experienced by individuals with ASD in their daily lives.

The discrepancy between performance on traditional tasks of executive function and everyday impairment was first highlighted in the field of head injury,

with the observation that some patients with frontal lobe injury showed deficits in multitasking and marked impairment in their daily lives but minimal impairment, if any, on traditional tasks of executive function (Shallice & Burgess, 1991). This led to the development of test batteries, such as the Behavioural Assessment of Dysexecutive Syndrome (BADS; Wilson, Evans, Emslie, Alderman, & Burgess, 1998) which was designed to have greater ecological validity compared to traditional neuropsychological tests such as the Tower of London. Another way to measure executive function in a more ecologically valid way is to utilise questionnaire-based measures.

Behavior Rating Inventory of Executive Function (BRIEF)

One such questionnaire measure, the Behavior Rating Inventory of Executive Function (BRIEF), was developed with the intention of having increased ecological validity (Gioia, Isquith, Guy, & Kenworthy, 2000). It is a parent- or teacher-report measure for children and adolescents aged 5-18. The BRIEF consists of eight subdomains of executive function: the scales Inhibit, Shift, and Emotional Control which together form the Behavioral Regulation Index (BRI), and the scales Initiate, Working Memory, Plan/Organize, Organization of Materials, and Monitor which together form the Metacognition Index (MI). The BRI and MI are combined to obtain an overall Global Executive Composite (GEC). Early research into profiles of different clinical groups on the BRIEF by the measure's authors found that the scores of children with ASD were significantly elevated across all eight scales compared to controls. Children with ASD showed a significantly higher elevation than any other group on the Shift scale which taps into cognitive flexibility (Gioia, Isquith, Kenworthy, & Barton, 2002).

Aims

To date, no systematic review has been carried out looking at the use of the BRIEF with children with ASD. It is therefore not known how sensitive the BRIEF is at detecting executive function difficulties in children with ASD and how outcomes

on the BRIEF compare to performance-based neuropsychological tests. Therefore, the aim of this review is to investigate the degree of executive function difficulty as measured by the BRIEF in children with ASD compared to typically developing (TD) children. This study focuses on the BRIEF as it is a widely-used informant-report measure of executive function, both clinically and within research (Roth, Isquith, & Gioia, 2014). Furthermore, focusing on only one measure reduces the heterogeneity between studies and allows the results of individual studies to be combined using a meta-analysis. Through a meta-analysis, an estimated effect size can be calculated. This review focuses on the two indices and overall composite score derived from the BRIEF: the BRI, MI and GEC.

Method

Eligibility Criteria

The following inclusion and exclusion criteria were used to identify relevant studies.

Inclusion criteria:

- Clinical sample of children and/or young people with a diagnosis of ASD.
- Used a typically developing comparison group (studies using solely other clinical groups were not included as they were few in number).
- Administered the child-version of the BRIEF (parent or teacher) to measure executive function with both groups (i.e. not pre-school, adult or self-report versions of BRIEF).
- Published in English in a peer reviewed journal.

Exclusion Criteria:

- Sample with an intellectual disability (i.e. IQ of less than 70). This criterion was set to limit group heterogeneity, as intellectual disability is likely to impact upon executive functioning.

- Studies which did not present new data, including reviews and book chapters.

Search Strategy

A systematic search of the literature was conducted in November 2016 using the databases PsycINFO, Web of Science and Medline. Search terms focused on both ASD (ASD, Asperger*, autis*, PDD-NOS) and the BRIEF ("Behavior Rating Inventory of Executive Function", "Behaviour Rating Inventory of Executive Function"). For ASD, both keyword and thesaurus searches were carried out.

Study Selection

The titles and abstracts of the identified studies were reviewed against the inclusion and exclusion criteria. If, after this screening, the paper met the eligibility criteria or it remained unclear, the full article was accessed and further assessed against the inclusion and exclusion criteria. Figure 1 depicts the process of study selection in a flow diagram according to the "Preferred Reporting Items for Systematic Reviews and Meta-Analyses" (PRISMA) guidelines. The search resulted in 176 articles and after the removal of duplicates, 140 articles remained. Their titles and abstracts were assessed against the eligibility criteria which resulted in the exclusion of a further 112 articles, leaving 28 articles to be assessed in full. After accessing the full-texts, 23 of the studies were deemed to meet the eligibility criteria for this review.

This review focused on the two indices and overall composite score derived from the BRIEF: the BRI, MI and GEC. It was beyond the scope of this review to investigate all individual sub-scales of the BRIEF. Furthermore, scores on the individual sub-scales are rarely used as outcomes in studies and therefore not reported. Of the 23 studies meeting eligibility for this review, only five reported data on the BRIEF composite (GEC) and both indices (BRI and MI) in the article. Eleven of the studies reported some data from the BRIEF and seven studies reported no data from the BRIEF. Therefore, the authors of 18 studies were contacted to request

the necessary data from the BRIEF. Additional data were provided by eight of the authors contacted. Six of the 23 studies were not included in the analysis due to the unavailability of any outcome data from the BRIEF. One additional study was not included as its data were included in a later study which was also included in the review. This resulted in 16 final studies.

Quality Appraisal

The quality of each study in the final sample was assessed using a custom quality appraisal tool. The studies in this review were cross-sectional in design and many published tools are not designed for this type of study, the decision was therefore made to use an adapted tool. Widely used tools, such as the checklists developed by the Critical Appraisal Skills Programme (http://www.casp-uk.net/casp-tools-checklists) were used to inform the development of the tool. The tool contained items that focused on methodology and reporting. The use of a custom tool allowed important factors specific to this type of research to be included, for example, whether autism was screened in the control group. The quality appraisal tool contained 12 items (see Appendix A). The studies were carefully assessed against these criteria, resulting in a score between 0 and 12.

Data Extraction

Relevant data were extracted from all studies identified as meeting inclusion criteria using a coding sheet designed specifically for this review (see Appendix B). Regarding the BRIEF, outcome data were extracted on the GEC, BRI and MI. Due to resource constraints it was not possible for the process of quality appraisal or data extraction to be completed by two independent raters.

Meta-Analysis Procedure

A random effects meta-analysis of standard effect sizes (Cohen's *d*) was conducted using the "metan" command in Stata version 14 (StataCorp., 2015). Cohen's *d* is calculated as the difference between the means of the clinical and control groups, divided by the pooled standard deviations of the clinical and control

group. The studies were weighted according to sample size and pooled effect sizes were calculated. Cohen's (1988) guidelines were used which suggest 0.2 is considered a small effect size, 0.5 a medium effect size and 0.8 a large effect size.

Between-study heterogeneity was assessed using the chi-square test of heterogeneity as well as visual inspection of the forest plots. A significant chi-square statistic provides evidence that there is variation amongst effect estimates that are beyond chance. Where heterogeneity was significant, the I^2 statistic was used to quantify its magnitude (Higgins & Thompson, 2002). The I^2 statistic provides an estimate of the variability in results across studies that can be attributed to heterogeneity between the studies. An I^2 value of 0% indicates no observed heterogeneity beyond that expected from sampling error. It is suggested that 25% indicates low heterogeneity, 50% moderate heterogeneity and 75% high heterogeneity (Higgins, Thompson, Deeks, & Altman, 2003).

Sub-group meta-analyses were conducted to explore any observed heterogeneity. Analyses were carried out grouping the studies according to the following characteristics: 1) Higher quality (quality score of ≥ 9 which was median) v lower quality (quality score < 9); 2) IQ matched v not IQ matched; 3) Higher average IQ of ASD group (> 102, which was median) or lower average IQ (< 102).

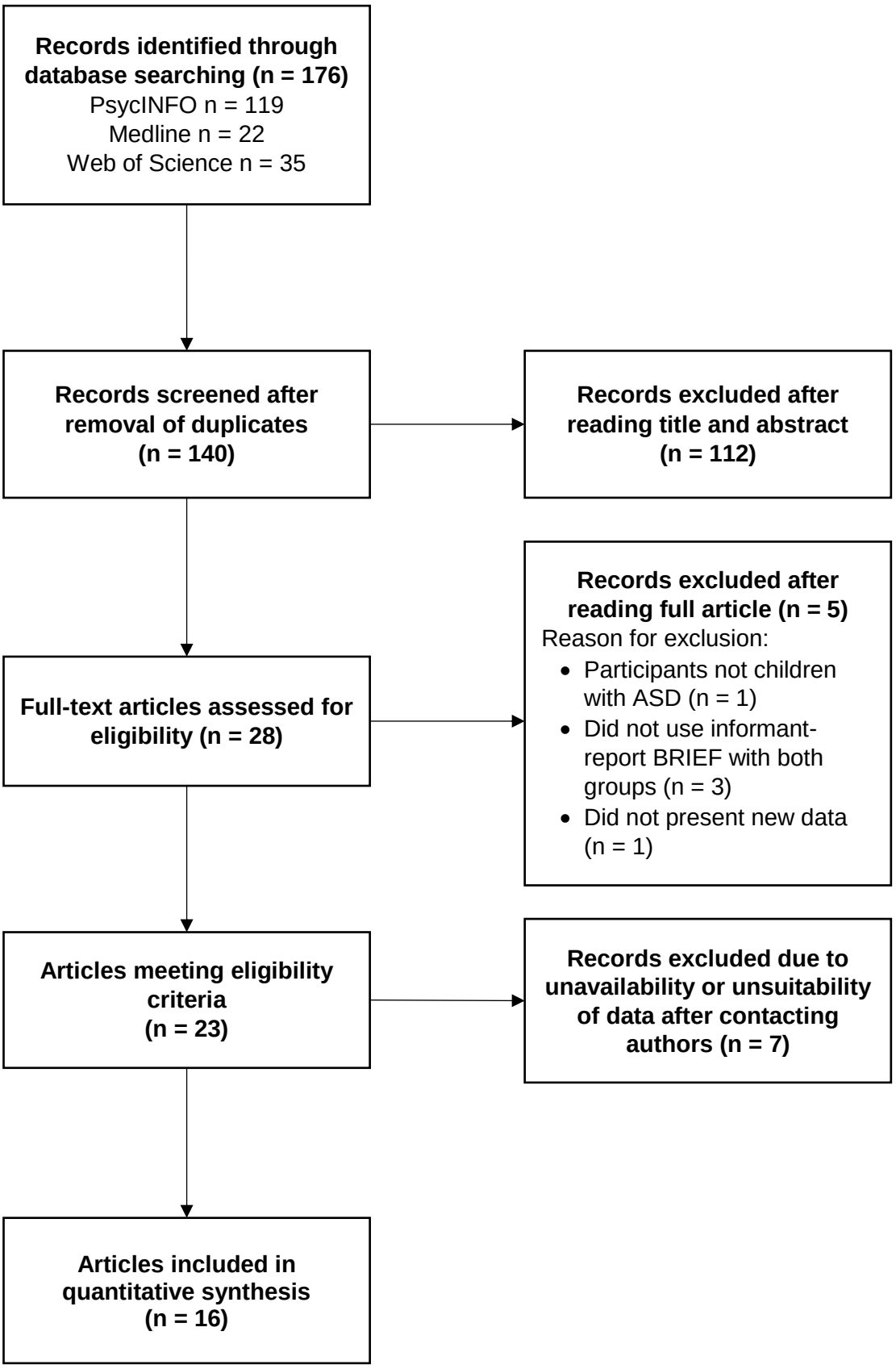

Figure 1. Flow Diagram of Study Selection.

Results

Corpus of Studies

The process of study selection and the reasons for exclusion are described in the methods and can be seen in Figure 1. Sixteen studies were included in the analyses, details of which can be seen in Table 1. Fifteen studies were included in the BRI and MI analyses, whilst 14 studies were included in the GEC analysis.

Quality Appraisal

Results of the quality appraisal can be seen in Table 2. The total scores range from 7 to 11 out of a possible 12, where higher scores indicate greater quality. Problems encountered with the studies' quality included: insufficient description of inclusion/exclusion criteria (n = 5, 31%); lack of screening of autism symptomatology in the control group (n = 9, 56%); insufficient matching of clinical and control groups on demographic variables (n = 11, 69%); insufficient description of all outcome variables (n = 1, 6%); insufficient reporting of results of all outcome variable (n = 7, 44%); and failure to report a consideration of power in sample size selection (n = 16, 100%).

Table 1

Summary of Studies Included in the Review and their Key Characteristics

Study	Total number of participants	% of sample male	Autism diagnoses Included	Age in years (M, SD)	IQ (M, SD)*	Group matching	BRIEF data available
Anthony, Kenworthy, Yerys, Jankowski, James, Harms…& Wallace (2013)	ASD = 109 TD = 76	ASD = 85% TD = 79%	Autism, AS, PDD-NOS	ASD = 12.7 (3.79) TD = 13.59 (3.85)	ASD = 111.56 (17.48) TD = 115.03 (11.72)	Matched on gender, age, FSIQ, SES	BRI, MI, GEC (all provided upon request)
Boyd, McBee, Holtzclaw, Baranek & Bodfish (2009)	ASD = 61 TD = 64	ASD = 93% TD = 95%	Autistic disorder, AS, PDD-NOS, ASD	ASD = 10.22 (2.78) TD = 11.75 (3.34)	ASD = 99.67 (17.30) TD = 111.16 (15.85)	Matched on gender and race; Significant differences in age and IQ.	BRI, MI, GEC
Chan, Cheung, Han, Sze, Leung, Man & To (2009)	ASD = 16 TD = 38	ASD = 88% TD = 74%	ASD	ASD = 10.54 (1.73) TD = 9.31 (2.2)	ASD = 96.75 (18.72) TD = 114.7 (16.6)	Matched on age; Significant difference in IQ (also completed matched sub-group analyses).	BRI, MI, GEC raw scores
de Vries & Geurts (2015)	ASD = 120 TD = 76	ASD = 90% TD = 57%	ASD	ASD = 10.2 (1.3) TD = 10.1 (1.2)	ASD = 110.9 (20.6) TD = 105.8 (18.4)	Matched on age and IQ; Significant	BRI, MI, GEC raw scores (BRI and MI

Study	Total number of participants	% of sample male	Autism diagnoses Included	Age in years (M, SD)	IQ (M, SD)*	Group matching	BRIEF data available
						difference in gender ratio	provided upon request)
Faja, Murias, Beauchaine & Dawson (2013)	ASD = 21 TD = 21	ASD = 71% TD = 71%	ASD	ASD = 6.83 (0.59) TD = 6.69 (0.63)	ASD = 104.0 (11.6) TD = 109.1 (7.2)	Matched on age, gender, IQ and SES	BRI, MI, GEC
Gioia, Isquith, Kenworthy & Barton (2002)	ASD = 54 TD = 208	ASD = 85% TD = 73%	HFA, AS, PDD-NOS	ASD = 10.8 (3) TD = 10.9 (3.3)	ASD = 102.7 (16.6) TD = not stated	Matched on age, sex, ethnicity and SES	BRI, MI, GEC (all provided upon request)
Hovik, Egeland, Isquith, Gioia, Skogli, Andersen & Øie (2014)	ASD = 34 TD = 50	ASD = 82% TD = 64%	AS, PDD-NOS	ASD = 11.9 (2.3) TD = 11.6 (2)	ASD = 98.2 (18.6) TD = 103.8 (12.9)	Matched on age and IQ	BRI, MI, GEC
Hutchins & Brien (2016)	ASD = 18 TD = 19	ASD = 83% TD = 79%	Autism, PDD-NOS, AS	ASD = 9.25 (1.55) TD = 8.67 (2.23)	ASD = 98.44 (20.66) TD = 112.84 (14.90)	Matched on age; Significant difference in IQ	BRI, MI, GEC (all provided upon request)

Study	Total number of participants	% of sample male	Autism diagnoses Included	Age in years (M, SD)	IQ (M, SD)*	Group matching	BRIEF data available
Irvine, Eigsti & Fein (2016)	ASD = 24 TD = 16	ASD = 88% TD = 88%	Autistic Disorder, PDD-NOS	ASD = 12.83 (2.4) TD = 13.33 (1.8)	VIQ: ASD = 103.5 (13.8); TD = 113.2 (12.9) NVIQ: ASD = 111.1 (14.7); TD = 115.1 (12.2)	Matched on age, gender, NVIQ; Significant difference in VIQ	BRI, MI, GEC (BRI and MI provided upon request)
Kloosterman, Kelley, Parker & Craig (2014)	ASD = 30 TD = 40	Male only	AS, PDD-NOS, HFA	ASD = 14.90 (2.25) TD = 14.23 (1.64)	ASD = 102.4 (13.28) TD = 109.43 (9.96)	Matched on age; Significant difference in IQ	GEC
Leung, Vogan, Powell, Anagnostou & Taylor (2016)	ASD = 70 TD = 71	ASD = 87% TD = 76%	ASD	ASD = 11.23 (2.47) TD = 11.69 (2.7)	ASD = 100.15 (15.54) TD = 110.09 (11.93)	Matched on age and gender; Significant difference in IQ	BRI and MI
Nair, Carper, Abbott, Chen, Solders, Nakutin, …& Müller (2015)	ASD = 37 TD = 38	ASD = 86% TD = 79%	ASD	ASD = 13.9 (2.6) TD = 13 (2.6)	NVIQ: ASD = 104.4 (16.9) TD = 107.5 (12.5)	Matched on age, gender, handedness, VIQ and NVIQ	BRI, MI, GEC (all provided upon request)

Study	Total number of participants	% of sample male	Autism diagnoses Included	Age in years (M, SD)	IQ (M, SD)*	Group matching	BRIEF data available
Semrud-Clikeman, Walkowiak, Wilkinson & Butcher (2010)	ASD = 15 TD = 32	ASD = 53% TD = 72%	AS	ASD = 10.6 (2.6) TD = 9.8 (2.1)	ASD = 100.8 (13) TD = 109.4 (10)	Matched on age and IQ	BRI, MI, GEC
Vanegas & Davidson (2015)	ASD = 24 (split into HFA = 13, AS =11) TD = 25	Not reported	HFA, AS	ASD = 9.70 (1.35) TD = 8.86 (1.09)	Non-verbal reasoning: HFA = 95.54 (15.41) AS = 105.91 (13.23) TD = 110.12 (14.59)	Matched on gender and ethnicity; Significant differences in age, non-verbal reasoning, core language abilities	BRI, MI, GEC (BRI and MI provided upon request)
Winsler, Abar, Feder, Schunn & Rubio (2007)	ASD = 33 TD = 28	ASD = 97% TD = 68%	HFA, AS, PDD-NOS	ASD = 11 (2.3) TD = 10.3 (3.2)	Not reported	Reports “no significant differences in child and family demographics”	BRI, MI, GEC raw scores (BRI and MI provided upon request)
Yerys, Wallace, Sokoloff, Shook, James & Kenworthy (2009)	ASD = 28 TD = 21	ASD = 71% TD = 62%	Autism, AS, PDD-NOS	ASD = 9.7 (2.12) TD = 10.3 (1.76)	ASD = 117.39 (18.68) TD = 116.24 (11.53)	Matched on age, SES, gender and IQ	BRI and MI

AS = Asperger Syndrome, ASD = Autism Spectrum Disorder, HFA = high-functioning autism, PDD-NOS = Pervasive Developmental Disorder Not Otherwise Specified, SES = Socioeconomic Status, TD = typically developing. *Full-scale IQ scores (FSIQ) unless stated otherwise. VIQ = verbal IQ, NVIQ = non-verbal IQ.

Table 2

Quality Appraisal Results

Study	Quality Appraisal Item												Quality Appraisal Total
	1	2	3	4	5	6	7	8	9	10	11	12	
Anthony et al. (2013)	✓	✓	✓	✓	X	✓	✓	X	X	✓	✓	✓	9
Boyd et al. (2009)	✓	✓	✓	✓	✓	X	✓	✓	X	✓	✓	✓	10
Chan et al. (2009)	✓	✓	X	✓	X	X	✓	✓	X	✓	✓	✓	8
de Vries & Geurts (2015)	✓	✓	✓	✓	✓	X	✓	X	X	✓	✓	✓	9
Faja et al. (2013)	✓	✓	✓	✓	✓	✓	✓	✓	X	✓	✓	✓	11
Gioia et al. (2002)	✓	✓	X	✓	X	X	✓	X	X	✓	✓	✓	7
Hovik et al. (2014)	✓	✓	✓	✓	✓	X	✓	✓	X	✓	✓	✓	10

Study	Quality Appraisal Item												Quality Appraisal Total
	1	2	3	4	5	6	7	8	9	10	11	12	
Hutchins & Brien (2016)	✓	✓	X	✓	X	X	✓	X	X	✓	✓	✓	7
Irvine et al. (2016)	✓	✓	✓	✓	✓	✓	✓	✓	X	✓	✓	✓	11
Kloosterman et al. (2014)	✓	✓	X	✓	X	X	✓	X	X	✓	✓	✓	7
Leung et al. (2016)	✓	✓	✓	✓	X	X	✓	✓	X	✓	✓	✓	9
Nair et al. (2015)	✓	✓	✓	✓	X	✓	X	X	X	✓	✓	✓	8
Semrud-Clikeman et al. 2010)	✓	✓	✓	✓	✓	X	✓	✓	X	✓	✓	✓	10
Vanegas & Davidson (2015)	✓	✓	X	✓	✓	X	✓	✓	X	✓	✓	✓	9
Winsler et al. (2007)	✓	✓	✓	✓	X	X	✓	X	X	✓	✓	✓	8

Study	Quality Appraisal Item												Quality Appraisal Total
	1	2	3	4	5	6	7	8	9	10	11	12	
Yerys et al. (2009)	✓	✓	✓	✓	X	✓	✓	✓	X	✓	✓	✓	10

✓ indicates yes
x indicates no or unknown (i.e. not reported)

Item 1 = Are the aims/objectives of the study clearly described?
Item 2 = Is the study design clearly described and appropriate?
Item 3 = Were the clinical and control groups recruited in appropriate way (with clearly defined inclusion and exclusion criteria)?
Item 4 = Did the clinical group have a diagnosed autism spectrum disorder meeting diagnostic criteria?
Item 5 = Was autism symptomatology screened in the control group using an appropriate method?
Item 6 = Were the clinical and control groups matched on age, gender and FSIQ?
Item 7 = Are the outcome variable(s) clearly defined and were they measured using appropriate tools and procedures?
Item 8 = Does the study report the results of all outcome variable(s) in full?
Item 9 = Was the sample size appropriate / did the study have sufficient power?
Item 10 = Are the analytic methods described and appropriate?
Item 11 = Are the main findings of the study clearly described?
Item 12 = Are the conclusions supported by the results?

Main Effects Meta-Analysis

Behavioral Regulation Index (BRI). Table 3 presents descriptive data for the BRI for each study and the standardised mean differences. The results show an overall significant positive effect with a pooled standardised mean difference of 2.19, 95% CI [1.91, 2.46]. Children with ASD showed significantly more difficulties on the BRI than typically developing children and the effect size is large. This is also illustrated with a forest plot in Figure 2. Heterogeneity between studies was significant (X^2 = 49.73, *df* = 14, $p < .001$). The I^2 statistic of 71.8% (variation in SMD attributable to heterogeneity) indicates a moderate-high level of heterogeneity.

Table 3

BRI: Descriptive Statistics and Meta-Analysis Results

Study	ASD N	ASD M (SD)	TD N	TD M (SD)	SMD	LCI	UCI	Weight
Anthony	101	64.72 (11.38)	71	43.85 (7.63)	2.09	1.71	2.46	8.33
Boyd	60	67.57 (11.29)	64	43.34 (5.66)	2.74	2.25	3.23	7.5
Chan (raw score)	16	60.75 (9.83)	38	47.74 (9.58)	1.35	0.71	1.99	6.44
de Vries (raw score)	120	62.2 (9.2)	74	37.1 (6.5)	3.03	2.61	3.45	8.02
Faja	19	61.4 (8.5)	21	50.1 (8.4)	1.34	0.65	2.03	6.09
Gioia	54	67.69 (12.8)	208	49.94 (7.63)	1.99	1.64	2.33	8.54
Hovik	34	64 (13.2)	50	40 (4)	2.69	2.09	3.29	6.72
Hutchins	18	69.89 (9.23)	19	48.84 (12.67)	1.89	1.11	2.67	5.49
Irvine	24	67.58 (10.48)	16	40.31 (4.54)	3.16	2.21	4.11	4.53

Study	ASD N	ASD M (SD)	TD N	TD M (SD)	SMD	LCI	UCI	Weight
Leung	70	70.15 (11.88)	71	45.61 (7.37)	2.49	2.04	2.93	7.87
Nair	24	68.79 (11.29)	23	44.87 (10.43)	2.2	1.47	2.93	5.82
Semrud-Clikeman	15	68.2 (10.6)	32	51.5 (9.5)	1.69	0.99	2.4	5.98
Vanegas	24	67.33 (10.77)	25	50.64 (9.44)	1.65	1	2.3	6.35
Winsler (raw score)	33	61.16 (9.8)	28	37.47 (8.65)	2.55	1.87	3.23	6.14
Yerys	28	59.92 (11.89)	21	41.62 (6.55)	1.83	1.16	2.51	6.18
Pooled SMD	-	-	-	-	2.19	1.91	2.46	100

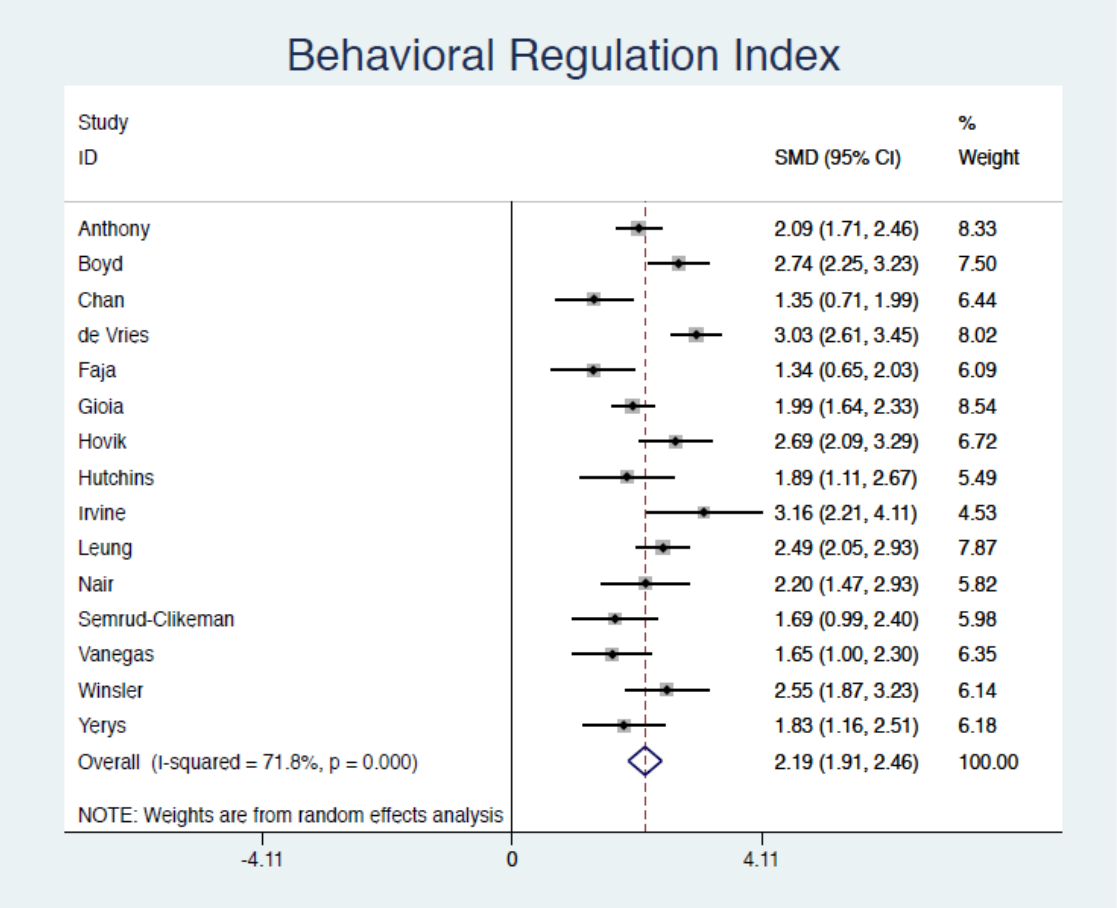

Figure 2. BRI Forest Plot.

Metacognition Index (MI). Table 4 presents descriptive data for the MI for each study and the standardised mean differences. The results show an overall significant positive effect with a pooled standardised mean difference of 2.04, 95% CI [1.78, 2.3]. Children with ASD showed significantly more difficulties on the MI than typically developing children and the effect size is large. This is also illustrated with a forest plot in Figure 3. Heterogeneity between studies was significant (X^2 = 45.72, *df* = 14, $p < .001$). The I^2 statistic of 69.4% indicates a moderate-high level of heterogeneity.

Table 4

MI: Descriptive Statistics and Meta-Analysis Results

Study	ASD N	ASD M (SD)	TD N	TD M (SD)	SMD	LCI	UCI	Weight
Anthony	100	64.79 (9.86)	71	46.14 (9.14)	1.95	1.58	2.32	8.5
Boyd	57	67.25 (9.64)	63	44.49 (8.14)	2.56	2.08	3.05	7.55
Chan (raw scores)	16	103.47 (11.59)	38	84.37 (14.34)	1.4	0.76	2.05	6.28
de Vries (raw scores)	120	102.6 (12.7)	74	67.6 (13.7)	2.67	2.28	3.07	8.29
Faja	19	66.5 (6.4)	21	54.6 (6.7)	1.81	1.07	2.56	5.56
Gioia	54	68.8 (10.71)	208	53.2 (8.78)	1.69	1.36	2.03	8.78
Hovik	34	62 (10.8)	50	40 (5.2)	2.77	2.16	3.38	6.55
Hutchins	18	68.39 (9.83)	19	49.05 (10.85)	1.87	1.09	2.64	5.31
Irvine	24	64 (9.31)	16	42.31 (7.04)	2.56	1.7	3.41	4.84
Leung	70	66.42 (9.12)	71	47.62 (8.75)	2.1	1.69	2.52	8.15
Nair	24	66.08 (10.13)	23	44.3 (10.29)	2.13	1.41	2.86	5.7
Semrud-Clikeman	15	70 (11.9)	32	52 (11)	1.59	0.9	2.29	5.89
Vanegas	24	63.67 (10.27)	25	51.72 (11.65)	1.09	0.48	1.69	6.6
Winsler (raw scores)	33	104.35 (12.45)	28	69.38 (13.62)	2.69	1.99	3.39	5.86
Yerys	28	58.19 (10.57)	21	43.29 (5.55)	1.69	1.03	2.36	6.14
Pooled SMD	-	-	-	-	2.04	1.78	2.3	100

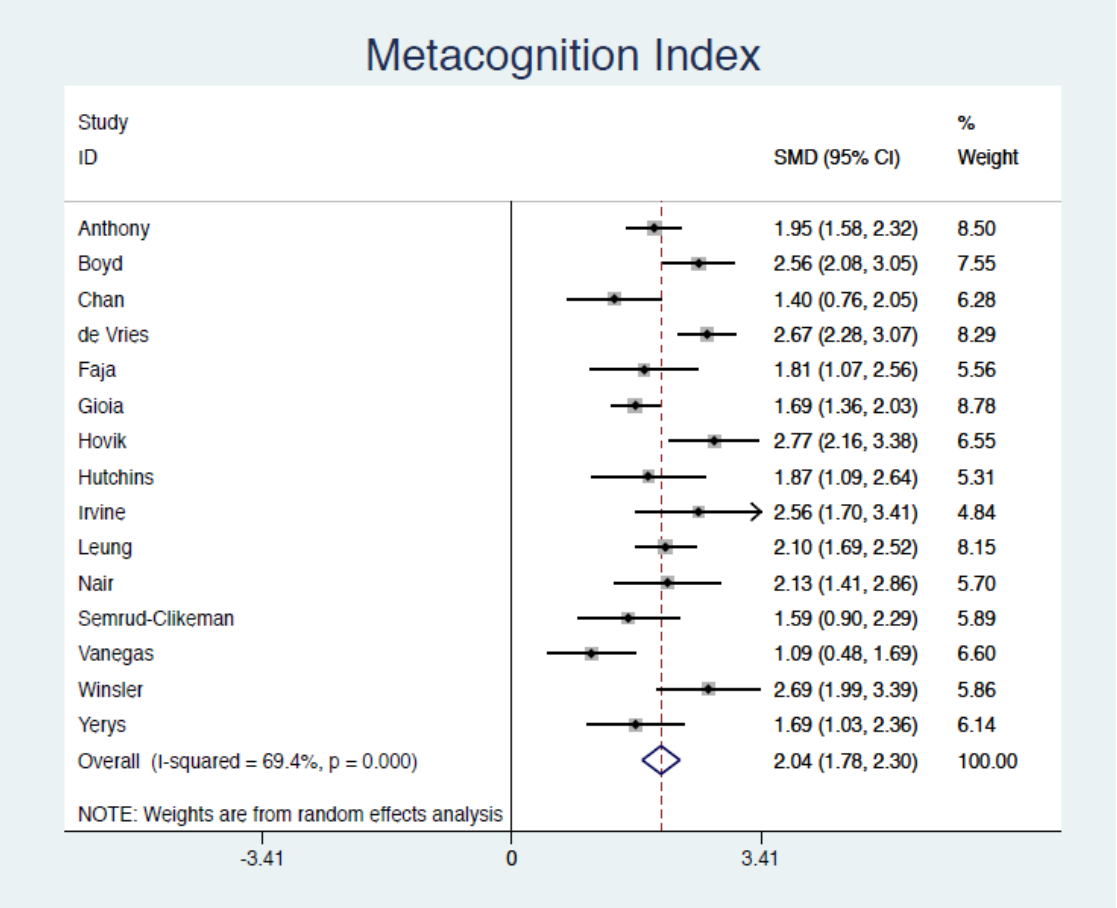

Figure 3. MI Forest Plot.

Global Executive Composite. Table 5 presents descriptive data for the GEC for each study and the standardised mean differences. The results show an overall significant positive effect with a pooled standardised mean difference of 2.38, 95% CI [2.06, 2.71]. Children with ASD showed significantly more difficulties on the GEC than typically developing children and the effect size is large. This is also illustrated with a forest plot in Figure 4. Heterogeneity between studies was significant (X^2 = 55.37, *df* = 13, $p < .001$). The I^2 statistic of 76.5% indicates a high level of heterogeneity.

Table 5

GEC: Descriptive Statistics and Meta-Analysis Results

Study	ASD N	ASD M (SD)	TD N	TD M (SD)	SMD	LCI	UCI	Weight
Anthony	100	66.06 (10.42)	71	44.92 (8.4)	2.19	1.81	2.58	8.68
Boyd	57	69.02 (9.71)	63	43.56 (6.23)	3.15	2.62	3.69	7.77
Chan (raw scores)	16	164.22 (17.56)	38	132.11 (20.68)	1.62	0.96	2.28	7.01
de Vries (raw scores)	120	164.9 (18.4)	74	104.7 (18)	3.3	2.86	3.74	8.37
Faja	19	66.1 (6.1)	21	53.2 (6.6)	2.03	1.26	2.8	6.36
Gioia	54	68.94 (10.67)	208	51.66 (8.09)	1.99	1.65	2.34	8.88
Hovik	34	64 (11.6)	50	39 (4.9)	3.02	2.38	3.66	7.18
Hutchins	18	70.22 (8.79)	19	47.42 (10.94)	2.29	1.45	3.13	5.97
Irvine	24	66.53 (9.29)	16	40.94 (6.17)	3.11	2.17	4.06	5.4
Kloosterman	30	72.1 (10.65)	40	51.87 (9.96)	1.97	1.39	2.55	7.53
Nair	24	68.54 (10.18)	23	44.91 (11.6)	2.17	1.44	2.89	6.62
Semrud-Clikeman	15	70.6 (8.8)	32	52.1 (7.7)	2.3	1.52	3.07	6.33
Vanegas	24	66.29 (10.14)	25	51.4 (10.79)	1.42	0.79	2.05	7.21
Winsler (raw scores)	33	165.51 (20.34)	28	106.84 (21.28)	2.82	2.11	3.54	6.68
Pooled SMD	-	-	-	-	2.38	2.06	2.71	100

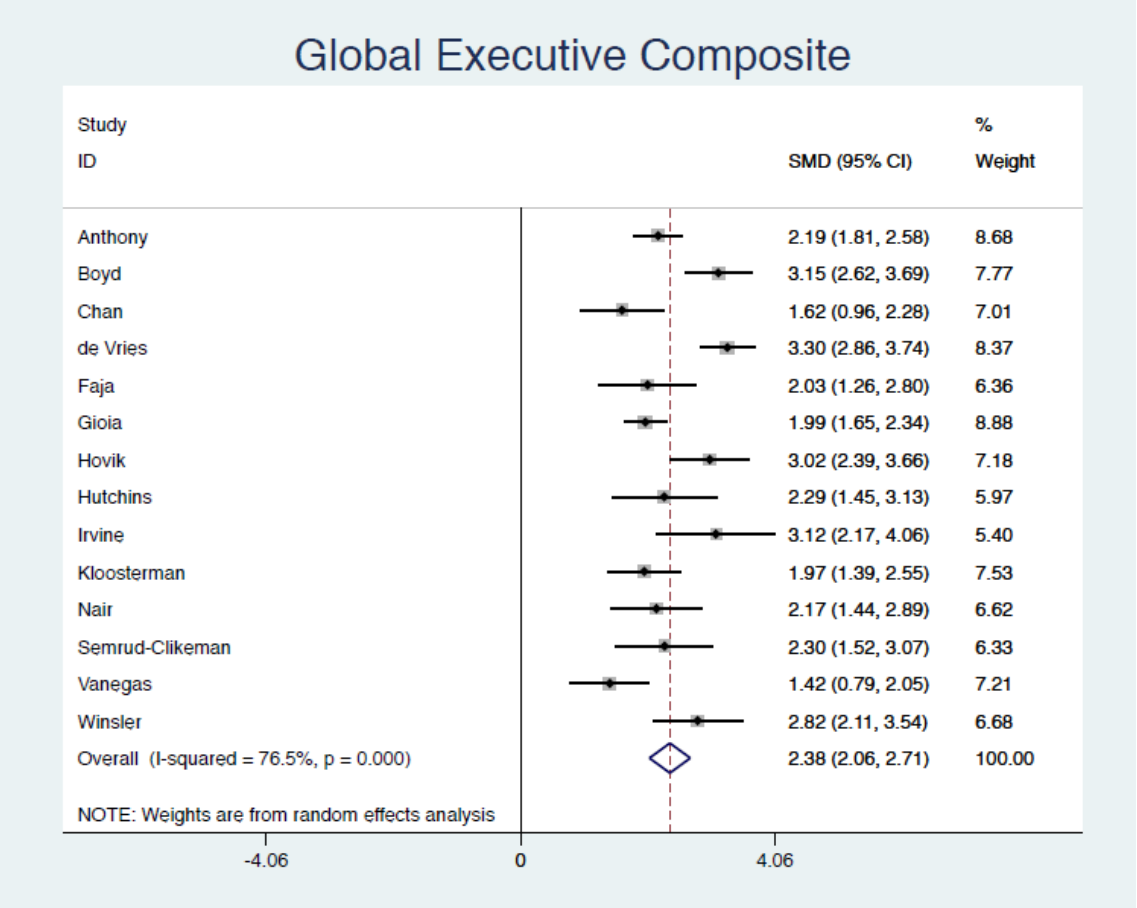

Figure 4. GEC Forest Plot.

Publication Bias

Publication bias was assessed visually using a funnel plot (see Figures 5, 6 and 7) and more formally using the Egger's test (Egger, Smith, Schneider, & Minder, 1997). The results did not provide evidence of publication bias in respect of the BRI (p = .53), MI (p = .93) or GEC (p = .85).

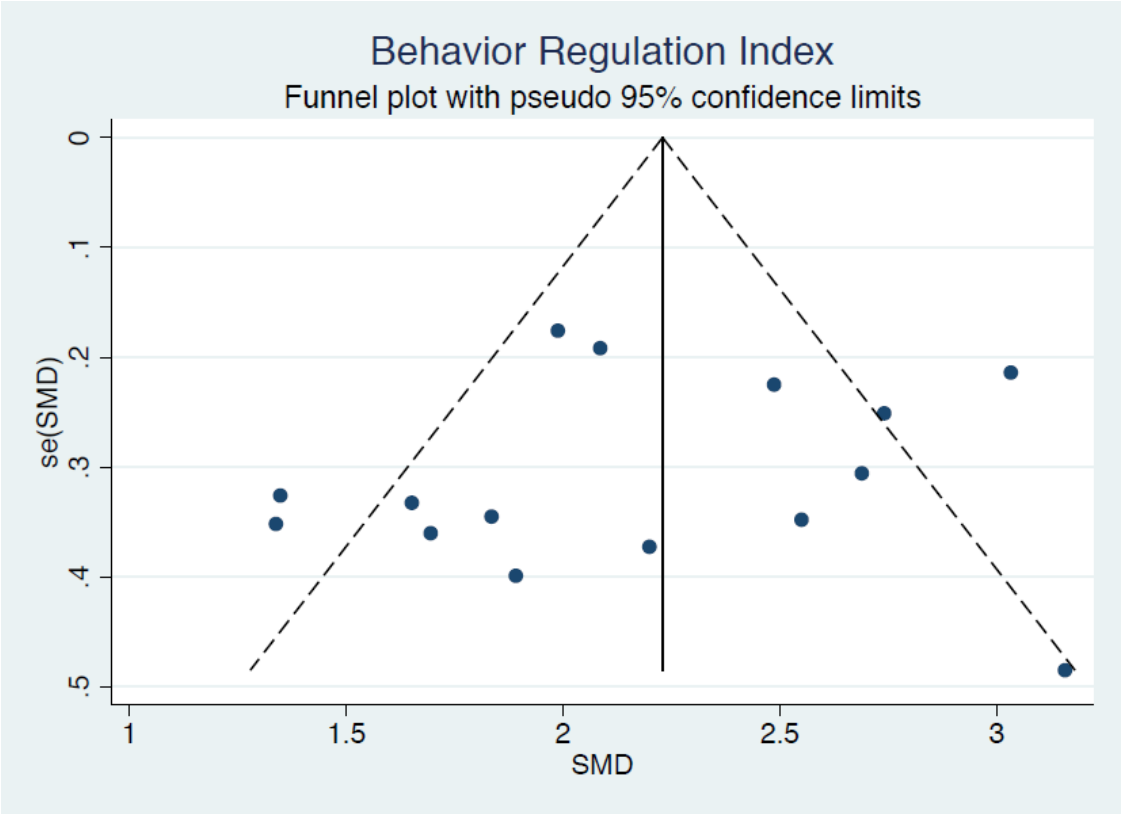

Figure 5. BRI Funnel Plot.

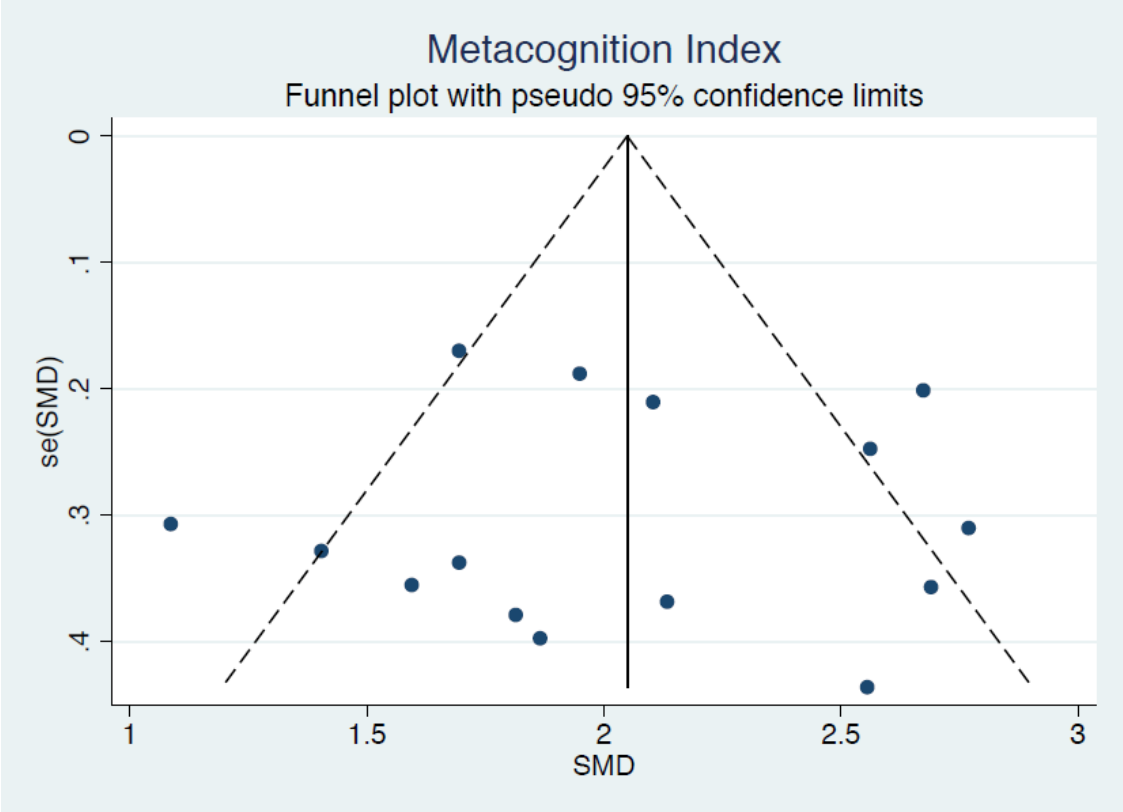

Figure 6. MI Funnel Plot.

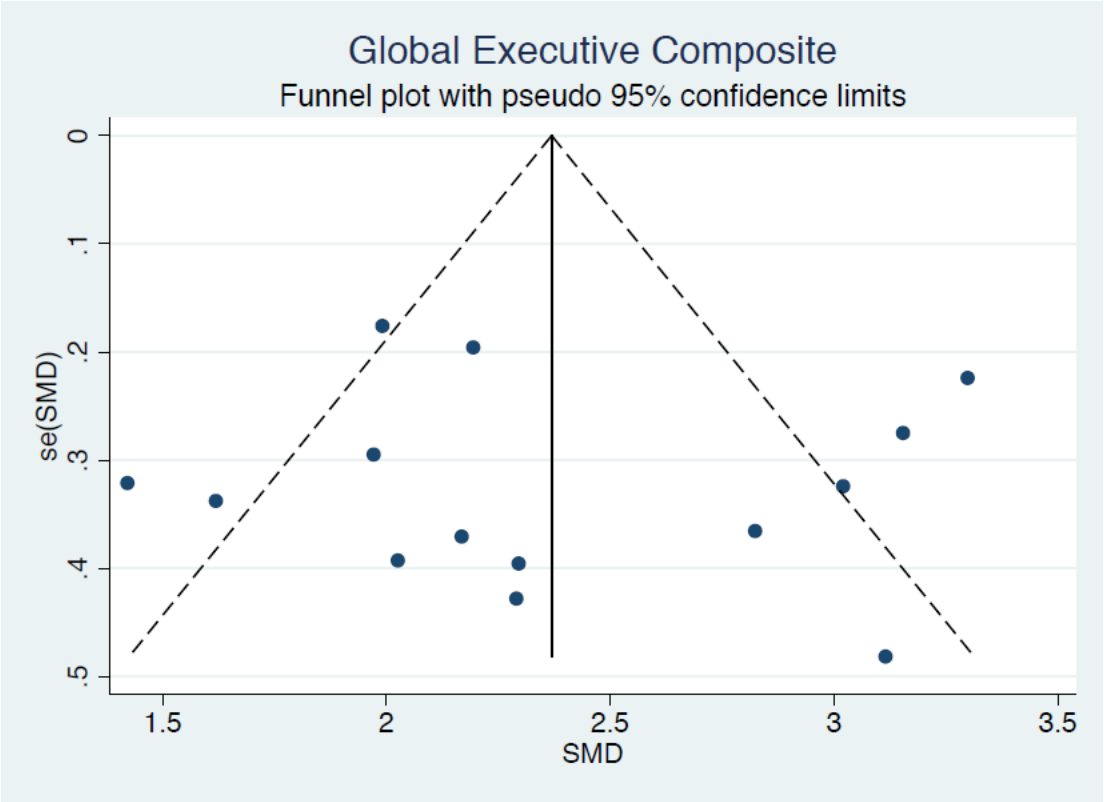

Figure 7. GEC Funnel Plot.

Sub-Group Analyses

Due to the high levels of heterogeneity, moderators of the effect were investigated. Study quality, average IQ and whether the groups were matched on IQ were investigated as possible moderators. Each of the three BRIEF outcomes were compared for the following groups:

1. Higher quality (9 or above) v lower quality (below 9).

2. IQ matched groups v non IQ matched groups.

3. Higher average IQ (>102 which was median) or lower average IQ (<102).

The results can be seen in Tables 6, 7 and 8. The pooled estimates for the two groups are not significant for any of the BRIEF outcomes or any of the factors, indicated by the overlapping confidence intervals. This means that, based on these exploratory (and underpowered analyses) we cannot attribute the variance between the studies to any of the three factors investigated.

Table 6

BRI: Results of Sub-Group Analyses

Analysis	N of studies	Pooled SMD	LCI	UCI	Weight	Heterogeneity $\chi 2$	*p*	I^2
All	15	2.19	1.91	2.46	100	49.73	<.001	71.8%
Quality								
High	10	2.28	1.92	2.64	67.57	37.07	<.001	75.7%
Low	5	1.98	1.63	2.34	32.43	6.84	.145	41.5%
IQ matching								
Yes	7	2.16	1.71	2.61	47.14	22.69	<.001	77.5%
No	8	2.21	1.84	2.57	52.86	22.76	.002	69.2%
Mean IQ								
Higher	8	2.25	1.86	2.65	53.65	28.74	<.001	75.6%
Lower	7	2.11	1.69	2.53	46.35	20.92	.002	71.3%

Table 7

MI: Results of Sub-Group Analyses

Analysis	N of studies	Pooled SMD	LCI	UCI	Weight	Heterogeneity $\chi 2$	Heterogeneity *p*	Heterogeneity I^2
All	15	2.04	1.79	2.3	100	45.72	<.001	69.4%
Quality								
High	10	2.1	1.77	2.42	68.06	32.66	<.001	72.4%
Low	5	1.92	1.51	2.32	31.94	8.81	.066	54.6%
IQ matching								
Yes	7	2.13	1.77	2.48	46.64	16.88	.01	64.5%
No	8	1.98	1.6	2.35	53.36	25.87	.001	72.9%
Mean IQ								
Higher	8	2.13	1.80	2.45	53.68	20.29	.005	65.5%
Lower	7	1.93	1.48	2.38	46.32	25.13	<.001	76.1%

Table 8

GEC: Results of Sub-Group Analyses

Analysis	N of studies	Pooled SMD	LCI	UCI	Weight	Heterogeneity		
						χ2	*p*	I^2
All	14	2.38	2.06	2.71	100	55.37	<.001	76.5%
Quality								
High	8	2.57	2.09	3.05	57.3	37.12	<.000	81.1%
Low	6	2.09	1.81	2.37	42.7	6.69	.245	25.2%
IQ matching								
Yes	6	2.53	2.06	3.01	43.54	19.77	.001	74.7%
No	8	2.27	1.83	2.73	56.46	29.47	<.001	76.2%
Mean IQ								
Higher	8	2.43	2.03	2.83	58.53	29.46	<.001	76.2%
Lower	6	2.31	1.69	2.93	41.47	25.9	<.001	80.7%

Discussion

Summary of Main Findings

The aim of this review was to collate studies which used the BRIEF with children with ASD and a TD control group, in order to assess the extent of informant-reported executive function deficits in children with ASD. This review focused on the BRIEF as it is a widely-used measure and no systematic review has previously been carried out looking at its use with children with ASD. Furthermore, focusing on a specific measure allowed the use of a meta-analysis, enabling a quantitative synthesis of the studies and therefore arguably a more rigorous review.

Sixteen studies met the inclusion criteria for this review and provided sufficient data to be included in the analysis. The main findings reveal a significant

difference between informant-reported executive function deficits in children with ASD and typically developing children. As anticipated, the direction of effect demonstrates that children with ASD are reported to have greater executive function deficits compared to typically developing children. What is most striking about these analyses is that the pooled effects are so large, for both indices on the BRIEF (the BRI and MI) and the overall composite (GEC). All three effects sizes were greater than 2, substantially greater than Cohen's (1988) proposed guide of 0.8 for a large effect. The largest effect found was for the GEC, however all three effects were similar in magnitude with overlapping confidence intervals, meaning they cannot be interpreted as significantly different. This review found no evidence of publication bias in the included studies, suggesting there is no threat to validity posed by a lack of publication.

A large amount of heterogeneity was found amongst the included studies for all three outcomes on the BRIEF, meaning that the effect sizes of the individual studies varied greatly. Exploratory analyses were therefore conducted to investigate factors that might moderate the effect, including the average IQ of the study's participants and whether the participant groups were matched on IQ. Neither was found to explain the observed heterogeneity, suggesting that differences in IQ did not account for the variation seen amongst the studies. However, as these analyses were exploratory and the number of studies in each group was small, they are underpowered and therefore unlikely to detect effects that may exist. The relationship between executive function and IQ is a complex one, with the extent to which they are distinct or related concepts a source of on-going debate (Blijd-Hoogewys, Bezemer, & van Geert, 2014).

Quality of the included studies was assessed using a customised tool and overall quality was moderate to high, with studies scoring between 7 and 11 out of a possible 12. Study quality was also investigated as a moderator, but did not reliably account for the observed heterogeneity between studies, although again this

analysis was likely underpowered. Although not significant, the trend was in the direction of studies of higher quality demonstrating greater effect sizes. This suggests that the higher quality studies may have had more sensitivity to the effect.

Relevance to Existing Evidence Base

These findings add support to previous narrative reviews which also find that children with ASD do show deficits in executive function (e.g. Hill, 2004; Pennington & Ozonoff, 1996). The current findings do not, however, reflect any of the conflicting findings in the neuropsychological literature which have been highlighted in recent reviews (Geurts et al., 2009; Hill, 2004). Every study in this review found a substantial effect of diagnosis (i.e. having ASD) on reported executive function deficits using the BRIEF. The smallest estimated effect size of any study in this review was 1.09 (MI in Vanegas & Davidson, 2005) which is still considered a large effect. The effect sizes found in this review are therefore greater and more consistent than those found in reviews of direct testing. A recent meta-analysis of neuropsychological measures of executive function in children with high-functioning ASD reported pooled effect sizes (*g*) ranging from 0.41 (inhibition) to 0.67 (verbal working memory) (Lai et al., 2016). The current review therefore highlights a discrepancy between studies using questionnaire-based informant-report measures and performance-based neuropsychological tests. This lack of concordance has been demonstrated in many studies of executive function (Isquith, Roth, Kenworthy, & Gioia, 2014) and several recent systematic reviews have demonstrated this with the ASD population (Demetriou et al., 2017; Gardiner, Hutchinson, Müller, Kerns, & Iarocci, 207). Since the present review was completed, a meta-analysis has been published which included 235 studies comparing children and adults with ASD to neurotypical controls. Although mainly focused on neuropsychological tests, it included consideration of BRIEF scores and provided strong evidence for a discrepancy between questionnaire measures and methods involving direct testing (Demetriou et al., 2017). They found that questionnaire measures resulted in a

much larger effect size (g = 1.84) than psychometric tests or experimental tasks. This effect size is broadly in line with the present finding on the BRIEF. After excluding questionnaire outcomes from the analyses, Demetriou et al. (2017) found the revised effect size was only moderate (g = 0.49).

Traditional neuropsychological tests attempt to explicitly assess specific executive functions, however, there are many factors that may limit their ecological validity and generalisability (Gioia et al., 2002). It has been argued that neuropsychological tests alone are not adequate to assess executive function because they artificially fractionate a complex and integrated executive function system (Burgess, 1997). The structured nature and explicit instructions of a typical neuropsychological assessment can reduce the demands on the executive system and the examiner may act as an external executive control. It has been suggested that the performance-based tasks in which children with ASD perform worse are those which have higher working memory load, are more unpredictable, and/or lack explicit task instructions (Geurts et al., 2014). These factors are highly likely to impact on the observed degree of day-to-day executive function difficulties seen in children and adults with ASD.

The BRIEF is a measure of the behavioural manifestation of executive function difficulties that children experience day-to-day, designed to have greater ecological validity. Proponents of the BRIEF argue that rating scales provide useful information clinically and can be particularly useful in developing interventions and measuring outcomes (Isquith et al., 2014). However, it is important to also hold in mind the possibility that the BRIEF over-estimates executive function difficulties in children with ASD. Anecdotal evidence of the research team is suggestive of a ceiling effect when using the BRIEF with children with ASD in clinical services. Further examination of the findings presented in this review may support that. For instance, Boyd et al. (2009) found a mean T-score of 69.02 on the GEC for the ASD group, which is almost at the 98th percentile, indicating that half of their sample

scored above this. There are several possible factors that could contribute to high scores seen on the BRIEF. Firstly, deficits as reported by parents may be higher, due to conscious and unconscious reporting biases. Secondly, the BRIEF may be tapping into something more than executive function deficits and impacted upon by autistic symptomatology, that is unrelated to executive function. For example, the question “interrupts other” which is designed to measure inhibition difficulties, is likely to be impacted upon by social communication deficits. Therefore, it is possible the BRIEF is confounded by factors other than executive function which mean it is assessing day-to-day impairment above and beyond executive functioning.

Limitations of Review

One limitation of this review and potential source of bias, is that it was not possible to utilise two independent raters. The use of two independent raters is recommended when assessing study inclusion, extracting data and when appraising a study’s quality, however, due to the time and resource constraints of this review that was not possible. Secondly, a more comprehensive quality appraisal tool may also have been useful. Given that the results indicated a trend towards higher quality studies demonstrating greater effect sizes, a more nuanced quality appraisal tool with a greater range of possible scores may have shed more light on this relationship.

Although there was no evidence of publication bias, a source of potential bias is the lack of inclusion of the six studies which met the inclusion criteria but did not provide data from the BRIEF. This highlights the importance of full reporting of outcome data in the literature in order to enable rigorous review and replication. However, given the word-limits and associated constraints of journals, it is apparent why this is not always feasible.

In this review, the developmental trajectory of executive function in children with ASD was not considered. Age was not included as a possible moderator in this review as the included studies did not vary greatly in terms of their mean age. There

is evidence to suggest, however, that age does impact on executive functioning. One study found that the pattern of executive function deficits as measured by the BRIEF varied according to age of children and adolescents aged between 6 and 18 (van den Bergh, Scheeren, Begeer, Koot, & Geurts, 2014).

Clinical Implications and Further Research

This review demonstrates that parents are reporting significant day-to-day executive function difficulties in their children with ASD on the BRIEF. This review also highlights the discrepancy between outcomes on the BRIEF and the evidence base utilising neuropsychological performance-based tests. Performance-based measures aim to assess specific executive function domains in isolation, while rating scales, such as the BRIEF, arguably assess the application of those skills in everyday life (Isquith, Roth, & Gioia, 2013). Therefore, the use of both types of measure in combination will provide a more comprehensive clinical picture of an individual's strengths and weaknesses.

This review focused on the BRIEF and the benefit of focusing only on one measure was that this allowed the results to be combined using a quantitative method. Focusing only on studies that used the BRIEF minimised heterogeneity in outcomes between the studies and therefore the threat to the validity of the meta-analysis. Although it is a widely-used informant-report measure, other measures of executive function exist, such as the DEX-C which is part of the BADS-C (Emslie, Wilson, Burden, Nimmo-Smith, & Wilson, 2003) and the Comprehensive Executive Function Inventory (CEFI; Naglieri & Goldstein, 2013). It would be informative to investigate whether other measures of executive function yield effect sizes as large as those generated through using the BRIEF.

Future research could also review the individual scales on the BRIEF to examine the profile of executive function difficulties of children with ASD. It would be interesting to see whether the relative strengths and weaknesses on the BRIEF

reflect those found on performance-based neuropsychological tasks, for example the conclusion that planning is generally impaired but inhibition is not (Hill, 2004).

Future research might also further explore the correlates of BRIEF outcomes in children with ASD. It would be useful to explore the degree to which scores on the BRIEF are impacted by other factors such as social communication deficits. Evidence suggests that scores on the BRIEF are correlated with social function, and that scores on the MI in particular may be predictive of social communication deficits in the ASD population, however the directionality of this relationship is unclear (e.g. Leung et al., 2016).

Conclusions

In conclusion, this review clearly indicates that parents of children with ASD are reporting a significant degree of day-to-day executive function impairment on the BRIEF as compared to parents of typically developing children. The effect size is very large which sits in contrast to evidence from some performance-based executive function measures. This review therefore highlights the discrepancy between different assessment measures and emphasises the importance of not relying solely on one mode of assessment which has been demonstrated in other recent reviews. The BRIEF provides useful information about how a child with ASD is managing in their day-to-day life which will be valuable in guiding interventions. However, the degree to which it is measuring only executive functioning or is impacted by other factors, such as broader ASD symptomatology, requires further investigation.

References

References marked with an asterisk indicate studies included in the meta-analysis.

Amaral, D. G., Schumann, C. M., & Nordahl, C. W. (2008). Neuroanatomy of autism. *Trends in Neurosciences, 31*(3), 137-145. doi:10.1016/j.tins.2007.12.005

American Psychiatric Association (2013). *Diagnostic and statistical manual of mental disorders (5th ed.).* Arlington, VA: American Psychiatric Association.

*Anthony, L. G., Kenworthy, L., Yerys, B. E., Jankowski, K. F., James, J. D., Harms, M. B., ... Wallace, G. L. (2013). Interests in high-functioning autism are more intense, interfering, and idiosyncratic than those in neurotypical development. *Development and Psychopathology, 25*(03), 643-652. doi:10.1017/S0954579413000072

Baird, G., Simonoff, E., Pickles, A., Chandler, S., Loucas, T., Meldrum, D., & Charman, T. (2006). Prevalence of disorders of the autism spectrum in a population cohort of children in South Thames: The special needs and autism project (SNAP). *The Lancet*, *368*(9531), 210-215. doi:10.1016/S0140-6736(06)69041-7

Baron-Cohen, S., Leslie, A. M., & Frith, U. (1985). Does the autistic child have a "theory of mind"? *Cognition, 21*(1), 37–46. doi:10.1016/0010-0277(85)90022-8

Blijd-Hoogewys, E. M. A., Bezemer, M. L., & van Geert, P. L. C. (2014). Executive functioning in children with ASD: An analysis of the BRIEF. *Journal of Autism and Developmental Disorders, 44*(12), 3089-3100. doi:10.1007/s10803-014-2176-9

*Boyd, B. A., McBee, M., Holtzclaw, T., Baranek, G. T., & Bodfish, J. W. (2009). Relationships among repetitive behaviors, sensory features, and executive functions in high functioning autism. *Research in Autism Spectrum Disorders, 3*(4), 959-966. doi:10.1016/j.rasd.2009.05.003

Brugha, T., Cooper, S. A., McManus, S., Purdon, S., Smith, J., Scott, F. J., … Tyrer, F. (2012). *Estimating the prevalence of autism spectrum conditions in adults: Extending the 2007 adult psychiatric morbidity survey.* Retrieved from NHS Digital: http://content.digital.nhs.uk/pubs/autism11

Burgess, P. W. (1997). Theory and methodology in executive function research. In P. Rabbitt (Ed.), *Methodology of frontal and executive function* (pp. 81–116). Hove, UK: Psychology Press.

Chakrabarti, S., & Fombonne, E. (2005). Pervasive developmental disorders in preschool children: Confirmation of high prevalence. *American Journal of Psychiatry, 162*(6), 1133-1141. doi:10.1176/appi.ajp.162.6.1133

*Chan, A. S., Cheung, M. C., Han, Y. M. Y., Sze, S. L., Leung, W. W., Man, H. S., & To, C. Y. (2009). Executive function deficits and neural discordance in children with autism spectrum disorders. *Clinical Neurophysiology, 120*(6), 1107-1115. doi:10.1016/j.clinph.2009.04.002

Cohen, J. (1988). *Statistical power analysis for the behavioral sciences*. Hillsdale, NJ: Lawrence Erlbaum Associates.

Damasio, A. R., & Maurer, R. G. (1978). A neurological model for childhood autism. *Archives of Neurology, 35*(12), 777–786. doi:10.1001/archneur.1978.00500360001001

*de Vries, M., & Geurts, H. (2015). Influence of autism traits and executive functioning on quality of life in children with an autism spectrum disorder. *Journal of Autism and Developmental Disorders, 45*(9), 2734-2743. doi:10.1007/s10803-015-2438-1

Demetriou, E. A., Lampit, A., Quintana, D. S., Naismith, S. L., Song, Y. J. C., Pye, J. E., ... Guastella, A. J. (2017). Autism spectrum disorders: A meta-analysis of executive function. *Molecular Psychiatry*, 1-17. doi:10.1038/mp.2017.75

Egger, M., Smith, G. D., Schneider, M., & Minder, C. (1997). Bias in meta-analysis detected by a simple, graphical test. *British Medical Journal, 315*, 629-634. doi:10.1136/bmj.315.7109.629

Emslie, H., Wilson, F. C., Burden, V., Nimmo-Smith, I., & Wilson, B. A. (2003). Behavioural assessment of the dysexecutive syndrome for children (BADS-C). London, UK: Harcourt Assessment/The Psychological Corporation.

*Faja, S., Murias, M., Beauchaine, T. P., & Dawson, G. (2013). Reward-based decision making and electrodermal responding by young children with autism spectrum disorders during a gambling task. *Autism Research, 6*(6), 494-505. doi:10.1002/aur.1307

Gardiner, E., Hutchison, S. M., Müller, U., Kerns, K. A., & Iarocci, G. (2017). Assessment of executive function in young children with and without ASD using parent ratings and computerized tasks of executive function. *The Clinical Neuropsychologist*, 1-23. doi:10.1080/13854046.2017.1290139

Geurts, H. M., Corbett, B., & Solomon, M. (2009). The paradox of cognitive flexibility in autism. *Trends in Cognitive Sciences, 13*(2), 74-82. doi:10.1016/j.tics.2008.11.006

Geurts, H. M., de Vries, M., & van den Bergh, S. F. W. M. (2014). Executive functioning theory and autism. In S. Goldstein & J. A. Naglieri (Eds.), *Handbook of executive functioning* (pp. 121-141). New York: Springer.

Gilbert, S. J., & Burgess, P. W. (2008). Executive function. *Current Biology, 18*(3), 110-114. doi:10.1016/j.cub.2007.12.014

Gioia, G. A., Isquith, P. K., Guy, S. C., & Kenworthy, L. (2000). Test review: Behavior rating inventory of executive function. *Child Neuropsychology, 6*(3), 235-238. doi:10.1076/chin.6.3.235.3152

*Gioia, G. A., Isquith, P. K., Kenworthy, L., & Barton, R. M. (2002). Profiles of everyday executive function in acquired and developmental disorders. *Child Neuropsychology, 8*(2), 121-137. doi: 10.1076/chin.8.2.121.8727

Goldstein, S., Naglieri, J. A., Princiotta, D., & Otero, T. M. (2014). Introduction: A history of executive functioning as a theoretical and clinical construct. In S. Goldstein & J. A. Naglieri (Eds.), *Handbook of executive functioning* (pp. 3-12). New York: Springer. doi:10.1007/978-1-4614-8106-5_1

Happé, F., & Frith, U. (2006). The weak coherence account: Detail-focused cognitive style in autism spectrum disorders. *Journal of Autism and Developmental Disorders, 36*(1), 5-25. doi:10.1007/s10803-005-0039-0

Higgins, J. P. T., & Thompson, S. G. (2002). Quantifying heterogeneity in a meta-analysis. *Statistics in Medicine, 21*(11), 1539-1558. doi: 10.1002/sim.1186

Higgins, J. P. T., Thompson, S. G., Deeks, J. J., & Altman, D. G. (2003). Measuring inconsistency in meta-analyses. *British Medical Journal, 327*, 557-560. doi:10.1136/bmj.327.7414.557

Hill, E. L. (2004). Evaluating the theory of executive dysfunction in autism. *Developmental Review, 24*(2), 189-233. doi:10.1016/j.dr.2004.01.001

*Hovik, K. T., Egeland, J., Isquith, P. K., Gioia, G., Skogli, E. W., Andersen, P. N., & Øie, M. (2014). Distinct patterns of everyday executive function problems distinguish children with tourette syndrome from children with ADHD or autism spectrum disorders. *Journal of Attention Disorders*, 1-13. doi:10.1177/1087054714550336

Hughes, C., Russell, J., & Robbins, T. W. (1994). Evidence for executive dysfunction in autism. *Neuropsychologia, 32*(4), 477-492. doi:10.1016/0028-3932(94)90092-2

*Hutchins, T. L., & Brien, A. (2016). Conversational topic moderates social attention in autism spectrum disorder: Talking about emotions is like driving in a snowstorm. *Research in Autism Spectrum Disorders, 26,* 99-110. doi:10.1016/j.rasd.2016.03.006

*Irvine, C. A., Eigsti, I. M., & Fein, D. A. (2016). Uh, um, and autism: Filler disfluencies as pragmatic markers in adolescents with optimal outcomes

from autism spectrum disorder. *Journal of Autism and Developmental Disorders, 46*(3), 1061-1070. doi:10.1007/s10803-015-2651-y

Isquith, P. K., Roth, R. M., & Gioia, G. (2013). Contribution of rating scales to the assessment of executive functions. *Applied Neuropsychology: Child, 2*(2), 125-132. doi:10.1080/21622965.2013.748389

Isquith, P. K., Roth, R. M., Kenworthy, L., & Gioia, G. (2014). Contribution of rating scales to intervention for executive dysfunction. *Applied Neuropsychology: Child, 3*(3), 197-204. doi:10.1080/21622965.2013.870014

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. doi:10.1007/s11065-008-9077-7

*Kloosterman, P. H., Kelley, E. A., Parker, J. D. A., & Craig, W. M. (2014). Executive functioning as a predictor of peer victimization in adolescents with and without an autism spectrum disorder. *Research in Autism Spectrum Disorders, 8*(3), 244-254. doi:10.1016/j.rasd.2013.12.006

Lai, C. L. E., Lau, Z., Lui, S. S. Y., Lok, E., Tam, V., Chan, Q., ... Cheung, E. F. C. (2016). Meta-analysis of neuropsychological measures of executive functioning in children and adolescents with high-functioning autism spectrum disorder. *Autism Research, 10*(5), 911–939. doi:10.1002/aur.1723

*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. doi:10.1080/09297049.2015.1005066

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 & Adolescent Psychiatry, 56*(6), 466-474. doi:10.1016/j.jaac.2017.03.013

Lopez, B. R., Lincoln, A. J., Ozonoff, S., & Lai, Z. (2005). Examining the relationship between executive functions and restricted, repetitive symptoms of autistic disorder. *Journal of Autism and Developmental Disorders, 35*(4), 445-460. doi:10.1007/s10803-005-5035-x

Maurer, R. G., & Damasio, A. R. (1982). Childhood autism from the point of view of behavioral neurology. *Journal of Autism and Developmental Disorders, 12*(2), 195-205. doi:10.1007/BF01531309

Naglieri, J., & Goldstein, S. (2013). *Comprehensive executive functioning inventory technical manual.* Toronto, Canada: Multi-Health Systems.

*Nair, A., Carper, R. A., Abbott, A. E., Chen, C. P., Solders, S., Nakutin, S., ... Müller, R. A. (2015). Regional specificity of aberrant thalamocortical connectivity in autism. *Human Brain Mapping, 36*(11), 4497-4511. doi:10.1002/hbm.22938

Ozonoff, S. (1995). Reliability and validity of the Wisconsin Card Sorting Test in studies of autism. *Neuropsychology, 9*(4), 491-500. doi:10.1037/0894-4105.9.4.491

Pellicano, E. (2010). Individual differences in executive function and central coherence predict developmental changes in theory of mind in autism. *Developmental Psychology, 46*(2), 530-544. doi:10.1037/a0018287

Pennington, B. F., & Ozonoff, S. (1996). Executive functions and developmental psychopathology. *Journal of Child Psychology and Psychiatry, 37*(1), 51–87. doi:10.1111/j.1469-7610.1996.tb01380.x

Roth, R. M., Isquith, P. K., & Gioia, G. A. (2014). Assessment of executive functioning using the Behavior Rating Inventory of Executive Function (BRIEF). In S. Goldstein & J. A. Naglieri (Eds.), *Handbook of executive functioning* (pp. 301-331). New York: Springer. doi:10.1007/978-1-4614-8106-5_18

*Semrud-Clikeman, M., Walkowiak, J., Wilkinson, A., & Butcher, B. (2010). Executive functioning in children with Asperger syndrome, ADHD-combined type, ADHD-predominately inattentive type, and controls. *Journal of Autism and Developmental Disorders, 40*(8), 1017-1027. doi:10.1007/s10803-010-0951-9

Shallice, T., & Burgess, P. W. (1991). Deficits in strategy application following frontal lobe damage in man. *Brain, 114*(2), 727-741. doi:10.1093/brain/114.2.727

StataCorp. (2015). Stata statistical software: Release 14. College Station, TX: StataCorp LP.

Turner, M. (1999). Annotation: Repetitive behaviour in autism: A review of psychological research. *Journal of Child Psychology and Psychiatry, 40*(6), 839-849. doi:10.1111/1469-7610.00502

van den Bergh, S. F. W. M., Scheeren, A. M., Begeer, S., Koot, H. M., & Geurts, H. M. (2014). Age related differences of executive functioning problems in everyday life of children and adolescents in the autism spectrum. *Journal of Autism and Developmental Disorders, 44*(8), 1959-1971. doi:10.1007/s10803-014-2071-4

*Vanegas, S. B., & Davidson, D. (2015). Investigating distinct and related contributions of weak central coherence, executive dysfunction, and systemizing theories to the cognitive profiles of children with autism spectrum disorders and typically developing children. *Research in Autism Spectrum Disorders, 11*, 77-92. doi:10.1016/j.rasd.2014.12.005

White, S. J., Burgess, P. W., & Hill, E. L. (2009). Impairments on “open-ended” executive function tests in autism. *Autism Research, 2*(3), 138-147. doi:10.1002/aur.78

Wilson, B. A., Evans, J. J., Emslie, H., Alderman, N., & Burgess, P. (1998). The development of an ecologically valid test for assessing patients with a

dysexecutive syndrome. *Neuropsychological Rehabilitation, 8*(3), 213-228. doi:10.1080/713755570

*Winsler, A., Abar, B., Feder, M. A., Schunn, C. D., & Rubio, D. A. (2007). Private speech and executive functioning among high-functioning children with autistic spectrum disorders. *Journal of Autism and Developmental Disorders, 37*(9), 1617-1635. doi:10.1007/s10803-006-0294-8

*Yerys, B. E., Wallace, G. L., Sokoloff, J. L., Shook, D. A., James, J. D., & Kenworthy, L. (2009). Attention deficit/hyperactivity disorder symptoms moderate cognition and behavior in children with autism spectrum disorders. *Autism Research, 2*(6), 322-333. doi:10.1002/aur.103

Part 2: Empirical Research Paper

Developing and Piloting a New Parent-Report Measure of Executive Function for Children with Autism Spectrum Disorder (ASD)

Abstract

Aims: The aim of this study was to develop and pilot a new parent-report measure of executive function for use with children with Autism Spectrum Disorder (ASD). This measure was intended to have good ecological validity and be sensitive to the day-to-day executive function difficulties experienced by children with ASD.

Method: This study is reported in two parts; Study 1 focuses on the development of the measure and Study 2 focuses on its piloting. In Study 1, parents and professionals were interviewed about the nature of executive function difficulties in children with ASD. These interviews were analysed using qualitative methods and this directly informed the development of the measure as a way of maximising its ecological validity. The measure was then piloted online with parents of children with ASD (N = 44) and parents of TD children (N = 55), enabling an examination of its psychometric properties.

Results: Study 1 presents a thematic analysis of the nature of executive function difficulties in children with ASD and the development of the new measure. The new measure is named the A-POD (Autism Planning and Organisation Difficulties) and initially contained 48 items. Piloting of the A-POD revealed a large difference between the ASD and TD groups, indicating that children with ASD are reported to have greater levels of executive dysfunction. The A-POD demonstrated good test-retest reliability and internal consistency. The items with the weakest reliability were removed and the final version of the A-POD contained 38 items. Preliminary positive evidence of its construct validity is presented. However, the distribution of the ASD group on the A-POD is suggestive of a ceiling effect.

Conclusion: The findings indicate that day-to-day executive function difficulties are substantial and widespread amongst children with ASD. The A-POD is a promising measure of these difficulties, demonstrating encouraging psychometric properties. One main limitation of the A-POD is the observed ceiling effect. Further research into its properties is required.

Introduction

Autism Spectrum Disorder

Autism Spectrum Disorder (ASD) is a life-long neurodevelopmental condition characterised by difficulties with social interaction and communication in combination with restricted, repetitive patterns of behaviour, interests or activities (American Psychiatric Association, 2013). ASD is a condition present from early childhood, although it may not be diagnosed until later into childhood or even adulthood. The prevalence of ASD in the population is estimated to be about 1% (Baird et al., 2006; Brugha et al., 2012) and around 30-50% of individuals with ASD also have an intellectual disability (Baird et al., 2006; Chakrabarti & Fombonne, 2005).

Numerous theories have been proposed over the last few decades that attempt to explain ASD in terms of cognitive strengths and weaknesses. These include the weak central coherence theory (e.g. Happé & Frith, 2006), the theory of mind account (e.g. Baron-Cohen, Leslie, & Frith, 1985) and the empathizing–systemizing theory (Baron-Cohen, 2009). Another prominent account, the executive dysfunction theory, proposes that ASD can be accounted for by deficits in executive function and the areas of the brain associated with these functions (i.e. the frontal cortex). This theory seeks to explain both the social and non-social (i.e. restrictive and repetitive behaviour) symptoms of ASD as the consequences of executive dysfunction and atypical functioning in the frontal lobes (Damasio & Maurer, 1978). Before exploring evidence of executive dysfunction in ASD, background to the concept of executive function will be presented.

Executive Function

Executive function is an umbrella term for a set of cognitive functions that control and regulate behaviour, which include working memory, inhibition, cognitive flexibility, monitoring, planning, and generativity (Kenworthy, Yerys, Antony, & Wallace, 2008). There is no universally agreed definition of executive function,

hence, numerous distinct theories of executive function exist (see Goldstein, Naglieri, Princiotta, & Otero (2014) for a review). There is consensus, however, that executive functions involve top-down regulation of lower processes according to the demands of the task (Gilbert & Burgess, 2008). Executive function is therefore important in purposeful, goal-directed behaviour and necessary when performing non-routine tasks, as opposed to when carrying out rehearsed behaviours (Gilbert & Burgess, 2008). Historically, the frontal lobes (in particular the pre-frontal cortex) were thought to regulate executive functions, as patients with acquired frontal damage showed impairment in executive function (e.g. Luria, 1966). It has now been shown that both frontal and non-frontal brain regions are required for optimal executive functioning (Alvarez & Emory, 2006).

Executive Dysfunction Theory of ASD

Early evidence for the executive dysfunction account of ASD came from the observation that patients with frontal lobe brain damage showed similar social difficulties and behaviours to individuals with ASD (Damsaio & Maurer, 1978). This included the observation that both groups had difficulty with switching between tasks and following social rules. This led to a neurological model which suggested that ASD was caused by impairment in the frontal lobes and resultant executive dysfunction. Amongst other areas of the brain, many recent imaging studies have demonstrated structural and functional abnormalities in the frontal cortex in individuals with ASD (e.g. Amaral, Schumann, & Nordahl, 2008). The executive dysfunction account has particular appeal in providing an explanation for some of the non-social features of ASD, such as the rigidity, desire for sameness, and repetitive behaviours that are part of its core features (Lopez, Lincoln, Ozonoff, & Lai, 2005). However, the model faces several criticisms as an explanation of ASD, including the non-specificity of executive function problems to ASD as other disorders such as ADHD also show executive function deficits (e.g. Pennington & Ozonoff, 1996). Another problem for the executive dysfunction account of ASD,

which will now be presented, is the inconsistencies in the research field regarding the profile of executive dysfunction observed in ASD.

ASD and Executive Functioning

Executive function difficulties are well-documented amongst individuals with ASD (e.g. Pennington & Ozonoff, 1996). An influential narrative review of the field found that individuals with ASD generally show impairments in the areas of planning, cognitive flexibility and generativity, with relatively intact inhibition skills (Hill, 2004). An earlier review supports this, demonstrating that the effect sizes for deficits in cognitive flexibility and planning amongst participants with ASD, typically measured using the Wisconsin Card Sorting Test (WCST) and Tower of Hanoi, were greater than for any other executive function measure amongst several other developmental disorders (Pennington & Ozonoff, 1996). Furthermore, inhibition and working memory were found to be relatively intact. However, as Hill's (2004) review highlights, the picture is complex and many studies present seemingly conflicting findings. Executive function deficits in ASD are not consistently found in experimental studies and findings are sometimes difficult to replicate. A more recent review of only cognitive flexibility, did not conclude that it was a core deficit in ASD (Geurts, Corbett, & Solomon, 2009), highlighting that only studies using the WCST reported clear deficits. Furthermore, studies looking at executive function difficulties on an individual level report executive function difficulties in some, but not all, children with ASD (Geurts, Sinzig, Booth, & Happé, 2014).

Executive function deficits are not universally found in individuals with ASD and effect sizes using performance-based measures are often relatively modest. It might therefore be reasonable to conclude that executive function deficits are not a universal feature of ASD, especially given that heterogeneity is an inherent feature of this condition (Mandy, Murin, & Skuse, 2015). However, anecdotally, many family members, clinicians, and teachers describe how individuals with ASD experience daily executive functioning difficulties in their lives (Kenworthy et al., 2008).

Clinically, problems with planning and organisation amongst children with ASD are reported to be substantial and widespread. Behavioural inflexibility is a core feature of ASD and perseverative behaviour (which may be linked to cognitive inflexibility) is widely seen. This therefore leads to a consideration of possible reasons for the observed discrepancies within the field.

Numerous factors have been identified which are likely to impact upon the varied outcomes of studies investigating executive functioning in the ASD population, including participant variables (e.g. IQ) and experimental factors (e.g. human vs computer administration) (Hill, 2004; Kenworthy et al., 2008). A demonstration of this can be seen in the planning literature. Planning was thought to be a robust deficit in ASD, however, Kenworthy et al. (2008) did not find consistent planning deficits in studies that were published since Hill's (2004) review. An examination of these findings at the task-level revealed that children with ASD performed poorly on standard forms of the Tower of London/Hanoi tasks when compared to controls. Many of the recent studies, however, had used a computerised version of the tower tasks and failed to find deficits in the ASD population (e.g. Goldberg et al., 2005). Removing the social element of administration may improve the performance of participants with ASD, suggesting it is the socially-mediated elements of these tasks which increase the difficulty. An additional factor concerns the degree to which the task presented is structured in nature. It has been found that children with ASD show greater impairment on "open-ended" or "ill-structured" tasks of executive function. These are tasks that present a less structured situation and require spontaneous strategy generation, and may therefore be more representative of real-life scenarios that rely on executive functioning (White, Burgess, & Hill, 2009).

Ecological Validity

The conflicting findings and observed discrepancies have raised questions about how we measure executive functioning in ASD and have highlighted potential

limitations with the use of more traditional neuropsychological tests. A discrepancy between performance on traditional neuropsychological tests of executive function and everyday functioning was first highlighted with patients with frontal lobe head injury. It was observed that some patients with frontal lobe injuries showed minimal impairment on traditional tests of executive function, despite showing marked everyday impairment and deficits in multi-tasking (Shallice & Burgess, 1991). This observation prompted the development of new tests designed to have greater ecological validity, such as the Behavioural Assessment of Dysexecutive Syndrome (BADS; Wilson, Evans, Emslie, Alderman, & Burgess, 1998). Ecological validity refers to the degree to which task performance corresponds to real world performance (Chaytor & Schmitter-Edgecombe, 2003). Ecological validity can be considered as "representativeness" (the extent to which a clinical test corresponds to a situation encountered outside the experimental setting) and the "generalisability" of test results (the degree to which performance on the test will be predictive of functioning outside the experimental setting). One way to increase the ecological validity of a measure is to use a bottom-up method of development, ensuring it is closely grounded in participant data. This method was used by Paul Burgess and colleagues when developing the BADS.

Questionnaire-Based Measures

An alternative way to measure neuropsychological constructs, such as executive function, is through the use of questionnaire-based measures. One such measure, the Behavior Rating Inventory of Executive Function (BRIEF) was developed by Gioia and colleagues (Gioia, Isquith, Guy, & Kenworthy, 2000) and was designed to have greater ecological validity than traditional performance-based measures. It is an informant-report measure for children and adolescents aged 5-18. The BRIEF consists of eight subdomains of executive function: the Inhibit, Shift, and Emotional Control scales which together form the Behavioral Regulation Index (BRI), and the Initiate, Working Memory, Plan/Organize, Organization of Materials,

and Monitor scales which together form the Metacognition Index (MI). The BRI and MI are combined to obtain an overall Global Executive Composite (GEC). Early research into the profiles of different clinical groups on the BRIEF by the measure's developers found that children with ASD were significantly elevated across all eight scales compared to controls. Children with ASD showed a significantly higher elevation than any other clinical group on the Shift scale which taps into cognitive flexibility (Gioia, Isquith, Kenworthy, & Barton, 2002). It is argued that rating scales provide useful information in guiding assessment and intervention and capture executive function difficulties as they manifest day-to-day (Isquith, Roth, & Gioia, 2013).

Limitations of the BRIEF in ASD

Since its development, the BRIEF has been widely used with children and adolescents with ASD. As shown in the Literature Review of this thesis, studies that have compared executive function as measured by the BRIEF in children with ASD and typically developing (TD) children have demonstrated large effect sizes (GEC: *d* = 2.38). Another recent meta-analysis provides similar evidence, demonstrating that effect sizes of studies using a questionnaire format, primarily the BRIEF, were very large (*g* = 1.84) and much greater than studies using performance-based tests (Demetriou et al., 2017). In fact, the clinical experience of the research team is suggestive of a ceiling effect when using the BRIEF with an ASD population, with many children in local Child and Adolescent Mental Health Services (CAMHS) scoring at the very top end of the scale. This observation is supported by an examination of the studies included in the literature review of this thesis; all 13 studies reporting *T* scores on the GEC, reported a mean score for the ASD group at or above the 92nd percentile.

These observations therefore lead us to question whether the BRIEF can accurately assess executive function problems in an ASD population. It is possible that the BRIEF is measuring everyday executive function impairment but we are

seeing a ceiling effect in the ASD population, meaning it cannot capture the variance observed amongst children who all display a fairly high degree of impairment. Another possibility is that having ASD inflates scores on the BRIEF, perhaps because some items on the BRIEF are actually tapping autistic symptomatology in this population. For example, the question he/she "does not realize that certain actions bother others", although designed to be tapping into the executive ability of self-monitoring, is clearly also impacted by the ability to mentalise or use theory of mind which is a characteristic deficit in ASD (Baron-Cohen et al., 1985). It has been suggested that the BRIEF is a better measure of general impairment and behavioural disruption than specifically executive function (McAuley, Chen, Goos, Schachar, & Crosbie, 2010).

Aims

Although the executive dysfunction account has short-comings as an explanatory model of ASD, it is widely accepted that executive function plays a role in some of the observed symptoms in ASD. The study of executive function in ASD remains worthwhile as better understanding the pattern of deficits will inform our understanding of the day-to-day difficulties that people with ASD experience. Executive function is particularly important as it has been found to correlate to adaptive functioning levels in children and young people (Gilotty, Kenworthy, Sirian, Black & Wagner, 2002; Pugliese et al., 2015). Therefore, the ability to accurately measure executive function in children with ASD in an ecologically valid way is essential for research and clinical purposes.

The present study aimed to develop a parent-report questionnaire measure of executive function with the following specific properties:

- The measure aims to be able to capture the everyday executive function difficulties experienced by children with ASD.
- The measure's ecological validity will be maximised through a bottom-up method of development, adhering closely to data provided by parents of

children with ASD and professionals working in the field. This will be achieved by collecting and analysing qualitative data on the nature of executive function difficulties in order to inform the development of the measure.

- The measure aims to be sensitive to the potential variability of executive functioning in the ASD population and aims to avoid a ceiling effect.
- The measure aims to be easy to administer and shorter than the BRIEF.

Study Overview

This study was carried out over two phases, which will be reported as Study 1, the development of the measure, and Study 2, piloting of the measure.

Study 1: Measure Development

The aim of Study 1 was to design a new parent-report measure of executive function difficulties in children with ASD which would be developed using methods intended to maximise its ecological validity. The process of developing a measurement scale set out by Streiner, Norman and Cairney (2015) was used throughout as a guide. In order to maximise the measure's ecological validity, a major source of data for its items came from informant interviews, in this study, parents of children with ASD and professionals working with children with ASD. These interviews were used to map the nature of executive function difficulties in children with ASD and then inform the generation of items for the new measure. The professionals were also consulted on their experience of using existing informant-report measures, in particular the BRIEF, and on what a helpful measure would be like. These interviews provided rich data that directly informed the development of the measure.

Study 1 Methods

Ethics

Ethical approval was obtained from the UCL Research Ethics Committee (Project ID: 8057/001). See Appendix C for confirmation of ethical approval. Informed consent was obtained from all participants before taking part in the study. Information sheets for Study 1 can be seen in Appendix D (parents and professionals) and in Appendix E for Study 2 (parents of children with ASD and parents of TD children). The consent form for both studies can be seen in Appendix F. The Ethics Guidelines for Internet-Mediated Research (British Psychological Society, 2017) were referred to in relation to Study 2.

Participants

Six parents of children with ASD and six professionals working in the field of autism were recruited via a convenience sampling method, through the research team's existing contacts. Details of the parent participants, all of whom were female, can be seen in Table 1. One parent had two children with ASD, meaning the parent interviews focused on seven children in total. The seven children with ASD were aged between 7 and 15 years (M = 11.28 years, SD = 3.30) and were not reported to have an intellectual disability. One child had suspected co-morbid ADHD, although undiagnosed; this child was not excluded as ADHD commonly occurs with ASD (Ronald, Simonoff, Kunsti, Asherson, & Plomin, 2008). The professionals were also recruited through the research team's existing contacts and comprised of clinical psychologists and researchers working with children and young people with ASD. Details of the professional participants can be seen in Table 2.

Table 1

Characteristics of Parent Participants in Interviews

	Re. child/ren		
Participant	Age	Gender	Diagnosis
1	15	Female	Asperger's Syndrome
2	15	Male	Asperger's Syndrome
3	13	Male	ASD
4	12	Female	High-Functioning Autism
5	7, 8	Both female	ASD
6	9	Female	ASD (possible ADHD)

Table 2

Characteristics of Professional Participants in Interviews

Participant	Gender	Profession	Place of work
7	Female	Clinical psychologist	ASD assessment service
8	Female	Senior clinical psychologist	ASD assessment service
9	Male	Clinical psychologist	University
10	Female	Clinical psychologist	Local CAMHS
11	Male	PhD researcher on executive function	University
12	Female	Clinical psychologist	University, ASD service

Interview Procedure

Participants were interviewed by the researcher, either in person or over the telephone, and the interviews were audio recorded. All of the parents were interviewed over the telephone, whereas four of the six professionals were interviewed in person, the remaining two over the telephone. The interviews lasted between 30 and 60 minutes. Parents were interviewed about their child's day-to-day difficulties with executive functioning. The professionals were interviewed about two main topics: the executive function difficulties they observe in children with ASD and their experience of using informant-report executive function measures.

Semi-structured interview. Data were collected using semi-structured interview schedules designed for this project. The interview schedules for parents and professionals can be seen in Appendix G. The schedules were guided by the research aims of the project, relevant theory, and clinical experience of the research team. Through prior review of the executive function literature, the interview schedule was based upon Naglieri and Goldstein's (2013) broad definition of executive function which is made up of nine domains: attention, emotion regulation, flexibility, inhibitory control, initiation, organization, planning, self-monitoring, and working memory. The interview schedule was designed to be used flexibly, allowing the interviewer to follow the participants' responses and therefore maximise the chances of gathering meaningful data from the interviewees (Smith, 1995). The parent interviews focused on a typical day and enquired about what children find difficult, homing in on executive function difficulties.

Interview Analysis

A Thematic Analysis was carried out on the parent and clinician interview data to address the question: what is the nature of executive function difficulties in children with ASD? The steps of a thematic analysis, as outlined by Braun and Clarke (2006), were broadly followed. Firstly, through immersion in the data, initial ideas were noted down. Interesting features were then coded and collated into

themes. Potential themes were defined and reviewed against the data and between the main researcher and supervisor. This analysis was intended to inform the item development, rather than constitute the main part of the study. Therefore, reflecting time and resource constraints, the analysis was based on audio recordings of interviews, rather than written transcripts, which is a deviation from Braun and Clarke's (2006) guidelines. The main researcher became familiar with the data via multiple listenings of the interviews and detailed note taking. During the analytic process, the evolving thematic framework was discussed between the main researcher and supervisor and re-formulated multiple times until a final framework was agreed upon. Paul Burgess was also consulted at this stage as an expert in executive function. A thematic framework was created which then informed the generation of questionnaire items, and the overall structure of the questionnaire.

Questionnaire Development

Item generation. The first stage in the development of a new measure is the generation of items (Streiner et al., 2015). The data for this came from the parent and professional interviews. This bottom-up method of development was intended to maximise the measure's ecological validity by grounding the questionnaire in the qualitative data obtained. Potential items for the new measure were generated, and grounded in the data, parent data particularly, using their language and examples as much as possible. Each item was broadly situated within one of the main themes generated by the qualitative analysis. Careful consideration was given to the wording of items and where possible negative wording was avoided (i.e. no, not) as this can make accurate responding more complicated for respondents, especially for those with reduced cognitive capacity (Streiner et al., 2015). Items were designed to be as short as possible whilst not losing the detail and meaning of the qualitative data.

Scale development. The initial item pool contained 91 items with the intention of being as inclusive as possible, as premature removal of items risks a

facet of the construct being missed (Streiner et al., 2015). The items were then gradually reduced through discussion with the project supervisor, colleagues, and professionals in other fields. The thematic framework was used as a guide to inform the spread of items, ensuring adequate coverage of each theme, however this was used as a guide only and did not rigidly dictate the inclusion or exclusion of items. Particular attention was given to the items' interpretability; checking for issues such as ambiguity and the level of jargon. Once a satisfactory list of items was reached, the response scale was decided upon, with consideration of issues such as whether the scale should be uni- or bi-polar, the number of response points and whether to have a mid-point.

Pre-testing. The measure was subsequently pre-tested with members of the target population (parents of children with ASD and parents of TD children). Parents who participated in the interviews and several parents of TD children were asked to complete the questionnaire and provide any feedback, related to the overall questionnaire and/or specific items. At this stage, some items were reverse coded, meaning that a response indicating executive function difficulties required disagreement with the statement. Reverse coding arguably decreases risk of acquiescence but may increase the risk of other biases. Inspection of responses on reverse coded items suggested that some parents had misunderstood the direction of the question, therefore, reverse coding was abandoned at this stage and not used in the piloting of the measure. Reverse coded items can contaminate the data through respondent inattention and confusion (van Sonderen, Sanderman, & Coyne, 2013) and may even produce a method factor related to the negatively worded items which is irrelevant to the construct (Harvey, Billings, & Nilan, 1985).

The final questionnaire contained 48 items which can be seen in Table 3 and will be discussed in the results.

Study 1 Results

Qualitative Analysis: Nature of Executive Function Difficulties in Children with ASD

The thematic analysis focused on the nature of executive function difficulties in children with ASD and can be seen in Figure 1. The following eight themes were derived from the parent and professional data: getting going, planning, self-awareness, rigidity/inflexibility, focus of attention, memory, managing emotions and consequences of executive function difficulties. The framework also depicts sixteen sub-themes and which of the main themes they are connected to. Given the complex, inter-related nature of executive functions (Otero & Barker, 2014), some of the sub-themes are connected to more than one of the main themes, therefore the framework is best represented in a visual format showing a net of themes that are conceptually distinct, yet often linked to each other. Each of the themes will be presented with illustrative quotes.

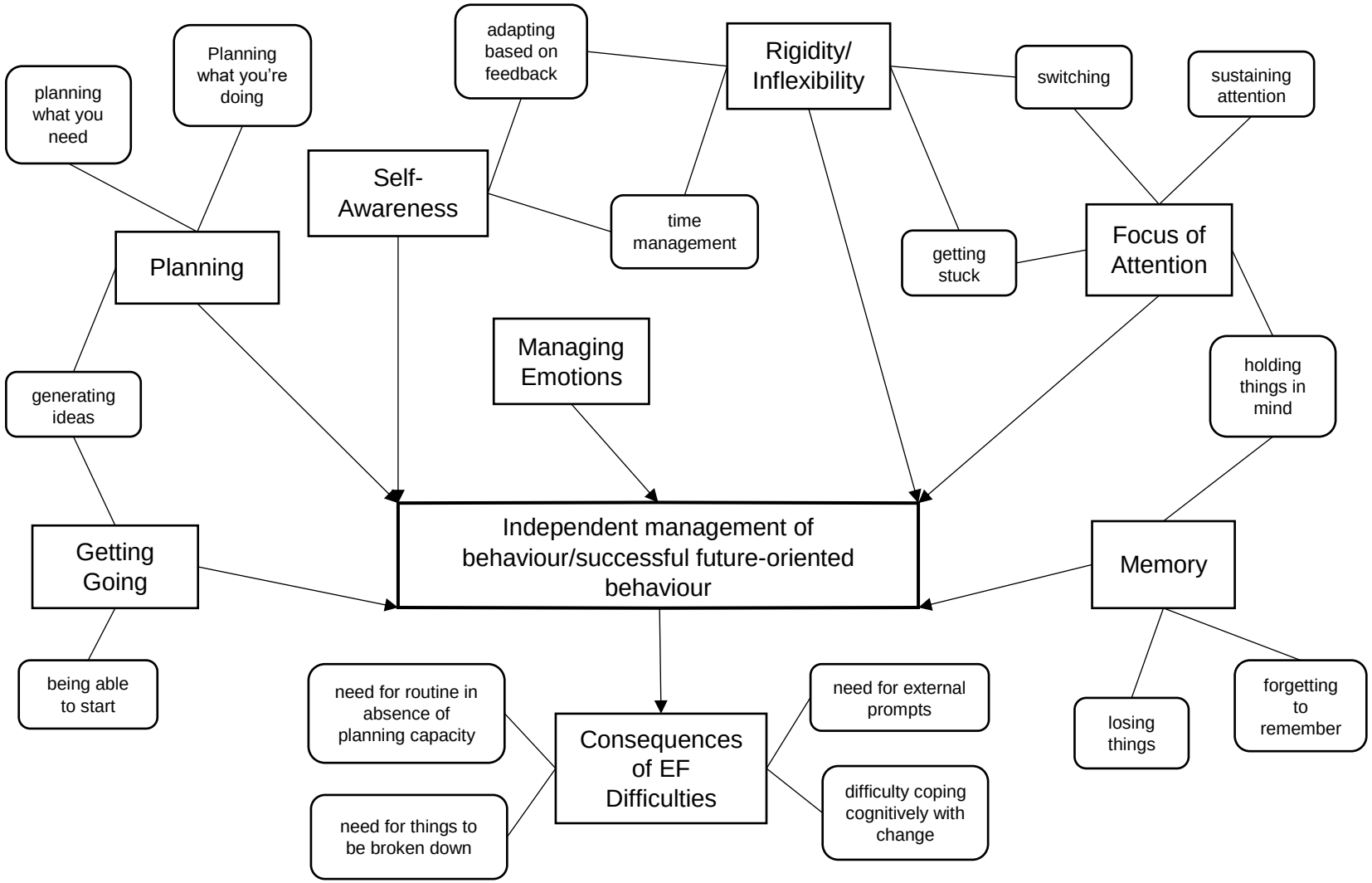

Figure 1. Thematic framework depicting the nature of executive function difficulties in children with ASD.

The first theme of **Getting Going** includes the difficulties that both parents and professionals reported children have with starting tasks and with generating ideas. Most of the parents described how their children find it difficult to start tasks, such as tidying their bedroom, especially if the task is broad or poorly defined. Several parents described the difficulty their child has with knowing how to start tasks at school, with one parent describing this as the blank page problem:

"I call it the blank page, a blank page to her is quite a terrifying thing because I think she thinks well what on earth do I do, how do I start this?" (Participant 6, parent)

Some of the parents also described how their children find routine daily tasks such as getting dressed difficult to start independently. The following quote depicts one parent's account of how their child finds getting started difficult in every aspect of his life:

"That's one of his biggest difficulties, as far as actually starting his work, and knowing what to do, he just needs it broken down. It's the same in every aspect of his life I think, same as getting dressed, he needs that prompt to start everything, he's not quite sure how to start it" (Participant 3, parent)

Linked to that, participants also described the difficulties children have with generating ideas for what to do, for example, in the context of what game to play or how to respond in a new situation:

[Talking about when son missed the school bus] "He was just stood on the bus stop, he didn't know what to do . . . he didn't know what to do because it was completely out of, it had never happened to him before" (Participant 3, parent)

Generating ideas was also connected to the next theme of **Planning**, as it was identified as a necessary part of making a plan. Further sub-themes included under planning were the difficulties children have in planning what they need and planning what they are doing. Parents commonly talked about the difficulty their child has with planning what they need for school and packing their school bag with the correct things:

"She would probably forget at least one thing, so it would be the fact that, if she had to remember her homework book, and her jotter, and her reading book, and her homework diary, she'll probably, one of those things at least, would be forgotten if we just said go and pack your bag. So, it's sometimes maybe the forward planning…" (Participant 6, parent)

Furthermore, planning what they are doing was described as an area of difficulty by some parents and professionals, for example, planning their route around school or planning their morning routine:

"And planning things like routes to take round the school, um, I had one child who was, a teenager, late for every lesson and they couldn't work it out cos he left the lesson at the same time as everybody else, then they realised in the end that he only knew how to get round the building by going back to the front door" (Participant 12, professional)

The next theme, **Self-Awareness**, incorporates difficulties with adapting based on feedback and with time management. Difficulty with adapting based on feedback was an area which was predominantly raised by professionals, rather than parents. Examples of this included, a difficulty modifying your opinion or behaviour in the light of new information or feedback:

[Talking about a child doing their schoolwork] "[You've] picked something up a bit wrong, but think that's how you've been, you're doing it right, because that's what you've been taught, and then can't see that because it's not working there must be a different way of doing it, so perhaps the other person who's telling you a different way of doing it might be right, which I think is about inflexibility as well" (Participant 12, professional)

Several parents and one professional raised the difficulties children can have with time management. This manifested in several ways including finding it difficult to get to lessons on time and lacking an awareness of how long things take them:

"If she's running late, she can't go faster, no concept of time at all" (Participant 1, parent)

Both sub-themes of time management and adapting based on feedback were also connected to the next theme of **Rigidity/Inflexibility**. Difficulties with time management can also relate to inflexibility, in the sense that some children struggle with effective management of their time as they have strict routines which they are not able to adapt. Adapting based on feedback was sometimes described by professionals as a difficulty that could be impacted upon by being rigid or inflexible, as children may be inflexible in adapting their behaviour based on feedback they receive, evident in the quote above. The next sub-theme of getting stuck is closely related to this. Again, getting stuck was talked about more by professionals than parents. One manifestation of getting stuck is that children can become very focused on one task but this may be at the expense of other things:

"Getting stuck, they can show very good attention on things they're good at but it's almost to an excessive extent" (Participant 7, professional)

Another example of getting stuck related to how children can get stuck on the first part of an instruction and go ahead with it despite the rest of the instructions:

"They really just grasp the first instruction and they're already focused on it and going ahead with it" (Participant 8, professional)

A further sub-theme of switching was also included under this theme which is closely related to getting stuck. Professionals commonly talked about switching in the context of being able to move between different environments or between tasks:

"Struggling perhaps with the transition between home and school as well, shifting between environments, people often talk about as a problem" (Participant 9, professional)

Given their relevance to attention, getting stuck and switching are also connected to the theme of **Focus of Attention.** Parents and professionals

frequently talked about children having difficultly sustaining their attention or flitting around. Getting easily distracted was brought up by several parents in relation to being in the classroom:

"He could be distracted by anything, staring out the window, people being silly in class" (Participant 3, parent)

Also related to focus of attention and to the next theme of **Memory** is the sub-theme of holding things in mind. Difficulty with holding things in mind was demonstrated in several parents' accounts of their children forgetting what they went upstairs for:

"You send her upstairs to put her pyjamas on, you go up 10 minutes later and she's sat on her bed, 'what you doing?', 'umm I forgot why I come up here'" (Participant 1, parent)

In addition to holding things in mind, further sub-themes connected to memory were losing things and forgetting to remember. Many parents and professionals talked about children losing or leaving their things:

"Having a set of things that you need and being able to hold on to them and keep them safe and have them when you need them, I think is something parents often talk about as a problem" (Participant 9, professional)

Connected to this, children were reported to struggle with remembering to remember, especially in the context of what they need to take to school:

"Organising his school bag, he always forgets something, and I'm always up the school taking his PE kit, he's left it behind even though I packed it" (Participant 3, parent)

The next theme, **Managing Emotions**, refers to the difficulty many children with ASD were reported to have in recognising and regulating their emotions. Many parents described emotional outbursts or meltdowns seeming to happen out of the blue or as though they built up very quickly:

"He gets quite angry very quickly, you don't really see it coming" (Participant 2, parent)

The final theme, **Consequences of Executive Function Difficulties**, became apparent in all conversations with parents. They frequently talked about the support their children need, for example, needing external prompts and a need for things to be broken down, evident in the next two quotes:

"We use little checklists so she knows what she has to do for the next day" (Participant 6, parent)

"If I give someone instructions that has multiple steps, they'll do the first bit or the last bit, and they might not actually remember all of the component parts, so parents talk about having to really break down instruction sets" (Participant 11, professional)

Some parents also described how their child's need for routine was related to an absence of being able to plan flexibly:

"I think the way that [she] would cope, cos she's done it before, so she just knows, like with the getting dressed and what order it happens in, once there's an order, even if it's quite random the first time you did it, I think she will latch on to that order…rather than following instructions, she's just doing what she's always done I think" (Participant 5, parent)

Related to that, a difficulty coping cognitively with change was also described in the context of finding it difficult to plan and problem-solve flexibly:

"She will have already received texts from my husband and me, saying don't forget, grandad's picking you up, you need to be at the other car park, so um, she probably wouldn't remember that change in the routine" (Participant 4, parent)

Consultation with Professionals

The professionals involved in the study were also consulted on their experience of using existing informant-report measures of executive function and what a helpful measure would be like. All of the professionals interviewed had experience of using the BRIEF.

Experience of using existing measures, including the BRIEF. The professionals consistently explained that their experience of using the BRIEF with an ASD population produces scores that are very elevated. They talked about how many children score at the ceiling on the BRIEF, therefore showing very little variation amongst children. This was highlighted as being different to what is described in the literature, where we might expect variability amongst executive functioning abilities in children with ASD. The particular discrepancy between cognitive tests and parent-report measures was raised as a particular issue by some of the professionals. There was consideration about what the BRIEF is measuring and showing us. Some professionals suggested it is measuring everyday impairment, whilst others suggested it may be demonstrating that there is in fact a very high level of executive dysfunction in the ASD population. Some professionals wondered whether the BRIEF is inflated by social difficulties and anxiety in the ASD population. It was noted that parents can clearly relate to the questions within the BRIEF and it is likely to be telling us something useful about how a child functions day-to-day. However, the potential pitfalls of using parent-report measures in general were also raised, including the risk or reporting bias and the non-specificity of what is being measured.

What a helpful measure would be like. It was felt a helpful measure would demonstrate more variability between children with ASD. This would be useful clinically as it would have the potential to highlight areas of strength and weakness in individual children. Furthermore, it may highlight particular areas to follow up in interview. Ideally, a new measure would not get scores heavily inflated by ASD symptoms and anxiety, therefore the focus of the questions may be more about non-social skills. An effective measure could be used to tailor interventions and provide an explanatory framework to parents about their child's behaviour.

Questionnaire Items

The final list of 48 items can be seen in Table 3, each falling under a primary theme from the thematic framework. The randomised order as administered during online piloting can be seen in Appendix H.

Table 3

Questionnaire Items

No.	Theme	Item
1	GG	My child takes a long time to get started on everyday tasks (e.g. washing, getting dressed)
2	GG	My child finds it difficult to independently get started on tasks or activities
3	GG	My child finds unstructured activities (e.g. tidying room, creative writing task) difficult as he/she doesn't know how to start
4	GG	My child finds non-specific instructions (e.g. "go and get ready") hard to put into action
5	GG	When my child has free time, he/she finds it difficult to come up with new ideas for what to do
6	GG	If something unexpected happens, my child has trouble coming up with ideas about how to react
7	GG	When my child loses something, he/she struggles to independently think of places it could be
8	P	My child finds it difficult to independently pack his/her school bag with what he/she needs

No.	Theme	Item
9	P	My child finds it difficult to plan for an overnight trip (e.g. pack clothes that are appropriate, consider how many changes of clothes are needed)
10	P	My child finds it difficult to have all the things he/she needs for a lesson at school
11	P	My child finds it difficult to get to lessons on time
12	P	My child has difficulty deciding what to do first when doing his/her homework
13	P	My child has difficulty organising him/herself to get ready in the morning
14	SA	My child has difficulty noticing his/her own mistakes when doing schoolwork/homework
15	SA	After starting a task, my child continues without pausing to check he/she is doing it right
16	SA	My child misjudges how long it will take to complete everyday tasks, such as getting ready to leave the house
17	SA	My child loses track of time easily
18	RI	My child finds it difficult to change his/her plan or point of view as a result of new information
19	RI	My child finds it difficult to adjust his/her behaviour based on feedback
20	RI	My child sticks with one way of doing something even when it is not working

No.	Theme	Item
21	RI	If my child can do a task (e.g. a maths problem) one way, he/she struggles to learn a different way of doing it
22	RI	My child finds it difficult to do a task on his/her way to doing something else (e.g. if asked to take out the bins on his/her way to get the bus)
23	RI	My child finds it difficult to switch between tasks and activities
24	FA	When given instructions for a task, my child goes ahead with the first part without paying attention to the rest of the instructions
25	FA	My child misunderstands tasks (e.g. in class, homework) as focuses on specific details rather than the overall picture
26	FA	My child has difficulty mentally putting something on hold (e.g. something that happened earlier) so that he/she can focus on something else instead
27	FA	My child has difficulty multi-tasking (e.g. listening to teacher's instructions and writing them down)
28	FA	If my child is interrupted whilst doing a task or activity, he/she has difficulty going back to it
29	FA	My child has difficulty sustaining his/her attention (e.g. in class)
30	FA	My child has difficulty focusing on important tasks he/she is not interested in
31	FA	My child interrupts others or blurts things out
32	FA	When my child wants to do something, he/she needs to do it immediately

No.	Theme	Item
33	M	My child finds instructions with three or more parts difficult to follow (e.g. “go upstairs and get x, y and z”)
34	M	If given instructions to do a task with several stages, my child may only do the first or last part
35	M	My child goes to another room and forgets what he/she went there for
36	M	My child forgets to take important items (e.g. completed homework) to school
37	M	My child forgets to check whether he/she has items that they need every day (e.g. lunch card, planner)
38	M	My child loses things or leaves things at school all the time (e.g. PE kit)
39	M	My child forgets where he/she has put things at home
40	ME	My child seems to get upset or angry very quickly when things do not go his/her way
41	ME	My child has meltdowns/extreme emotional outbursts
42	C	My child needs many prompts when getting ready for school in the morning
43	C	My child needs everyday tasks and instructions (e.g. getting dressed, tidying room) broken down into small steps
44	C	My child has to follow routines in daily life, in order to remember what he/she needs to do
45	C	My child does everyday activities in a set way (e.g. getting dressed in a particular order)

No.	Theme	Item
46	C	My child relies on routines rather than trying new ways, as he/she struggles to come up with new ideas (e.g. always goes the same route around school even though this is not the most efficient)
47	C	Changes to my child's routine can throw his/her whole day off course
48	C	My child finds it difficult to adjust his/her behaviour when plans change (e.g. if being picked up from school in new location would still go to usual location)

C = consequences of EF difficulties, FA = focus of attention, GG = getting going, M = memory, ME = managing emotions, P = planning, RI = rigidity/inflexibility, SA = self-awareness

Study 1 Discussion

The aim of study 1 was to develop a new parent-report measure of executive function in a way that would promote its ecological validity. This was done by interviewing parents and professionals in order to map the nature of executive function difficulties in children with ASD, generating data which closely informed the generation of items for the measure. The interviews generated rich data which was organised into a network of eight main inter-related themes. Seven of those focused on particular areas of difficulty related to executive functioning (getting going, planning, self-awareness, rigidity/inflexibility, focus of attention, memory, managing emotions) and one on the consequences of those difficulties.

All of the parents who were interviewed highlighted significant executive function difficulties that their children face day-to-day, across a broad range of areas. All of the children were reported by their parents to have difficulties across at least several of the themes identified, and in most cases, across the majority of the themes. The professionals each emphasised particular areas but also described

difficulties across all of the themes. Subtle differences emerged between the accounts of parents and professionals, for instance, the difficulty of adapting based on feedback was raised by professionals rather than parents, whereas difficulties with sustaining attention and being easily distracted were much more often raised by parents. The consequences of executive function difficulties became apparent particularly through the parents' accounts, describing the impact of their child's executive difficulties and the daily adaptations that they make in order to support their child.

Consultation with the professionals regarding existing measures provided evidence of the problems that arise when using the BRIEF with an ASD population. All of the professionals interviewed who had experience of using the BRIEF with this population talked about the high scores seen on the measure and the lack of variation therefore seen amongst children with ASD. This corroborates the findings of the literature review of this thesis which demonstrated very large effect sizes when using the BRIEF with children with ASD compared to TD children. The professionals interviewed emphasised the need for a measure that is not inflated by having ASD and is able to capture the range of difficulties seen amongst children with ASD.

The interviews led to the generation of items and development of the measure. The final version of the measure comprised 48 items and pre-testing provided evidence that is was acceptable to the target population and easy to interpret. Study 2 will now focus on piloting the new measure and a preliminary examination of its psychometric properties. Further discussion of both studies will be covered in the main discussion.

Study 2: Online Piloting

The main aim of study 2 was to pilot the measure online in order to collect data and examine the measure's psychometric properties. The measure was piloted

online with parents of children with ASD and parents of TD children. This allowed examination of the patterns of responding on the measure and its reliability on both an individual item- and scale-level. The measure was then shortened on the basis of these analyses and validity at a scale-level was explored. Specific aims were as follows:

- To identify items that need removing from the measure and assess the measure's reliability:
    - Via examination of endorsement frequency
    - Via individual item test-retest reliability
    - Via tests of internal consistency (item- and scale-level)
- To shorten the scale and examine its validity and distribution of scores.

Study 2 Methods

Design

The study aims were met using a design that had cross-sectional and longitudinal elements, comparing two independent groups: parents of children with ASD and parents of TD children.

Procedures

The 48-item measure, which we have named the measure of Autism Planning and Organisational Difficulties (A-POD), was piloted online with parents of children with ASD and parents of TD children. Participants were recruited via existing contacts of the research team and through adverts placed on social media, including on pages visited by parents and those specific to parents of children with ASD. The study was also advertised by Research Autism, a charity that specialises in promoting research in the field of autism. An example advert can be seen in Appendix I. Entry into a prize draw and a charity donation for every questionnaire completed were offered as incentives for participation.

The online survey was created and administered through the online software Opinio (https://opinio.ucl.ac.uk). Participants accessed the survey by following the link in an advert and were subsequently directed to an online version of the information sheet and then consent form. Consent statements had to be agreed with before the participant could proceed. Separate tailored survey sites existed for the ASD and TD groups. After giving consent, participants were asked to provide demographic information on themselves and their child. They then completed between one and three separate measures (details of which are provided below). A sub-group of participants were asked to complete the new measure of executive function on a second occasion to assess its test-retest reliability. The mean time interval between completions was 21.9 days.

Participants

The following inclusion criteria applied to all participants: parent of a child aged between 8 and 16; parent speaks fluent English; child without an intellectual disability. This project focused on high-functioning children with ASD in order to limit group heterogeneity, as intellectual disability is likely to impact upon executive functioning. Children in the ASD group were required to have a parent-reported formal diagnosis of autism given by a qualified professional, such as a child and adolescent psychiatrist, clinical psychologist or paediatrician.

ASD. Fifty-two parents of children with ASD participated in the online study. Of those, a total of four were excluded, due to the child having a reported intellectual disability. An additional four responses were incomplete and therefore not included, as per the Ethics Guidelines for Internet-Mediated Research (British Psychological Society, 2017) consent cannot be assumed if the participant fails to complete. This resulted in a final sample of 44 (all respondents were mothers). Forty-two parents were located in the UK, two in other European countries. The ASD group comprised of children with reported diagnoses of Autism Spectrum Disorder (n = 15, 34%), Asperger's Syndrome/Asperger's Disorder (n = 22, 50%),

and High-Functioning Autism (n = 7, 16%). All autism diagnoses were reportedly received from CAMHS, specialist ASD assessment teams, or appropriate health care professionals including psychiatrists, clinical psychologists, educational psychologists, paediatricians, and speech and language therapists. Twenty children were reported to have one or more developmental comorbidity: ADHD (n = 8, 18%); specific learning difficulty (n = 7, 16%); other (n = 8, 18%; including sensory processing disorder and dyspraxia). Thirty-seven of the children were reported to have one or more emotional difficulty: anxiety (n = 36, 82%); behaviour/conduct problems (n = 16, 36%); depression (n = 17, 39%); other (n = 2, 4%; including self-harm and pathological demand avoidance).

TD. Fifty-five parents of TD children participated in the online study; 49 of the respondents were mothers and 6 were fathers, all were located in the UK. The majority of the children had no reported developmental problems (n = 50, 91%) or emotional difficulties (n = 47, 85%). The following developmental and emotional difficulties were reported: specific learning difficulty (n = 3, 5%); other developmental difficulty (n = 2, 4%; stammer, visual processing disorder); anxiety (n = 7, 13%); other emotional difficulties (n = 1, 2%).

Group comparisons. Table 4 displays demographic information for the final sample included the analyses. Group comparisons using the Mann-Whitney test showed that the groups differed significantly in terms of age ($U = 715$, $p < .001$), with the ASD participants being older than the TD controls. The groups did not differ in terms of gender ratio ($U = 990$, $p = .073$) or parent education ($U = 975$, $p = .116$).

Table 4

Participant Demographics in Study 2

		Group	
		ASD	TD
N		44	55
Gender ratio (male:female)		28:16* (64% male)	25:30 (45% male)
Age of child (years)			
M (SD); range		12.7 (2.39); 8-16	10.87 (2.55); 8-16
Parent education	Some secondary education (e.g. GCSEs)	8 (18.2%)	7 (12.7%)
	Completed secondary education (e.g. A levels)	9 (20.4%)	4 (7.3%)
	Undergraduate degree (e.g. BSc)	15 (34.1%)	26 (47.3%)
	Postgraduate degree (e.g. masters, PhD)	11 (25%)	18 (32.7%)
	Other	1 (2.3%)	-

*One male is transgender

Measures

Measure of Autism Planning and Organisational Difficulties (A-POD). All participants completed the new questionnaire measuring day-to-day executive function difficulties. The questionnaire had 48 items and the version as administered (in a quasi-randomised order) can be seen in Appendix H. Subsequent questionnaire analyses will refer to this order of items. The questionnaire used a five-point response scale: strongly disagree (1), slightly disagree (2), neither agree nor disagree (3), slightly agree (4), strongly agree (5). Higher scores indicate a greater level of difficulty.

Behavior Rating Inventory of Executive Function-2 (BRIEF-2**,** Gioia et al., 2000). The BRIEF-2 is an informant-report rating scale of everyday executive functioning for children aged 5-18. The second version of the BRIEF contains 63 items and yields a composite score and three index scores: Behavior Regulation, Emotion Regulation, and Cognitive Regulation. A sub-sample of participants completed the BRIEF-2 (ASD n = 26, TD n = 25); these were the first parents who participated in the study. The Global Executive Composite (GEC) raw score was used as the outcome; higher scores indicate a greater level of executive dysfunction. The BRIEF-2 was administered as an established existing measure of executive functioning.

Autism Quotient (AQ). The AQ child (Auyeung, Baron-Cohen, Wheelwright, & Allison, 2007) was administered to parents of children with ASD aged 8-11 (n = 13) and the AQ adolescent (Baron-Cohen, Hoekstra, Knickmeyer, & Wheelwright, 2006) administered to those aged 12-16 (n = 30). AQ data was missing for one adolescent participant. The AQ contains 50 items and is a measure of autistic traits.

Data Analysis

Data analyses were conducted in Excel and SPSS version 24. Preliminary analyses were carried out to assess normality of the variables and decide on the most appropriate statistical tests to use. Group differences on demographic

variables were analysed using the Mann Whitney U test. The psychometric properties of the 48-item scale were examined with the ASD and TD group data combined. This allowed an appropriately large sample size for these analyses to be sufficiently powered. A sample size of at least 50 is recommended to begin considering the psychometric properties of a scale (Streiner et al., 2015). Based on these analyses, the scale was shortened in length and its validity was examined.

A sequential process was carried out on the pilot data based on the ideas of Streiner et al. (2015):

Step 1: Endorsement frequency. The first analysis of the scale's properties carried out was an examination of endorsement frequency at an item-level. The frequency of endorsement is the proportion of people who endorse each response alternative to an item (Streiner et al., 2015). The frequency of endorsement of each response on the scale was examined. Firstly, it was ensured that "neither agree nor disagree" was not highly endorsed (>20%) as that would indicate an item that was difficult to answer and therefore less clinically useful. An acceptable endorsement rate for the other four responses was set at between 5 and 95% (Streiner et al., 2015).

Step 2: Item- and scale-level reliability. Reliability tests were conducted on the 48-item scale. Reliability was assessed at both the individual item-level and scale-level.

Step 2a: test-retest reliability. The stability over time of each item was assessed using the intra-class correlation coefficient (2-way mixed, absolute agreement, single measure). An acceptable limit for test-retest reliability was set at ≥ .75. There is no universally accepted guide for test-retest reliability as it is dependent on multiple factors, including the length of time between testing and the stability of the trait being measured.

Step 2b. Internal consistency. Cronbach's alpha was used as a measure of internal consistency for the whole scale and each subscale, and "Cronbach's if

item deleted" was examined for each item. An acceptable cut-off for Cronbach's alpha was set at 0.8, as this is conventionally deemed to be a good level. Any item which increased the overall scale or sub-scale Cronbach's if it were deleted was removed.

Internal consistency was also assessed through inspection of the mean inter-item correlation and each individual item-total correlation. The mean inter-item correlation was set at an acceptable limit of ≥.25 (Streiner et al., 2015). The item-total correlation of each item was checked and an acceptable limit of ≥ .2 was set (Streiner et al., 2015).

Step 3: Item removal. The scale was shortened by removing weak items based on the preceding analyses.

Step 4: Validity analyses. Preliminary validity analyses were conducted on the shortened scale. Construct validity was assessed by comparing the scores of the two groups on the new measure (using a Mann-Whitney U test), criterion validity was assessed by comparing scores on the new measure to scores on the BRIEF, and discriminant validity was assessed by comparing scores on the new measure to scores on a measure of ASD symptoms (both using a Spearman's rank correlation).

Step 5: Analysis of distribution. The final analyses concerned an examination of both the ASD and TD group's distribution on the new measure.

Study 2 Results

Endorsement Frequency

Endorsement frequencies for each item can be seen in Appendix J. The frequencies of endorsement for each response option ranged from 2% to 41%. The highest rate of endorsement for "neither agree nor disagree" was 19% (items 28 and 30), not above the proposed 20% limit, therefore deemed as acceptable. Frequencies of endorsement for the remaining four response options (strongly disagree, slightly disagree, slightly agree, strongly agree) ranged from 7% to 41%,

falling within the acceptable 5%-95% range. As a result, no items were removed on the basis of endorsement frequency.

Item- and Scale-Level Reliability

Test-retest reliability. A sub-group of parents (ASD n = 26, TD n = 11) completed the A-POD on two occasions, with a mean time interval of 21.9 days. The test-retest correlation coefficients ranged from .521-.964. (*M* = .818, *SD* = .097). Reliability analyses can be seen in Table 5. Eight items were removed at this stage due to poor test-retest reliability (items 13, 30, 35, 11, 22, 34, 18, and 32). See Table 6 for details of all items removed.

Internal consistency. Cronbach's alpha for the 48-item scale was .988, indicating very high internal consistency. Cronbach's alpha for the subscales ranged from .825-.943. Values for Cronbach's alpha if each item was deleted can also be seen in Table 5, demonstrating that the removal of three items would lead to an increased sub-scale Cronbach's alpha (items 40, 35, and 29). As such, these were removed from the measure (see Table 6). The mean inter-item correlation was high at .624 (range = .166-.885). The item-total correlations ranged from .612-.91 for the total scale and .489-.911 for the sub-scales.

Table 5

Results of Reliability Analyses on 48-Item Scale

				Measures of Internal Consistency				
				Total scale		Sub-scale		
Item no.	Item description	Sub-scale	Test-retest ICC	Item-total correlation	Cronbach's α if item deleted	Sub-scale Cronbach's α	Item-total correlation	Cronbach's α if item deleted
10	My child needs everyday tasks and instructions (e.g. getting dressed, tidying room) broken down into small steps	C	.938*	0.86	0.987	0.921	0.798	0.904
14	My child finds it difficult to adjust his/her behaviour when plans change (e.g. if being picked up from school in new location would still go to usual location)	C	.904*	0.835	0.987	-	0.862	0.897
22	My child does everyday activities in a set way (e.g. getting dressed in a particular order)	C	.705*	0.622	0.988	-	0.659	0.918
36	My child has to follow routines in daily life, in order to remember what he/she needs to do	C	.840*	0.764	0.987	-	0.785	0.906

				Measures of Internal Consistency				
				Total scale		Sub-scale		
Item no.	Item description	Sub-scale	Test-retest ICC	Item-total correlation	Cronbach's α if item deleted	Sub-scale Cronbach's α	Item-total correlation	Cronbach's α if item deleted
40	My child needs many prompts when getting ready for school in the morning	C	.843*	0.647	0.988	-	0.489	0.934
42	Changes to my child's routine can throw his/her whole day off course	C	.941*	0.84	0.987	-	0.861	0.897
46	My child relies on routines rather than trying new ways, as he/she struggles to come up with new ideas (e.g. always goes the same route around school even though this is not the most efficient)	C	.873*	0.804	0.987	-	0.834	0.901
3	If my child is interrupted whilst doing a task or activity, he/she has difficulty going back to it	FA	.785*	0.826	0.987	0.943	0.805	0.934

				Measures of Internal Consistency				
				Total scale		Sub-scale		
Item no.	Item description	Sub-scale	Test-retest ICC	Item-total correlation	Cronbach's α if item deleted	Sub-scale Cronbach's α	Item-total correlation	Cronbach's α if item deleted
19	My child misunderstands tasks (e.g. in class, homework) as focuses on specific details rather than the overall picture	FA	.871*	0.864	0.987	-	0.846	0.932
20	My child has difficulty focusing on important tasks he/she is not interested in	FA	.756*	0.774	0.987	-	0.771	0.937
30	When given instructions for a task, my child goes ahead with the first part without paying attention to the rest of the instructions	FA	.524*	0.703	0.988	-	0.695	0.940
32	When my child wants to do something, he/she needs to do it immediately	FA	.743*	0.74	0.988	-	0.730	0.938
34	My child interrupts others or blurts things out	FA	.705*	0.674	0.988	-	0.666	0.942

				Measures of Internal Consistency				
				Total scale		Sub-scale		
Item no.	Item description	Sub-scale	Test-retest ICC	Item-total correlation	Cronbach's α if item deleted	Sub-scale Cronbach's α	Item-total correlation	Cronbach's α if item deleted
37	My child has difficulty multi-tasking (e.g. listening to teacher's instructions and writing them down)	FA	.964*	0.901	0.987	-	0.883	0.929
39	My child has difficulty sustaining his/her attention (e.g. in class)	FA	.904*	0.78	0.987	-	0.790	0.935
43	My child has difficulty mentally putting something on hold (e.g. something that happened earlier) so that he/she can focus on something else instead	FA	.773*	0.861	0.987	-	0.817	0.934
5	My child finds unstructured activities (e.g. tidying room, creative writing task) difficult as he/she doesn't know how to start	GG	.840*	0.85	0.987	0.922	0.822	0.903

				Measures of Internal Consistency				
				Total scale		Sub-scale		
Item no.	Item description	Sub-scale	Test-retest ICC	Item-total correlation	Cronbach's α if item deleted	Sub-scale Cronbach's α	Item-total correlation	Cronbach's α if item deleted
17	When my child loses something, he/she struggles to independently think of places it could be	GG	.757*	0.746	0.988	-	0.721	0.913
18	My child takes a long time to get started on everyday tasks (e.g. washing, getting dressed)	GG	.724*	0.773	0.987	-	0.751	0.911
21	My child finds non-specific instructions (e.g. “go and get ready”) hard to put into action	GG	.895*	0.882	0.987	-	0.861	0.899
26	If something unexpected happens, my child has trouble coming up with ideas about how to react	GG	.897*	0.77	0.987	-	0.739	0.912
33	My child finds it difficult to independently get started on tasks or activities	GG	.818*	0.856	0.987	-	0.848	0.9

				Measures of Internal Consistency				
				Total scale		Sub-scale		
Item no.	Item description	Sub-scale	Test-retest ICC	Item-total correlation	Cronbach's α if item deleted	Sub-scale Cronbach's α	Item-total correlation	Cronbach's α if item deleted
35	When my child has free time, he/she finds it difficult to come up with new ideas for what to do	GG	.661*	0.612	0.988	-	0.555	0.929
2	My child forgets to take important items (e.g. completed homework) to school	M	.817*	0.861	0.987	0.941	0.847	0.928
4	If given instructions to do a task with several stages, my child may only do the first or last part	M	.929*	0.876	0.987	-	0.861	0.927
9	My child forgets where he/she has put things at home	M	.894*	0.693	0.988	-	0.766	0.935
15	My child finds instructions with three or more parts difficult to follow (e.g. “go upstairs and get x, y and z”)	M	.901*	0.865	0.987	-	0.783	0.934
16	My child loses things or leaves things at school all the time (e.g. PE kit)	M	.937*	0.689	0.988	-	0.758	0.936

				Measures of Internal Consistency				
				Total scale		Sub-scale		
Item no.	Item description	Sub-scale	Test-retest ICC	Item-total correlation	Cronbach's α if item deleted	Sub-scale Cronbach's α	Item-total correlation	Cronbach's α if item deleted
25	My child goes to another room and forgets what he/she went there for	M	.859*	0.721	0.988	-	0.754	0.936
38	My child forgets to check whether he/she has items that they need every day (e.g. lunch card, planner)	M	.910*	0.86	0.987	-	0.87	0.926
12	My child has meltdowns/extreme emotional outbursts	ME	.905*	0.773	0.987	0.898	0.816	-
45	My child seems to get upset or angry very quickly when things do not go his/her way	ME	.757*	0.729	0.988	-	0.816	-
1	My child finds it difficult to independently pack his/her school bag with what he/she needs	P	.852*	0.813	0.987	0.941	0.859	0.926
6	My child has difficulty organising him/herself to get ready in the morning	P	.815*	0.816	0.987	-	0.859	0.926

				Measures of Internal Consistency				
				Total scale		Sub-scale		
Item no.	Item description	Sub-scale	Test-retest ICC	Item-total correlation	Cronbach's α if item deleted	Sub-scale Cronbach's α	Item-total correlation	Cronbach's α if item deleted
23	My child finds it difficult to have all the things he/she needs for a lesson at school	P	.864*	0.91	0.987	-	0.911	0.92
28	My child finds it difficult to get to lessons on time	P	.778*	0.817	0.987	-	0.843	0.929
29	My child has difficulty deciding what to do first when doing his/her homework	P	.789*	0.786	0.987	-	0.698	0.945
48	My child finds it difficult to plan for an overnight trip (e.g. pack clothes that are appropriate, consider how many changes of clothes are needed)	P	.905*	0.802	0.987	-	0.782	0.935
11	If my child can do a task (e.g. a maths problem) one way, he/she struggles to learn a different way of doing it	RI	.690*	0.693	0.988	0.933	0.724	0.93

				Measures of Internal Consistency				
				Total scale		Sub-scale		
Item no.	Item description	Sub-scale	Test-retest ICC	Item-total correlation	Cronbach's α if item deleted	Sub-scale Cronbach's α	Item-total correlation	Cronbach's α if item deleted
24	My child finds it difficult to switch between tasks and activities	RI	.851*	0.864	0.987	-	0.825	0.917
27	My child finds it difficult to adjust his/her behaviour based on feedback	RI	.758*	0.766	0.987	-	0.765	0.925
31	My child finds it difficult to change his/her plan or point of view as a result of new information	RI	.762*	0.83	0.987	-	0.868	0.912
41	My child finds it difficult to do a task on his/her way to doing something else (e.g. if asked to take out the bins on his/her way to get the bus)	RI	.910*	0.848	0.987	-	0.763	0.925
44	My child sticks with one way of doing something even when it is not working	RI	.795*	0.832	0.987	-	0.869	0.912

				Measures of Internal Consistency				
				Total scale		Sub-scale		
Item no.	Item description	Sub-scale	Test-retest ICC	Item-total correlation	Cronbach's α if item deleted	Sub-scale Cronbach's α	Item-total correlation	Cronbach's α if item deleted
7	My child loses track of time easily	SA	.852*	0.728	0.988	0.825	0.714	0.749
8	My child misjudges how long it will take to complete everyday tasks, such as getting ready to leave the house	SA	.788*	0.754	0.987	-	0.71	0.751
13	My child has difficulty noticing his/her own mistakes when doing schoolwork/homework	SA	.521*	0.663	0.988	-	0.557	0.82
47	After starting a task, my child continues without pausing to check he/she is doing it right	SA	.760*	0.73	0.988	-	0.624	0.791

*p<.05

C = consequences of EF difficulties, FA = focus of attention, GG = getting going, M = memory, ME = managing emotions, P = planning, RI = rigidity/inflexibility, SA = self-awareness

Removal of Items

Following the above analyses, ten items were removed from the scale according to the guidelines described in the methods. Eight items were removed due to poor test-retest reliability and two items were removed as their removal led to an increased Cronbach's alpha.

Table 6

Items Removed with Reasons

Item no.	Item	Sub-scale	Reason for removal
13	My child has difficulty noticing his/her own mistakes when doing schoolwork/homework	SA	Test-retest < .75
30	When given instructions for a task, my child goes ahead with the first part without paying attention to the rest of the instructions	FA	Test-retest < .75
35	When my child has free time, he/she finds it difficult to come up with new ideas for what to do	GG	Test-retest < .75
11	If my child can do a task (e.g. a maths problem) one way, he/she struggles to learn a different way of doing it	RI	Test-retest < .75
22	My child does everyday activities in a set way (e.g. getting dressed in a particular order)	C	Test-retest < .75
34	My child interrupts others or blurts things out	FA	Test-retest < .75
18	My child takes a long time to get started on everyday tasks (e.g. washing, getting dressed)	GG	Test-retest < .75
32	When my child wants to do something, he/she needs to do it immediately	FA	Test-retest < .75
40	My child needs many prompts when getting ready for school in the morning	C	Cronbach's increase if item deleted

Item no.	Item	Sub-scale	Reason for removal
29	My child has difficulty deciding what to do first when doing his/her homework	P	Cronbach's increase if item deleted

C = consequences of EF difficulties, FA = focus of attention, GG = getting going, P = planning, RI = rigidity/inflexibility, SA = self-awareness

Validity Tests

Validity tests were conducted on the shortened 38-item scale.

Construct validity. The ASD group's raw score on the new measure (M = 161.95, SD = 25.46) was significantly and substantially greater than the TD group (M = 82.98, SD = 29.78) indicating a higher level of executive function problems (U = 94.5, $p < .001$, r = .79). This is in line with predictions, therefore providing initial evidence of the new measure's construct validity. Figure 2 shows the overall distribution of scores broken down into the two groups.

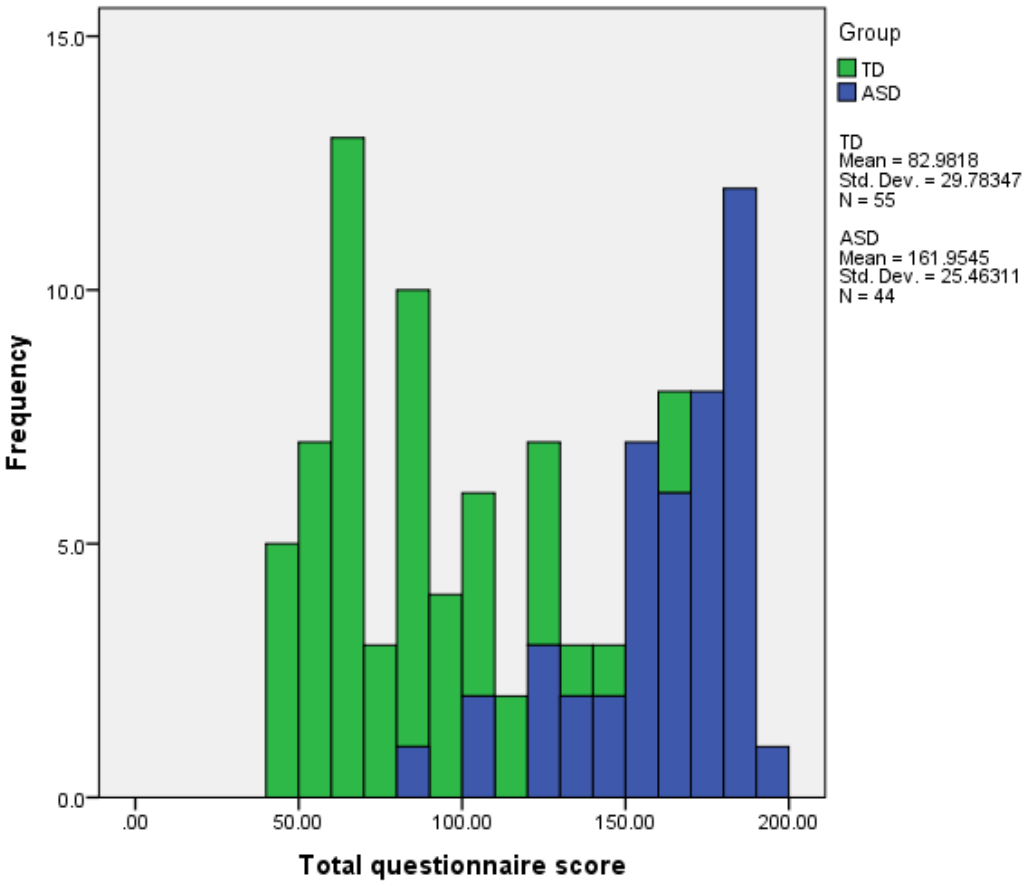

Figure 2. Stacked Histogram Depicting Distributions of TD and ASD Groups on the 38-Item A-POD.

Criterion validity. Raw scores on the new measure and raw scores on the BRIEF (GEC) were significantly positively correlated (r_s = .926, p < .001).

Discriminant validity. Scores on the new measure were not correlated to scores on the AQ-child (r_s = .037, p = .904) or AQ-adolescent (r_s = .147, p = .437). However, the sample sizes included were small and therefore these analyses may be underpowered.

Analysis of Distribution

The distributions of both groups on the A-POD were found to significantly deviate from a normal distribution (ASD: $D(44) = 0.164$, p = .005; TD: $D(55) = 0.135$, p = .014). The ASD distribution was significantly negatively skewed (S = -1.27, z = -3.56, p < .01), whilst the TD distribution was significantly positively skewed

(*S* = .881, *z* = 2.73, *p* < .01) as evident from visual inspection of Figure 2. Kurtosis was significant in the ASD distribution (*z* = 2, *p* < .05) but not significant in the TD distribution (z = 0.47, p > .05).

These findings were compared to distributions on the BRIEF GEC raw score which can be seen in Figure 3. Analyses showed that the distributions of both groups on the BRIEF also significantly deviated from a normal distribution (ASD: *D*(26) = .171, *p* = .048; TD: *D*(25) = 0.196, *p* = .014). However, unlike on the A-POD, the distribution of the ASD group on the BRIEF was not found to have a significant problem with skewness (*S* = -.357, *z* = -0.78, *p* > .05) or kurtosis (*z* = -1.29, *p* > .05), despite looking as though it is negatively skewed. The distribution of the TD group is significantly positively skewed (*S* = .941, *z* = 2.03, *p* < .05) but does not have a problem with kurtosis (*z* = 1.2, *p* > .05). It is of note that skewness statistics are impacted upon by sample size, with larger samples more likely to produce significant findings of skewness. Although the distribution of the ASD group was not found to be significantly skewed, visual inspection of Figure 3 shows that is highly skewed. Visual inspection of Figures 2 and 3 suggest that the A-POD may be more discriminative than the BRIEF between the ASD and TD groups as the two groups appear to overlap slightly less. However, it is not possible to draw any substantive claims at this stage as the distributions are very similar.

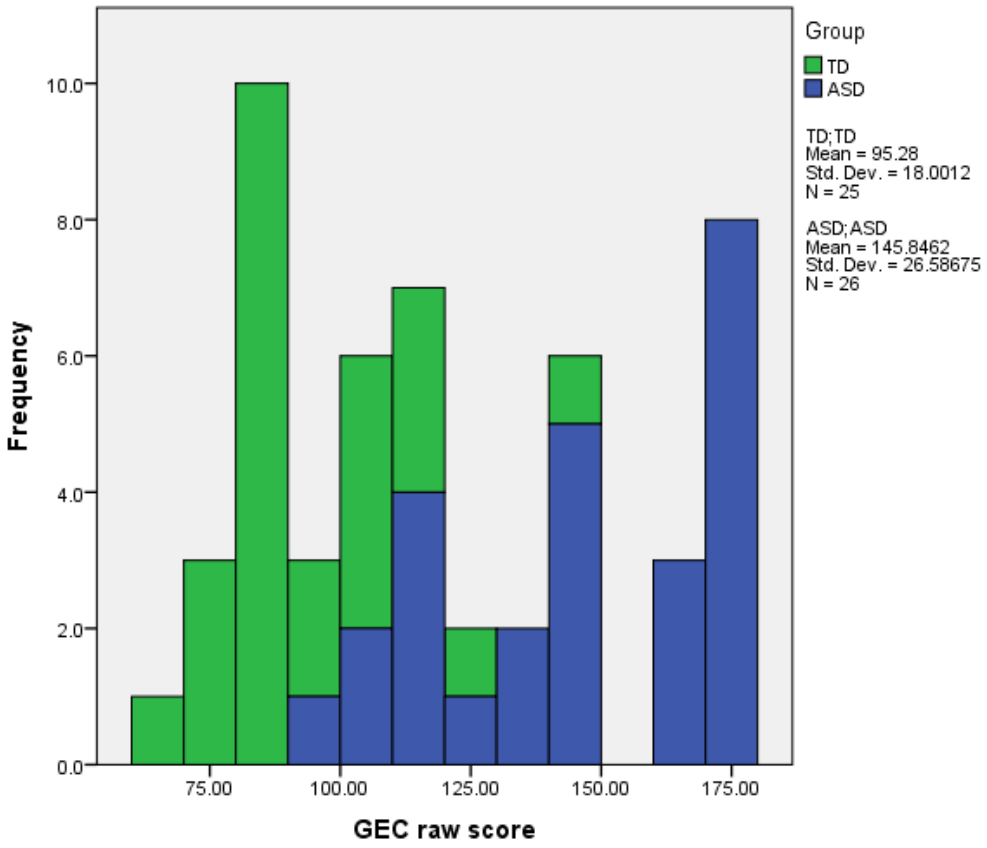

Figure 3. Stacked Histogram Depicting Distributions of TD and ASD Groups on the BRIEF (GEC raw score).

Study 2 Discussion

In Study 2, the 48-item questionnaire developed in Study 1 was piloted online with parents of children with ASD and parents of TD children. Following piloting, the psychometric properties of the scale were examined. Analysis of the endorsement frequencies and reliability were conducted and used to inform shortening of the scale. Preliminary validity tests were then carried out on the shortened 38-item scale and the distributions of the two groups were examined.

The 48-item measure was found to have good stability over time, with a mean test-retest reliability of .82. The measure demonstrated high internal consistency, measured by Cronbach's alpha (.988). The measure's items were highly correlated to each other and to the overall total, providing further evidence of

internal consistency. Ten items with the poorest reliability properties were removed, resulting in the shortened 38-item scale.

The ASD group scored significantly higher on the new measure compared to the TD group, indicating a greater level of executive function difficulty, and the effect size was very large (r = .79). This can therefore be taken as evidence of the new measure's construct validity, as we would expect children with ASD to demonstrate greater executive dysfunction than TD children. Further evidence can be taken from the finding that the measure correlates highly with the BRIEF, providing evidence of concurrent validity (a form of criterion validity). The measure demonstrated preliminary discriminant validity, in that autistic symptoms were not related to outcomes on the new measure, however, these analyses were underpowered. Overall, the new measure demonstrates good psychometric properties. An examination of the response distribution on the new measure reveals a distinct distribution for each group, suggesting the measure is able to discriminate well between children with and without ASD. The distribution of responses in the ASD group shows a positive skew, meaning that scores are bunched up at the top end of the scale. This is therefore suggestive of a ceiling effect.

General Discussion

Overall Aims

The overall aim of this study was to develop and pilot a parent-report questionnaire measure of executive function for use with children with ASD. This measure was intended to have the following characteristics: be able to capture the day-to-day executive function difficulties experienced by children with ASD; be sensitive to the variation amongst children with ASD and therefore avoid a ceiling effect; maximise ecological validity through a bottom-up method of development; be easy to administer and relatively short. This project was reported in two phases. The focus of Study 1 was development of the new measure in which a major source of

data came from interviewing parents and professionals. The focus of Study 2 was piloting of the new measure online, allowing an examination of the measure's psychometric properties. In this general discussion, further consideration to the findings of the qualitative work will first be presented, before going on to discuss the measure and its psychometric properties. Limitations of the study, clinical implications and future directions will then be discussed.

Interviews, Qualitative Findings and Resultant Measure

This is to our knowledge the first study which has used qualitative methods to try and build a picture of everyday executive function difficulties in children with ASD. A bottom-up approach was taken to map these difficulties and directly inform the development of the new measure. This approach was used as a way of maximising the measure's ecological validity. A very broad range of executive function difficulties were spoken about in the parent and professional interviews, meaning the subsequent thematic analysis and resultant questionnaire items therefore cover a wide range of executive function difficulties. Naglieri and Goldstein's (2013) comprehensive definition, comprised of nine domains of executive function, was used as a guide to structure the interviews. The difficulties raised by parents and professionals in the interviews related to all nine of those domains: attention, emotion regulation, flexibility, inhibitory control, initiation, organization, planning, self-monitoring, and working memory. This is particularly interesting as it does not reflect the evidence from performance-based neuropsychological tests which suggest some areas of executive function may not be impaired in individuals with ASD. For example, Pennington & Ozonoff's (1996) influential review did not find evidence of inhibition or working memory deficits in samples of children with ASD. Given the heterogeneity seen within ASD, the current qualitative findings may in part represent the individual differences in executive function abilities between children with ASD. It is possible that patterns of strength

and weakness vary significantly between individuals and there is no clear universal profile of executive dysfunction.

The analysis and interpretation of the interview data was intended to be inductive and grounded in the data as much as possible. Interestingly, the resultant thematic analysis appears to resemble theoretical models of executive function. It is important to recognise that, when conducting a thematic analysis, researchers cannot free themselves of their prior knowledge and theoretical position (Braun & Clarke, 2006). The present thematic analysis is inevitably impacted by my training as a clinical psychologist and the theoretical stance I approached the interviews with. Upon reflection, the complex and poorly defined nature of executive function as a construct meant that careful consideration of what related to the research question was needed. Coding the data in relation to executive function required taking a position on what constituted executive function. No qualitative analysis can be entirely independent of existing knowledge and theory, but interpreting data in relation to such a complex and hard to define topic may ultimately entail more reliance on pre-existing assumptions.

Furthermore, analysis of the interview data highlighted the complex, interrelated nature of executive functions, reflecting what is found in the executive function literature (Otero & Barker, 2014). It proved difficult to think about one type of executive function without also considering other types. Many of the examples given by parents could relate to several of the executive function domains, for example, a commonly reported difficulty related to correctly following task instructions at school. Numerous executive functions could impact upon being able to successfully do this, including: appropriately paying attention to the entire instructions; inhibiting the desire to go straight ahead with the first part of the instructions; the ability to self-monitor in order to check you are doing the correct thing; using working memory to hold the instructions in mind whilst deciding how to proceed. This therefore led to the complex thematic framework in which each sub-

theme was not neatly located under a main theme, which is the conventional method of thematic analysis. Furthermore, the overlap between executive function and ASD symptoms became apparent through the interviews and was raised as a complex issue by several professionals. An example where this frequently arose was the relationship between rigidity or a need for routines and a difficulty with cognitive flexibility. Furthermore, parents often talked about other areas of difficulty, including anxiety, where the overlap with executive function was at times hard to decipher. Anxiety is likely to impact on successful executive functioning (e.g. Ansari, Derakshan, & Richards, 2008) and, as demonstrated in Study 2, a large number of high-functioning children with ASD experience anxiety.

Psychometric Properties of the New Measure

The piloting results of Study 2 indicate that the A-POD is a promising measure of executive functioning in ASD. Firstly, it is highly reliable, demonstrating very good stability over time and high internal consistency. Secondly, we have initial evidence of the measure's construct validity. The finding of a large difference between children with ASD and TD children on the new measure is in line with theory-based predictions and the relative levels of executive function difficulties in these two groups (e.g. Pennington & Ozonoff, 1996). A high score on the new measure is therefore very strongly associated with having ASD at a group level. Visual inspection of the distribution of scores on the measure (Figure 2) suggests it is fairly good at discriminating between children with ASD and TD children, although there is some overlap between the two groups' distributions. Although scores on the measure are strongly associated with group membership (ASD or TD), they are not associated with level of ASD symptoms within the ASD group, as measured by the AQ. This can be taken as preliminary evidence of discriminant validity, further support in favour of the measure's construct validity status, as it indicates the measure is tapping into something distinct from ASD symptoms. These results

should be treated with some caution as they may be underpowered, however, there was no suggestion of a relationship between scores on the AQ and the A-POD.

The effect size for the difference between the ASD group and the TD group on the new measure was very large (r = .79; Cohen's (1988) guidelines specify an r of .5 is considered a large effect). The literature review of this thesis demonstrated a very large effect size of 2.38 (d) for 14 studies using the BRIEF with an ASD group and TD control group (this is equivalent to an r value of .76). The effect size on the new measure is therefore in concordance with this. The present study therefore further highlights the discrepancy between executive function assessment using questionnaire-based measures and performance-based neuropsychological tests, identified in much of the literature (e.g. Demetriou et al., 2017). This discrepancy was spontaneously raised as an issue by many of the professionals interviewed in Study 1.

How does the A-POD compare to the BRIEF?

Scores on the new measure were very highly correlated to scores on the BRIEF, providing evidence of its criterion validity. On the one hand, this is promising and demonstrates that the new measure is tapping into what is also measured by the BRIEF. However, the fact that the correlation was approaching a value of one, leads us to consider what the new measure adds above and beyond the BRIEF.

One of the main aims of the new measure was to avoid a ceiling effect amongst the ASD group, however, examination of the distribution suggests this has not been achieved. The distribution of the ASD group on the A-POD is indicative of a ceiling effect, as also found with the BRIEF. It was hoped that this would be avoided by careful consideration of the question content and designing the measure specifically for the ASD population. Nevertheless, this tells us that the difficulties described in the new measure are widespread amongst children with ASD. Visual inspection of the distributions on the new measure and the BRIEF suggest that the new measure may be more discriminative between the TD and ASD groups.

However, it is too early to concretely say whether the new measure is more useful than the BRIEF for use with the ASD population. Further exploration of the new measure will be needed and ideas for this are presented at the end of this discussion.

Gathering data from parents themselves clearly has great benefit and was chosen as a way of maximising the measure's ecological validity. However, this method of generating items may have contributed to the ceiling effect as the difficulties listed are clearly very familiar to parents. Additional sources of information to aid in the generation of items, such as direct observation of children with ASD, may have proved useful.

In future research, it will be useful to explore additional ways of avoiding the observed ceiling effect. One way may to be use a different response scale with a greater range of options for providing an affirmative response. Another alternative would be re-wording of items so that they are a more extreme version of their current form and therefore more able to pick up variation amongst children who potentially all have reported difficulties in that area.

Limitations

One limitation of this study was the use of a convenience sample in Study 1. This resulted in an over-representation of girls with ASD in the interviews which is not representative of the prevalence of ASD in the population in which more boys are diagnosed (Baird et al., 2006). Furthermore, research shows that girls with ASD may present differently and use more strategies to camouflage and compensate (Lai & Baron-Cohen, 2015). A limitation of the sample used in Study 2 is the significant difference in age between the two groups, with the TD children being younger than the children with ASD, which may have impacted on the findings. Research suggests that children with ASD show age-related improvements in executive functioning (Happé, Booth, Charlton, & Hughes, 2006), therefore age-matched groups may have resulted in an even greater effect size on the A-POD.

A second limitation of this study concerns the thematic analysis. Given the time constraints of the project, a pragmatic decision was made to analyse the interviews directly from the audio recordings, rather than transcribing as recommended by Braun and Clarke (2006). Although, the analytic process was a joint endeavour between the lead researcher and project supervisor, formal credibility checks where a second researcher checks the results against the full data set were not carried out. Although the thematic analysis was otherwise carried out in a methodical way, it is acknowledged that the lack of these elements of a formal qualitative analysis may have allowed more room for bias and ultimately impacted on the resultant thematic framework.

Aside from the observed ceiling effect, an additional limitation of the measure is that it demonstrates overly high reliability. A very high Cronbach's alpha is not recommended (Streiner et al., 2015) and it is advised that individual inter-item correlations should generally be between .15 and .5 (Clark & Watson, 1995). Both factors indicate highly related items, therefore suggestive of redundant items. Furthermore, it is important to note that a greater number of items in a test can artificially inflate the value of Cronbach's alpha (Streiner et al., 2015).

Clinical and Research Implications

The current measure provides a useful and comprehensive description of the difficulties that many high-functioning children with ASD are reported to be having day-to-day. The findings highlight the extent of everyday executive function difficulties in this population. Therefore, the clinical implication of this is that these difficulties need to be routinely assessed in children with ASD. However, the extent to which the measure is capturing executive function impairment rather than everyday impairment is unclear and an issue that requires on-going consideration in research in this area. The value of parent-report should not be underestimated however, as such questionnaire-based measures provide valuable information about how a child copes day-to-day. Everyday life is inherently unpredictable and complex

to navigate, therefore traditional neuropsychological tests which are presented in a structured experimental setting can only tell us so much about how a child manages day-to-day. Clinically, it will continue to be important to utilise varied assessment methods to inform a comprehensive picture of a child's executive functioning.

In future research, the A-POD needs further refinement and assessment of its psychometric properties to improve its utility with an ASD population. Testing the measure with a larger sample would enable an exploratory factor analysis to be conducted, allowing us to look at the factor structure of the measure and consider the validity of the proposed sub-scales. Furthermore, it would be useful to re-examine the psychometric properties of the individual items and shorten the scale. At present, the scale is longer than desirable and the various indicators of internal reliability are suggestive of highly overlapping items. With a larger ASD sample, further examination of whether the measure correlates to a measure of ASD symptoms would be possible. Sensitivity and specificity analyses would also provide useful information about the measure's utility with an ASD population. It would also be interesting to explore includes whether the measure correlates with measure of everyday functioning/adaptive functioning.

Conclusions

This study has successfully developed a new parent-report measure of executive functioning that is grounded in the everyday experiences of children with ASD. The measure captures large differences between children with ASD and TD children and demonstrates very promising psychometric properties. The findings of the interviews and piloting data are suggestive of substantial everyday executive function problems in children with ASD.

References

Alvarez, J. A., & Emory, E. (2006). Executive function and the frontal lobes: A meta-analytic review. *Neuropsychology Review, 16*(1), 17-42. doi:10.1007/s11065-006-9002-x

Amaral, D. G., Schumann, C. M., & Nordahl, C. W. (2008). Neuroanatomy of autism. *Trends in Neurosciences, 31*(3), 137-145. doi:10.1016/j.tins.2007.12.005

American Psychiatric Association (2013*). Diagnostic and statistical manual of mental disorders (5th ed.).* Arlington, VA: American Psychiatric Association.

Ansari, T. L., Derakshan, N., & Richards, A. (2008). Effects of anxiety on task switching: Evidence from the mixed antisaccade task. *Cognitive, Affective, & Behavioral Neuroscience, 8*(3), 229-238. doi:10.3758/CABN.8.3.229

Auyeung, B., Baron-Cohen, S., Wheelwright, S., & Allison, C. (2007). The Autism Spectrum Quotient: Children's Version (AQ-Child). *Journal of Autism and Developmental Disorders, 38*(7), 1230–1240. doi:10.1007/s10803-007-0504-z

Baird, G., Simonoff, E., Pickles, A., Chandler, S., Loucas, T., Meldrum, D., & Charman, T. (2006). Prevalence of disorders of the autism spectrum in a population cohort of children in South Thames: The special needs and autism project (SNAP). *The Lancet, 368*(9531), 210-215. doi:10.1016/S0140-6736(06)69041-7

Baron-Cohen, S. (2009). Autism: The empathizing–systemizing (E-S) theory. *Annals of the New York Academy of Sciences, 1156*(1), 68-80. doi:10.1111/j.1749-6632.2009.04467.x

Baron-Cohen, S., Hoekstra, R. A., Knickmeyer, R., & Wheelwright, S. (2006). The Autism-Spectrum Quotient (AQ) - Adolescent Version. *Journal of Autism and Developmental Disorders, 36*, 343-350. doi:10.1007/s10803-006-0073-6

Baron-Cohen, S., Leslie, A. M., & Frith, U. (1985). Does the autistic child have a "theory of mind"? *Cognition, 21*(1), 37–46. doi:10.1016/0010-0277(85)90022-8

Braun, V., & Clarke, V. (2006). Using thematic analysis in psychology. *Qualitative Research in psychology, 3*(2), 77-101. doi:10.1191/1478088706qp063oa

British Psychological Society (2017). *Ethics Guidelines for Internet-mediated Research*. Leicester: Author. Available from: www.bps.org.uk/publications/policy-and-guidelines/research-guidelines-policy-documents/researchguidelines-poli

Brugha, T., Cooper, S. A., McManus, S., Purdon, S., Smith, J., Scott, F. J., … Tyrer, F. (2012). *Estimating the prevalence of autism spectrum conditions in adults: Extending the 2007 adult psychiatric morbidity survey.* Retrieved from NHS Digital: http://content.digital.nhs.uk/pubs/autism11

Chakrabarti, S., & Fombonne, E. (2005). Pervasive developmental disorders in preschool children: Confirmation of high prevalence. *American Journal of Psychiatry, 162*(6), 1133-1141. doi:10.1176/appi.ajp.162.6.1133

Chaytor, N., & Schmitter-Edgecombe, M. (2003). The ecological validity of neuropsychological tests: A review of the literature on everyday cognitive skills. *Neuropsychology Review, 13,* 181–1297. doi:10.1023/B:NERV.0000009483.91468.fb

Clark, L. A., & Watson, D. (1995). Constructing validity: Basic issues in objective scale development. *Psychological Assessment, 7*(3), 309-319. doi:10.1037/1040-3590.7.3.309

Cohen, J. (1988). *Statistical power analysis for the behavioral sciences*. Hillsdale, NJ: Lawrence Erlbaum Associates.

Damasio, A. R., & Maurer, R. G. (1978). A neurological model for childhood autism. *Archives of Neurology, 35*(12), 777–786. doi:10.1001/archneur.1978.00500360001001

Demetriou, E. A., Lampit, A., Quintana, D. S., Naismith, S. L., Song, Y. J. C., Pye, J. E., . . . Guastella, A. J. (2017). Autism spectrum disorders: A meta-analysis of executive function. *Molecular Psychiatry*, 1-17. doi:10.1038/mp.2017.75

Geurts, H. M., Corbett, B., & Solomon, M. (2009). The paradox of cognitive flexibility in autism. *Trends in Cognitive Sciences, 13*(2), 74-82. doi:10.1016/j.tics.2008.11.006

Geurts, H., Sinzig, J., Booth, R., & Happé, F. (2014). Neuropsychological heterogeneity in executive functioning in autism spectrum disorders. *International Journal of Developmental Disabilities, 60*(3), 155-162. doi:10.1179/2047387714Y.0000000047

Gilbert, S. J., & Burgess, P. W. (2008). Executive function. *Current Biology, 18*(3), 110-114. doi:10.1016/j.cub.2007.12.014

Gilotty, L., Kenworthy, L., Sirian, L., Black, D. O., & Wagner, A. E. (2002). Adaptive skills and executive function in autism spectrum disorders. *Child Neuropsychology, 8*(4), 241-248.

Gioia, G. A., Isquith, P. K., Guy, S. C., & Kenworthy, L. (2000). Test review: Behavior rating inventory of executive function. *Child Neuropsychology, 6*(3), 235-238. doi:10.1076/chin.6.3.235.3152

Gioia, G. A., Isquith, P. K., Kenworthy, L., & Barton, R. M. (2002). Profiles of everyday executive function in acquired and developmental disorders. *Child Neuropsychology, 8*(2), 121-137. doi: 10.1076/chin.8.2.121.8727

Goldberg, M. C., Mostofsky, S. H., Cutting, L. E., Mahone, E. M., Astor, B. C., Denckla, M. B., & Landa, R. J. (2005). Subtle executive impairment in children with autism and children with ADHD. *Journal of Autism and Developmental Disorders, 35*(3), 279-293. doi:10.1007/s10803-005-3291-4

Goldstein, S., Naglieri, J. A., Princiotta, D., & Otero, T. M. (2014). Introduction: A history of executive functioning as a theoretical and clinical construct. In S.

Goldstein & J. A. Naglieri (Eds.), *Handbook of executive functioning* (pp. 3-12). New York: Springer.

Happé, F., Booth, R., Charlton, R., & Hughes, C. (2006). Executive function deficits in autism spectrum disorders and attention-deficit/hyperactivity disorder: examining profiles across domains and ages. *Brain and Cognition, 61*(1), 25-39. doi:10.1016/j.bandc.2006.03.004

Happé, F., & Frith, U. (2006). The weak coherence account: Detail-focused cognitive style in autism spectrum disorders. *Journal of Autism and Developmental Disorders, 36*(1), 5-25. doi:10.1007/s10803-005-0039-0

Harvey, R. J., Billings, R. S., & Nilan, K. J. (1985). Confirmatory factor analysis of the Job Diagnostic Survey: Good news and bad news. *Journal of Applied Psychology, 70*(3), 461-468. doi:10.1037/0021-9010.70.3.461

Hill, E. L. (2004). Evaluating the theory of executive dysfunction in autism. *Developmental Review, 24*(2), 189-233. doi:10.1016/j.dr.2004.01.001

Isquith, P. K., Roth, R. M., & Gioia, G. (2013). Contribution of rating scales to the assessment of executive functions. *Applied Neuropsychology: Child, 2*(2), 125-132. doi:10.1080/21622965.2013.748389

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. doi:10.1007/s11065-008-9077-7

Lai, M. C., & Baron-Cohen, S. (2015). Identifying the lost generation of adults with autism spectrum conditions. *The Lancet Psychiatry, 2*(11), 1013-1027. doi:10.1016/S2215-0366(15)00277-1

Lopez, B. R., Lincoln, A. J., Ozonoff, S., & Lai, Z. (2005). Examining the relationship between executive functions and restricted, repetitive symptoms of autistic disorder. *Journal of Autism and Developmental Disorders, 35*(4), 445-460. doi:0.1007/s10803-005-5035-x

Luria, A. R. (1966). *Higher cortical functions in man.* New York, USA: Basic Books.

Mandy, W., Murin, M., & Skuse, D. (2015). The cognitive profile in autism spectrum disorders. In M. Leboyer M & P. Chaste (Eds.), *Autism spectrum disorders. Phenotypes, mechanisms and treatments* (pp. 34-45). Basel, Switzerland: Karger Publishers.

McAuley, T., Chen, S., Goos, L., Schachar, R., & Crosbie, J. (2010). Is the behavior rating inventory of executive function more strongly associated with measures of impairment or executive function? *Journal of the International Neuropsychological Society, 16*(3), 495-505. doi:10.1017/S1355617710000093

Naglieri, J., & Goldstein, S. (2013). *Comprehensive executive functioning inventory technical manual.* Toronto, Canada: Multi-Health Systems.

Otero, T. M., & Barker, L. A. (2014). The frontal lobes and executive functioning. In S. Goldstein & J. A. Naglieri (Eds.), *Handbook of executive functioning* (pp. 29-44). New York, NY: Springer.

Pennington, B. F., & Ozonoff, S. (1996). Executive functions and developmental psychopathology. *Journal of Child Psychology and Psychiatry, 37*(1), 51–87. doi:10.1111/j.1469-7610.1996.tb01380.x

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. doi:10.1007/s10803-014-2309-1

Ronald, A., Simonoff, E., Kuntsi, J., Asherson, P., & Plomin, R. (2008). Evidence for overlapping genetic influences on autistic and ADHD behaviours in a community twin sample. *Journal of Child Psychology and Psychiatry, 49*(5), 535–542. doi:10.1111/j.1469-7610.2007.01857.x

Shallice, T., & Burgess, P. W. (1991). Deficits in strategy application following frontal lobe damage in man. *Brain, 114*(2), 727-741. doi:10.1093/brain/114.2.727

Smith, J. A. (1995). Semi structured interviewing & qualitative analysis. In J. A. Smith, R. Harre, & L. Van Langenhove (Eds.), *Rethinking methods in psychology* (pp. 9–26). London, UK: Sage.

Streiner, D. L., Norman, G. R., & Cairney, J. (2015). *Health measurement scales: A practical guide to their development and use* (5th edition). Oxford, UK: Oxford University Press.

Van Sonderen, E., Sanderman, R., & Coyne, J. C. (2013). Ineffectiveness of reverse wording of questionnaire items: Let's learn from cows in the rain. *PLOS ONE, 8*(7), e68967. doi:10.1371/journal.pone.0068967

White, S. J., Burgess, P. W., & Hill, E. L. (2009). Impairments on "open-ended" executive function tests in autism. *Autism Research, 2*(3), 138-147. doi:10.1002/aur.78

Wilson, B. A., Evans, J. J., Emslie, H., Alderman, N., & Burgess, P. (1998). The development of an ecologically valid test for assessing patients with a dysexecutive syndrome. *Neuropsychological Rehabilitation, 8*(3), 213-228. doi:10.1080/713755570

Part 3: Critical Appraisal

Introduction

The following is a critical appraisal of the empirical research I undertook as part of the Doctorate in Clinical Psychology. I will begin by presenting an expanded discussion of both the study's and the measure's limitations, before then discussing methodological challenges that arose throughout the process. I will also present a consideration of broader conceptual issues, before finally offering reflections on the research process and my own personal development.

Further Discussion of Limitations

Sampling Limitations

A convenience sample was used for the interviews in Study 1 which resulted in an over-representation of parents of girls with ASD (five out of seven children with ASD were female). This was likely impacted upon by the project supervisor's research interest in the female phenotype and therefore having contacts comprising of many women with ASD and parents of girls with ASD. Research suggests that girls and women with ASD may present differently to males, for example, demonstrating less repetitive stereotyped behaviours (Mandy et al., 2012). There is some preliminary evidence that males and females may differ in terms of degree of executive function difficulty (Lai et al., 2012). The resultant thematic analysis and questionnaire were based upon the interviews, and may therefore be biased by the number of girls in the parent interviews. Interestingly, girls were also over-represented in the online piloting sample (Study 2). The online sample was made up of 63.6% boys, therefore, approaching two boys to every one girl. The commonly cited sex ratio across all Autism Spectrum Disorders is four males to every female (Baird et al., 2006), with an even higher prevalence of males in higher functioning groups (eight or nine to one; Mandy et al., 2012). It is likely that ASD is under-recognised in females and therefore the true rate of ASD may not be so biased towards males. However, children in the current project were required to have an established diagnosis of ASD. Again, it is likely this was in part impacted upon by

the recruitment methods; adverts were placed on social media accounts of the research team, meaning a greater number of people interested in gender differences and the presentation of ASD in females may have been reached.

Nevertheless, many participants were recruited via social media from sources unconnected to the research team's contacts, therefore, I wonder if other factors also impacted upon the observed gender ratio in Study 2. The reasons for this are unclear, so we can only speculate. Autism Spectrum Disorders are more likely to be misdiagnosed and therefore later diagnosed in girls and women (Lai & Baron-Cohen, 2015). Therefore, parents of girls with ASD are more likely than parents of boys to have actively sought a diagnosis, and it is possible that they may be more motivated to engage in research. The difficulties of correctly getting an ASD diagnosis were in fact raised by several of the parents of girls in the interviews. Gender differences on the A-POD were not explored but this would be an interesting avenue to pursue in future research.

This raises a more general issue of the factors that influence participants when deciding to take part in research. Where a participant sample is self-selecting through voluntary participation, we will always face the question of how representative the sample is of the population under investigation. The participants motivations for taking part are unknown and we may see biases in their presentation or demographic characteristics. Participants who respond to research participation requests are likely to differ from those who do not participate in terms of interest, motivation, and educational level (Barker, Pistrang, & Elliott, 2016). In Study 2, we can see that the sample does has a very high level of educational attainment. Eighty percent of the typically developing sample in Study 2 had undergraduate or postgraduate level education, whereas, the 2011 census found that 27% of adults in the UK had obtained degree-level education (Office for National Statistics, 2016). Furthermore, although internet recruitment is a convenient way to reach a large number of potential participants, economically disadvantaged members of the

population are less likely to have internet access (Dillman, Smyth, & Christian, 2014), although this is ever changing as more and more people have internet access. The impact of these biases may be that the sample obtained is not as representative of the ASD and TD populations as we would like. This is a recognised problem across psychological research, in particular as the vast majority of participants are drawn from white, educated, industrialised, rich, and democratic (WEIRD) societies. People from societies with these characteristics represent as much as 80% of study participants, but only 12% of the world's population (Henrich, Heine, & Norenzayan, 2010).

Service-User Involvement

Although the involvement of service users is recommended in research, unfortunately this study did not directly involve individuals with ASD. Consultation with individuals from the target population can provide valuable experiential knowledge and assist in shaping research. In this project, individuals with ASD were not consulted, although parents of children with ASD were a crucial part of the development of the measure. If time had not been so limited, I would have consulted more parents at the pre-testing phase, maybe running a focus group to gather feedback on the measure. Furthermore, children or adults with ASD could have been interviewed directly about the executive function difficulties that they experience day-to-day as part of the development phase.

A significant amount of research is carried out on the topic of ASD but an examination of the focus suggests there may be a mismatch between the type of research being carried out and what people directly affected by autism perceive as important. It has been demonstrated that individuals in the autism community are dissatisfied by the lack of breadth in current autism research and the bias towards basic science and are most interested in research that will make a difference to people's day-to-day lives (Pellicano, Disnmore, & Charman, 2014). I hope that the current study would be perceived as valuable by the autism community for its

relevance to clinical practice. The accurate assessment of executive function is essential to offering tailored interventions with the aim of improving a child's everyday life.

Limitations of the Measure

Our new measure, the A-POD, faces two main challenges in its current form. Firstly, the indications of overly high internal consistency and secondly, the observed ceiling effect. I will further discuss the A-POD's high internal consistency, which is both a strength and weakness. The 48-item measure was found to have a Cronbach's alpha of .988 which is very high. On one hand, this is a very encouraging finding as it provides strong evidence that the measure's items are tapping into the same underlying construct. An acceptable or desirable limit for Cronbach's alpha is a source of on-going discussion in the psychometric literature. Nunnally (1978) suggests a value of at least .8 for basic research and .9 for a clinical instrument. However, an overly high Cronbach's alpha is not entirely desirable as it may suggest a high level of item redundancy, indicating that some items are unnecessary and the scale may be too narrow to have much validity (Streiner, Norman, & Cairney, 2015). Barker et al. (2016) suggest that a reliability statistic of .8 or above is good, but a value of more than .9 may indicate overkill (too many items) or triviality (selection of superficial but easily rateable variables). It is therefore also desirable for individual inter-item correlations and item-total correlations to not be overly high. It is suggested that that inter-item correlations should generally fall between .15 and .5 (Clark & Watson, 1995) and individual item-total correlations should not be greater than 0.7 (Kline, 1979). These are further indications of an overly narrow and specific scale. Many of the A-POD's inter-item correlations were above .5, with the highest being .885. Furthermore, the majority of the item-total correlations were greater than .7. Overall, these analyses indicate that we have developed a measure with highly related items. However, the findings are highly suggestive of redundant items. It is important to note that Cronbach's alpha is

affected by the length of the scale and the sample size, with more items or a large sample artificially increasing the value of alpha. It will be of interest to re-assess measures of internal consistency when the measure is shortened in future research.

Methodological Challenges and Dilemmas

Interviews and Thematic Analysis

The use of qualitative data from parents and professionals in developing the measure was intended to maximise its ecological validity. Before beginning the process, I had no experience of carrying out qualitative research and several notable challenges arose during the process. Firstly, in developing the semi-structured interview schedule I became aware of the tension between bottom-up and top-down methods of collecting qualitative data. The interviews and subsequent thematic analysis were intended to de data-driven as much as possible, however, it was not possible for this to be totally independent of theory (Braun & Clarke, 2006). A difficult balance needed to be struck to ensure that the interviews were not overly directed by my pre-existing ideas and assumptions, yet that the data were relevant to research question. This tension feels particularly relevant to this project as the construct of executive function is complex and not clearly defined, which I will go on to discuss later. This meant that during the interviews and in their analysis, constant judgements had to be made about whether the data were or were not related to executive function. A decision was taken to be relatively inclusive when judging whether something related to executive function during the interviewing, in order to not limit the experiences of parents and capture as much rich data as possible. The data collected during the interviews were incredibly valuable in developing the measure and the examples provided were often rich and informative.

The resultant thematic framework appears to adhere relatively closely to theoretical models of executive function. The extent to which this was reflected in the data or how much it resembles my existing theoretical stance is interesting to reflect upon. When conducting qualitative research, there will always be an

interaction between what the researcher brings and what the participants present. It would be interesting to know whether another researcher, in particular someone who has not previously studied executive function, would come up with a similar framework given the same data. However, this is an unavoidable part of qualitative research; the researcher's own assumptions and existing knowledge cannot be ignored and will ultimately affect interpretation of the data, in a way that is less of an issue in quantitative research. In this respect, the experience of conducting qualitative research was new and interesting for me.

What Constitutes Executive Function?

The difficulty of teasing apart the different areas of executive function from each other became apparent during analysis of the interviews, as the different domains of executive function are so overlapping and inter-related (Otero & Barker, 2014). Furthermore, I became very aware of the overlap between executive functions and the core impairments of ASD, both social and non-social. Prior to conducting the interviews, the hypothesised relationship between rigid behaviours and executive functioning (in particular cognitive inflexibility) was apparent to me. The interviews also highlighted to me the potential relationship between executive function and the core social difficulties seen in ASD. For example, a social communication deficit such as a difficulty in turn-taking, which likely relates to theory of mind, may also relate to a deficit in executive function, in particular, inhibition. Proponents of the executive dysfunction account of ASD (e.g. Damsaio & Maurer, 1978) would argue that executive dysfunction does relate to these social symptoms, however, in this study, we were not interested in trying to tease apart different theories of ASD. Therefore, examples of executive functioning which did not relate to social skills were focused upon in the interviews. This is the one of the potential limitations of the BRIEF with an ASD population, several items do relate to social interactions, which will be impacted upon by the social difficulties characteristic of ASD, whether or not they relate to executive function.

Questionnaire Development

During the development of the measure, many methodological dilemmas arose. Firstly, a balance needed to be struck between the general and specific nature of the questionnaire items. Parents provided many descriptive and rich examples of executive function difficulties during the interviews. We chose to often present examples within the questionnaire items, so that they were closely grounded in the data. However, the items needed to be broad enough that respondents could think of other ways the particular difficulty might manifest beyond the specific example. It was felt that examples were useful to clarify items, especially when the particular difficulty might be hard to conceptualise.

The interpretability and wording of items was given careful consideration to ensure the scale was as accessible as possible to the general population, this included avoiding ambiguity, double-barrelled questions and jargon (Streiner et al., 2015). Negatively worded items were avoided wherever possible, one main reason for this, is that some people, including lower-functioning adults may have difficulty understanding they need to disagree with an item in order to indicate a positive answer. Furthermore, when scales with both positively and negatively worded items are factor analysed, two factors often emerge which relate to the positive and negative worded items, irrespective of their content (Streiner et al., 2015). As the measure is further developed, it would be useful to formally assess its reading level using a measure such as the Flesch–Kincaid readability test.

It is recommended by some in the psychometric literature that reverse coding be used, meaning that half of the items tap the opposite direction of the trait being measured. The usefulness of reverse coding of items was carefully considered in the development of the measure. Although reverse coding can reduce the risk of response biases, such as acquiescence, it can introduce other problems. Pre-testing of the A-POD raised cause for concern regarding the reverse coded items. From examination of individual response sets it was clear that some parents

had misinterpreted some items that were reverse coded. Therefore, the decision was made that the potential disadvantages of reverse coding outweighed the advantages. On reflection, I wonder whether reverse coded items would have impacted on the ceiling effect as it can reduce risk of acquiescent responding.

Broader Issues

Concept of Executive Function

In hindsight, my knowledge of executive functioning prior to beginning this project was relatively simplistic. As I became engaged in the literature and learnt much more about executive function, I came to quickly recognise the complexity of the field and of executive function as a construct. Even trying to define executive function was difficult; there is no universally agreed definition, hence multiple definitions of executive function exist. Goldstein, Naglieri, Princiotta and Otero (2014) recently reviewed the field in their comprehensive book, this proved useful and provided a good overview of the many different definitions in existence. Although executive function is widely accepted as a construct and commonly used, it has been criticised for "conceptual under-specification" (Geurts, Corbett, & Solomon, 2009). There is a lot of on-going debate about whether executive function represents a single central executive or whether it is made up of multiple processes. Some have argued that executive function is a unitary construct that cannot be artificially fractionated and therefore neuropsychological tests which attempt to assess one aspect, such as cognitive flexibility, are attempting to break up a complex and integrated system (Burgess, 1997). Others query whether executive function should be seen as a unitary construct and question its usefulness and validity as a construct, given how broad and poorly defined it is (Salthouse, 2005). During this process, as mentioned previously, it was hard to tease apart executive functioning from other aspects of functioning and it sometimes felt as though executive function encapsulated all higher level cognitive functions.

Questionnaire Measures

This project has successfully developed a new measure that demonstrates promising psychometric properties and appears to tap into the day-to-day difficulties faced by children with ASD. One important question that the project raises is what is the A-POD actually measuring? This is a source of on-going discussion with respect to the BRIEF. This study provides further evidence of the discrepancy between parent-report measures and performance-based neuropsychological tests. We find a very large effect size on the A-POD for a comparison between the ASD and TD groups, as found with the BRIEF. This contrasts with the moderate effect sizes and mixed findings demonstrated on performance-based neuropsychological tests. Arguably, the BRIEF and other parent-report measures of executive functioning tap the integrated, multidimensional skills that are often demanded in real-world situations, whereas performance-based measures assess discrete parts of this executive function system in controlled settings (Isquith, Roth, & Gioia, 2013). Some argue the BRIEF is a better measure of everyday impairment than executive function (McAuley, Chen, Goos, Schachar, & Crosbie, 2010). Given the relationship between executive functioning and everyday functioning, it may be that executive function difficulties manifest as significant everyday impairment. Scores on the A-POD correlate very highly with scores on the BRIEF, so it is likely they are tapping into the same difficulties. In future research, it would be interesting to use the A-POD with other measures of executive function and to examine their relationship.

Aside from what questionnaire measures are actually tapping into, they will also be impacted by biases that are inherent in self- or informant-report methods. Factors including the rater's emotional state or personality characteristics may influence their ratings (Isquith et al., 2013). Furthermore, questionnaire measures used in clinical settings may be influenced by the rater's motivations and the message they wish to communicate, for example, one of significant distress or

impairment. As mentioned previously, response biases, such as acquiescence, can also affect how an individual answers a questionnaire measure.

A Deficit Narrative

As this project advanced and I simultaneously progressed in my clinical psychology training, it became more apparent to me that the nature of the project was very much focused on deficits. This is not necessarily a criticism of the project but more a personal reflection. This became particularly apparent when conducting the parent interviews. The purpose of the interviews was to gather data on the nature of executive function difficulties in ASD, understandably they were therefore very much focused on "deficit". Some parents also talked about things that their children were good at, examples included playing a musical instrument to a high standard or being very conscientious. At the time of conducting the interviews and developing the measure I was on a clinical placement in a Child and Adolescent Mental Health Service. During this time, I was developing my interest and knowledge of systemic approaches (e.g. Hedges, 2005). In my work with children and young people, I was often listening out for alternative narratives, picking up on strengths and resources. This sat somewhat in contrast to the focus on deficits in my research interviews. It was difficult not to pursue stories of resource and strength in the parent interviews. This was interesting to reflect upon as I developed my own identity as a clinical psychologist.

In the context of this research project, however, the focus on difficulties is justified and necessary as we were trying to develop a good assessment tool which would enable accurate assessment of day-to-day difficulties and inform targeted interventions. Furthermore, given the close association between executive functioning and independent functioning in ASD (Pugliese et al., 2015), the importance of good assessment tools is essential as it may directly impact the daily lives of children with ASD. However, it is important to simultaneously hold in mind the idea that societal narratives around deficit and dysfunction can have negative

impacts on individuals who are different from the “norm”. A shift in research and social narratives to a focus on strengths as well as difficulties will be beneficial going forward.

Research Process: Reflections and Personal Development

I chose this research topic predominantly due to my previous research experience with ASD as part of my undergraduate degree research project. Furthermore, I have a long-standing personal interest in developmental disorders and ASD which has been shaped by my personal and family experiences. When beginning the project, I had little understanding of the process of developing a measure. I have found the process relatively challenging, yet rewarding. At times during the process I felt anxious to make the correct decisions, for example about methodological choices. I gradually came to the realisation that there was often not one right choice, but many ways of doing things. I grew in confidence in my ability to make decisions based on the evidence available. Furthermore, as the project was part of the DClinPsy, decisions were often made in the context of resource and time constraints. An example of this, was the decision not to transcribe the interviews of Study 1. The reality of conducting research involves trying to achieve an acceptable balance between rigorous gold-standard methodology and the constraints in which you are operating. It is therefore essential that decisions are made in informed and explicit way, with clear rationale. This has parallels with working in an NHS clinical setting, in which constraints may limit what you are able to offer as a clinician.

The scope of this project became larger than expected and developed into two distinct parts which may have been suitable for two separate projects. However, the focus of the project and the mixed methods employed mean I have really developed my existing research skills and developed many new skills in areas I was not familiar with. Furthermore, during the period of conducting this research project, I have faced personal challenges. This all contributes to a real sense of achievement in having successfully completed a project I am proud of.

References

Baird, G., Simonoff, E., Pickles, A., Chandler, S., Loucas, T., Meldrum, D., & Charman, T. (2006). Prevalence of disorders of the autism spectrum in a population cohort of children in South Thames: The special needs and autism project (SNAP). *The Lancet, 368*(9531), 210-215. doi:10.1016/S0140-6736(06)69041-7

Barker, C., Pistrang, N., & Elliott, R. (2016). Research methods in clinical psychology: An introduction for students and practitioners (3rd ed.). Chichester, UK: John Wiley & Sons.

Braun, V., & Clarke, V. (2006). Using thematic analysis in psychology. *Qualitative Research in psychology, 3*(2), 77-101. doi:10.1191/1478088706qp063oa

Burgess, P. W. (1997). Theory and methodology in executive function research. In P. Rabbitt (Ed.), *Methodology of frontal and executive function* (pp. 81–116). Hove, UK: Psychology Press.

Clark, L. A., & Watson, D. (1995). Constructing validity: Basic issues in objective scale development. *Psychological Assessment, 7*(3), 309-319. doi:10.1037/1040-3590.7.3.309

Damasio, A. R., & Maurer, R. G. (1978). A neurological model for childhood autism. *Archives of Neurology, 35*(12), 777–786. doi:10.1001/archneur.1978.00500360001001

Dillman, D. A., Smyth, J. D., & Christian, L. M. (2014). *Internet, phone, mail, and mixed-mode surveys: The tailored design method* (4th ed.). Hoboken, NJ: John Wiley & Sons.

Geurts, H. M., Corbett, B., & Solomon, M. (2009). The paradox of cognitive flexibility in autism. *Trends in Cognitive Sciences, 13*(2), 74-82. doi:10.1016/j.tics.2008.11.006

Goldstein, S., Naglieri, J. A., Princiotta, D., & Otero, T. M. (2014). Introduction: A history of executive functioning as a theoretical and clinical construct. In S.

Goldstein & J. A. Naglieri (Eds.), *Handbook of executive functioning* (pp. 3-12). New York: Springer.

Hedges, F. (2005). *An introduction to systemic therapy with individuals: A social constructionist approach.* Basingstoke, UK: Palgrave Macmillan.

Henrich, J., Heine, S. J., & Norenzayan, A. (2010). The weirdest people in the world? *Behavioral and Brain Sciences, 33*(2-3), 61-83. doi:10.1017/S0140525X0999152X

Isquith, P. K., Roth, R. M., & Gioia, G. (2013). Contribution of rating scales to the assessment of executive functions. *Applied Neuropsychology: Child, 2*(2), 125-132. doi:10.1080/21622965.2013.748389

Kline, P. (1979). *Psychometrics and psychology*. London, UK: Academic Press.

Lai, M. C., & Baron-Cohen, S. (2015). Identifying the lost generation of adults with autism spectrum conditions. *The Lancet Psychiatry, 2*(11), 1013-1027. doi:10.1016/S2215-0366(15)00277-1

Lai, M. C., Lombardo, M. V., Ruigrok, A. N., Chakrabarti, B., Wheelwright, S. J., Auyeung, B., ... Baron-Cohen, S. (2012). Cognition in males and females with autism: similarities and differences. *PLoS ONE, 7*(10), e47198. doi:10.1371/journal.pone.0047198

Mandy, W., Chilvers, R., Chowdhury, U., Salter, G., Seigal, A., & Skuse, D. (2012). Sex differences in autism spectrum disorder: Evidence from a large sample of children and adolescents. *Journal of Autism and Developmental Disorders, 42*(7), 1304-1313. doi:10.1007/s10803-011-1356-0

McAuley, T., Chen, S., Goos, L., Schachar, R., & Crosbie, J. (2010). Is the behavior rating inventory of executive function more strongly associated with measures of impairment or executive function? *Journal of the International Neuropsychological Society, 16*(3), 495-505. doi:10.1017/S1355617710000093

Nunnally, J. C. (1978). *Psychometric theory* (2nd ed.). New York, NY: McGraw-Hill.

Office for National Statistics (2016). 2011 Census aggregate data. Retrieved from: https://www.ons.gov.uk/census/2011census

Otero, T. M., & Barker, L. A. (2014). The frontal lobes and executive functioning. In S. Goldstein & J. A. Naglieri (Eds.), *Handbook of executive functioning* (pp. 29-44). New York, NY: Springer.

Pellicano, E., Dinsmore, A., & Charman, T. (2014). What should autism research focus upon? Community views and priorities from the United Kingdom. *Autism, 18*(7), 756-770. doi:10.1177/1362361314529627

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. doi:10.1007/s10803-014-2309-1

Salthouse, T. A. (2005). Relations between cognitive abilities and measures of executive functioning. *Neuropsychology, 19*(4), 532-545. doi:10.1037/0894-4105.19.4.532

Streiner, D. L., Norman, G. R., & Cairney, J. (2015). *Health measurement scales: A practical guide to their development and use* (5th edition). Oxford, UK: Oxford University Press.

Appendix A

Quality Appraisal

Study ID:

Criteria		Yes (1)	No (0)	Can't tell/not reported (0)	Comments
1	Are the aims/objectives of the study clearly described?				
2	Is the study design clearly described and appropriate?				
3	Were the clinical and control groups recruited in appropriate way (with clearly defined inclusion and exclusion criteria)?				
4	Did the clinical group have a diagnosed autism spectrum disorder meeting diagnostic criteria?				
5	Was autism symptomatology screened in the control group using an appropriate method?				
6	Were the clinical and control groups matched on age, gender and FSIQ?				
7	Are the outcome variable(s) clearly defined and were they measured using appropriate tools and procedures?				
8	Does the study report the results of all outcome variable(s) in full?				
9	Was the sample size appropriate / did the study have sufficient power?				
10	Are the analytic methods described and appropriate?				
11	Are the main findings of the study clearly described?				
12	Are the conclusions supported by the results?				

Total Score: /12

Appendix B

Data Extraction Form

Background:

Reference number	
Title	
First author	
Publication year	
Country	

Participant Recruitment:

	ASD group	TD group
ASD diagnoses included		N/A
Validity of diagnosis (inc. diagnostic manual)		N/A
Diagnostic screening/verification of ASD diagnosis		
IQ inclusion criteria		
Independent verification of IQ		
Age inclusion criteria		
Further inclusion/exclusion criteria		
Recruitment method		

Sample Demographics:

	ASD group	TD group
Number of participants		
Age (M, SD, range)		
Male:female ratio		
IQ (M, SD, range)		
Groups matched (IQ, gender, age, SES)		
BRIEF parent or teacher report		

BRIEF results:

Variable	N	ASD mean (SD)	N	TD mean (SD)
BRI				
MI				
GEC				

Form that data is in e.g. *T* score	
Data reported in paper	

Appendix C

Ethical Approval

UCL RESEARCH ETHICS COMMITTEE
ACADEMIC SERVICES

7 March 2016

Dr William Mandy
Research Department of Clinical, Educational and Health Psychology
UCL

Dear Dr Mandy

Notification of Ethical Approval
Project ID: 8057/001: Developing an ecologically-valid informant report measure of executive function for use with children with Autistic Spectrum Disorder

I am pleased to confirm in my capacity as Chair of the UCL Research Ethics Committee that I have approved your study for the duration of the project i.e. until **1st September 2017**.

Approval is subject to the following conditions:

1. You must seek Chair's approval for proposed amendments 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

2. 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. The adverse event will be considered at the next Committee meeting and a decision will be made on the need to change the information leaflet and/or study protocol.

   For non-serious adverse events the Chair or Vice-Chair of the Ethics Committee should again be notified via the Ethics Committee Administrator (ethics@ucl.ac.uk) within ten days of an adverse 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.

On completion of the research you must submit a brief report of your findings/concluding comments to the Committee, which includes in particular issues relating to the ethical implications of the research.

Yours sincerely

[redacted]

Professor John Foreman
Chair of the UCL Research Ethics Committee

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

Appendix D

Participant Information Sheets – Study 1

Parent Participants

Participant Information Sheet – for parents taking part in interviews

Title: Developing a new questionnaire to measure executive function in children with Autism Spectrum Disorder

What is the study about?

This study is interested in planning and organisation abilities (also known as 'executive functioning') in children with Autism Spectrum Disorder (ASD). Executive functioning includes things like planning, organisation and working memory. Research shows that children with ASD often find these areas difficult. There are already tests and questionnaires which measure executive functioning but none of them were designed specifically for children with ASD. This study, therefore, aims to develop and pilot a new questionnaire looking at these skills in children with ASD.

Who can take part?

- We invite parents of school-aged children with an Autism Spectrum Disorder (ASD) to participate. We ask that your child has received a diagnosis such as ASD, Autism, or Asperger's Syndrome from a mental health or medical professional.
- In this study we are looking to recruit parents of children with ASD who do not have a general intellectual disability.
- You need to be able to speak fluent English.

What will happen if I agree to take part?

We want to ask parents about how their child with ASD plans and organises their behaviour to help us design a new questionnaire focused on this. If you agree to take part, you will be asked to attend an interview with the researcher in which you will be asked questions about your child and their day-to-day functioning. This will include questions about how your child plans and organises his/her behaviour. This interview will be audio recorded and will take place at UCL or at a location more convenient for you.

Are there any risks or inconveniences of taking part?

We do not anticipate any particular risks involved in taking part in this study. If, however, you need support after taking part please contact the researcher. This study has been approved by the UCL Research Ethics Committee (Project ID Number): 8057/001

What are the possible benefits of taking part?

We hope that this study will help us to further understand executive functioning in children with ASD. A new questionnaire will help us to more accurately assess the nature of children's difficulties and strengths in this area. If you would like to receive a copy of the final report please let us know.

Do I have to take part?

No, it is totally up to you to decide whether or not to take part; choosing not to take part will not disadvantage you in any way. If you do decide to take part you are still free to withdraw at any time and without giving a reason. If you decide to take part you will be given this information sheet to keep and be asked to sign a consent form.

University College London, Gower Street, London WC1E 6BT
Tel: +44 (0)20 7679 2000
email@ucl.ac.uk
www.ucl.ac.uk

What will happen to the information I provide?

Interviews will be audio recorded and stored securely in encrypted files. Once they have been analysed the recordings will be deleted. All data will be collected and stored in accordance with the Data Protection Act (1998). This means the information collected will be anonymised and stored confidentially.

How to contact us?

Please discuss the information above with others if you wish. Please ask us if there is anything that is not clear or if you would like more information.

Laurie Ledger-Hardy, Trainee Clinical Psychologist,
Research Department of Clinical, Educational and Health Psychology,
1-19 Torrington Place,
London, WC1E 6BT
[redacted]

Supervised by Dr Will Mandy, Senior Lecturer and Research Director,
UCL Clinical Psychology Course
Email: [redacted]

Thank you for taking the time to read this information sheet and considering taking part in this research.

Professional Participants

Participant Information Sheet – for clinicians in the interview phase

Title: Developing a new questionnaire to measure executive function in children with Autism Spectrum Disorder

What is the study about?

This study is interested in executive functioning skills in children with Autism Spectrum Disorder (ASD). Executive functioning includes things like planning, organisation and working memory, and research shows that children with ASD often find these areas difficult. However, none of the existing tests or questionnaires measuring executive functioning were designed specifically for children with ASD. This study, therefore, aims to develop and pilot a new questionnaire to measure executive functioning in school-aged children with ASD. We plan to consult clinicians and parents in the development of this questionnaire and hope that this method leads to a more ecologically valid measure.

Who can take part?

- In this stage of the study we are looking to consult clinicians who work with children with ASD and have experience using measures of executive function.
- The current study is focused on school-aged children with a diagnosis of a ASD who do not have a learning disability.

What will happen if I agree to take part?

If you agree to take part, you will be asked to attend a short interview with the researcher. We want to ask you for your experience of using existing measures of executive functioning, such as the BRIEF (Behaviour Rating Inventory of Executive Functioning) when working with children with ASD. We are particularly interested to hear what you feel is and isn't captured in existing measures and how useful they are for this population. This interview will be audio recorded and can take place at UCL or at a location more convenient for you.

Are there any risks or inconveniences of taking part?

We do not anticipate any particular risks to clinicians taking part in this study. This study has been approved by the UCL Research Ethics Committee (Project ID Number): 8057/001

What are the possible benefits of taking part?

We hope that a new questionnaire will help us to more accurately assess the nature of children's executive functioning difficulties, in particular in their day-to-day lives. If you would like to receive a copy of the final report please let us know.

Do I have to take part?

No, it is totally up to you to decide whether or not to take part; choosing not to take part will not disadvantage you in any way. If you do decide to take part you are still free to withdraw at any time and without giving a reason. If you decide to take part you will be given this information sheet to keep and be asked to sign a consent form.

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How to contact us?

Please discuss the information above with others if you wish. Please ask us if there is anything that is not clear or if you would like more information.

Laurie Ledger-Hardy, Trainee Clinical Psychologist,
Research Department of Clinical, Educational and Health Psychology,
1-19 Torrington Place,
London, WC1E 6BT
[redacted]

Supervised by Dr Will Mandy, Senior Lecturer and Research Director,
UCL Clinical Psychology Course
[redacted]

Thank you for taking the time to read this information sheet and considering taking part in this research.

Appendix E

Information Sheets – Study 2

Parents of Children with ASD

Participant Information Sheet – for parents taking part in the online questionnaire

Title: Developing a new questionnaire to measure executive function in children with Autism Spectrum Disorder

What is the study about?
This study is interested in planning and organisation abilities (also known as 'executive functioning') in children with Autism Spectrum Disorder (ASD). Executive functioning includes things like planning, organisation and working memory. Research shows that children with ASD often find these areas difficult. There are already tests and questionnaires which measure executive functioning but none of them were designed specifically for children with ASD. This study, therefore, aims to develop and pilot a new questionnaire looking at these skills in children with ASD.

Who can take part?
- We invite parents of school-aged children with an Autism Spectrum Disorder (ASD) to participate. We ask that your child has received a diagnosis such as ASD, Autism, or Asperger's Syndrome from a mental health or medical professional.
- In this study we are looking to recruit parents of children with ASD who do not have a general intellectual disability.
- You need to be able to speak fluent English.

What will happen if I agree to take part?
We want to pilot a new questionnaire online which is looking at the executive functioning skills of children with ASD. If you agree to take part, you will first be asked to provide some background details about yourself and your child. You will then be asked to complete the online questionnaire. The questionnaire we are piloting will ask you questions about your child's day to day behaviours, such as how they plan and organise their behaviour. A second questionnaire will ask about your child's symptoms of ASD. A sub-group of parents will be asked to complete the online questionnaires on two separate occasions.

Are there any risks or inconveniences of taking part?
We do not anticipate any particular risks involved in taking part in this study. If, however, you need support after taking part please contact the researcher. This study has been approved by the UCL Research Ethics Committee (Project ID Number): 8057/001

What are the possible benefits of taking part?
We hope that this study will help us to further understand executive functioning in children with ASD. A new questionnaire will help us to more accurately assess the nature of children's difficulties and strengths in this area. If you would like to receive a copy of the final report please let us know.

Do I have to take part?
No, it is totally up to you to decide whether or not to take part; choosing not to take part will not disadvantage you in any way. If you do decide to take part you are still free to withdraw at any time and without giving a reason. If you decide to take part you will be given this information sheet to keep and be asked to sign a consent form.

University College London, Gower Street, London WC1E 6BT
Tel: +44 (0)20 7679 2000
email@ucl.ac.uk
www.ucl.ac.uk

What will happen to the information I provide?
All data will be collected and stored in accordance with the Data Protection Act (1998). This means the information collected will be anonymised and stored confidentially.

How to contact us?
Please discuss the information above with others if you wish. Please ask us if there is anything that is not clear or if you would like more information.

Laurie Ledger-Hardy, Trainee Clinical Psychologist,
Research Department of Clinical, Educational and Health Psychology,
1-19 Torrington Place,
London, WC1E 6BT
E[redacted]

Thank you for taking the time to read this information sheet and considering taking part in this research.

Parents of TD Children

Participant Information Sheet – for parents of typically developing children taking part in the online questionnaire

Title: Developing a new questionnaire to measure executive function in children with Autism Spectrum Disorder

What is the study about?
This study is interested in planning and organisation abilities (also known as 'executive functioning') in children with Autism Spectrum Disorder (ASD). Executive functioning includes things like planning, organisation and working memory. Research shows that children with ASD often find these areas difficult. There are already tests and questionnaires which measure executive functioning but none of them were designed specifically for children with ASD. This study, therefore, aims to develop and pilot a new questionnaire looking at these skills in children with ASD. To check whether the new questionnaire is useful we also want to pilot it with children who do not have ASD.

Who can take part?
- We invite parents of "typically developing" school-aged children to take part – this means children who do not have a developmental or learning disorder such as Autism Spectrum Disorder (ASD), Attention Deficit Hyperactivity Disorder (ADHD), or a learning disability.
- You need to be able to speak fluent English.

What will happen if I agree to take part?
We want to pilot a new questionnaire online which is looking at the executive functioning skills of children with and without ASD. If you agree to take part, you will first be asked to provide some background details about yourself and your child. You will then be asked to complete the online questionnaire. The questionnaire we are piloting will ask you questions about your child's day to day behaviours, such as how they plan and organise their behaviour. A sub-group of parents will be asked to complete another questionnaire asking similar questions. Another sub-group of parents will be asked to complete the new online questionnaires on two separate occasions.

Are there any risks or inconveniences of taking part?
We do not anticipate any particular risks involved in taking part in this study. If, however, you need support after taking part please contact the researcher. This study has been approved by the UCL Research Ethics Committee (Project ID Number): 8057/001

What are the possible benefits of taking part?
We hope that this study will help us to further understand executive functioning in children with ASD. A new questionnaire will help us to more accurately assess the nature of children's difficulties and strengths in this area. If you would like to receive a copy of the final report please let us know.

Do I have to take part?
No, it is totally up to you to decide whether or not to take part; choosing not to take part will not disadvantage you in any way. If you do decide to take part you are still free to withdraw at any time and without giving a reason. If you decide to take part you will be given this information sheet to keep and be asked to sign a consent form.

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What will happen to the information I provide?
All data will be collected and stored in accordance with the Data Protection Act (1998). This means the information collected will be anonymised and stored confidentially.

How to contact us?
Please discuss the information above with others if you wish. Please ask us if there is anything that is not clear or if you would like more information.

Laurie Ledger-Hardy, Trainee Clinical Psychologist,
Research Department of Clinical, Educational and Health Psychology,
1-19 Torrington Place,
London, WC1E 6BT
[redacted]

Supervised by Dr Will Mandy, Senior Lecturer and Research Director,
UCL Clinical Psychology Course
[redacted]

Thank you for taking the time to read this information sheet and considering taking part in this research.

Appendix F

Consent Form

Informed Consent Form for Participation in Research Studies

Please complete this form after you have read the Information Sheet and/or listened to an explanation about the research.

Title of Project: Developing a new questionnaire to measure executive function in children with Autism Spectrum Disorder

This study has been approved by the UCL Research Ethics Committee (Project ID Number): 8057/001

Thank you for your interest in taking part in this research. Before you agree to take part, the person organising the research must explain the project to you.

If you have any questions arising from the Information Sheet or explanation already given to you, please ask the researcher before you to decide whether to join in. You will be given a copy of this Consent Form to keep and refer to at any time.

Participant's Statement

I

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

Signed: Date:

Name:

Appendix G

Interview Schedules

General interview guidelines:

- Start broad but can narrow as go along - home in on EF difficulties
- Acknowledge other difficulties, listen carefully to whether there are any EF problems within what participant is saying
- Let EF difficulties emerge, use follow up questions re. specific EF areas as appropriate

Semi-structured interview schedule – parents

- Introductions (trainee CP, supervised by Will Mandy); thank you for agreeing to take part; purpose of study – looking at executive function in children with autism, will develop a new questionnaire measure
- Interview today will take between 30 and 60 minutes – is that ok?
- Give information sheet or check participant has read it
- Reiterate confidentiality, recording, data protection, right to withdraw etc.
- Re-check eligibility for study – school aged children (8-16), ASD, no LD
- Sign/send consent form
- Start recording
- Tell me some background about your child – name, age, ASD diagnosis, when received diagnosis, where received diagnosis, type of school attended, any learning difficulties?
- Give parent a brief and broad definition of executive function – ask them to focus on this area
    - We want to learn about EF in autism and how it manifests day to day.
    - Executive functions are a broad set of skills that help us manage life tasks.
    - EF can be viewed like the conductor of an orchestra – organising other cognitive skills in the brain.
    - Executive functions help us plan and organise our behaviour, especially when doing non-routine tasks or working towards a goal.
    - For example, if I need to plan a trip for the next day – EFs used to decide what I need to take, how I will get there, change my plans as go along if necessary, remember things etc.
- Tell me about a typical school day – what things does X have difficulty with?
    - Start with getting ready in the morning
    - During the school day
    - After school
    - Evening/going to bed
- Tell me about a typical non-school/weekend day - what things does X have difficulty with? Anything different to school day?
- Focus in on specific EF areas as they emerge. Use follow up questions around specific EFs as necessary

Semi-structured interview schedule – professionals

- Introductions (trainee CP, supervised by Will Mandy); thank you for agreeing to take part; purpose of study – looking at executive function in children with autism, will develop a new questionnaire measure
- Interview today will take between 30 and 60 minutes – is that ok?
- Give information sheet or check they've read it
- Reiterate confidentiality, recording, data protection, right to withdraw etc.
- Re-check eligibility for study – working in field of ASD
- Sign/send consent form
- Start recording
- From your clinical work what EF difficulties do you observe in children with ASD?
    - Focus in on specific EFs as necessary
- Have you used informant-report questionnaire measures such as the BRIEF? Strengths and weaknesses of the BRIEF/existing measures?
- What would a useful measure of EF in ASD need to capture? What would it need to be like?

Appendix H

Questionnaire Order as Administered

Table H1

Original 48 items in Order Administered

No	Question	Scale
1	My child finds it difficult to independently pack his/her school bag with what he/she needs	P
2	My child forgets to take important items (e.g. completed homework) to school	M
3	If my child is interrupted whilst doing a task or activity, he/she has difficulty going back to it	FA
4	If given instructions to do a task with several stages, my child may only do the first or last part	M
5	My child finds unstructured activities (e.g. tidying room, creative writing task) difficult as he/she doesn't know how to start	GG
6	My child has difficulty organising him/herself to get ready in the morning	P
7	My child loses track of time easily	SA
8	My child misjudges how long it will take to complete everyday tasks, such as getting ready to leave the house	SA
9	My child forgets where he/she has put things at home	M
10	My child needs everyday tasks and instructions (e.g. getting dressed, tidying room) broken down into small steps	C
11	If my child can do a task (e.g. a maths problem) one way, he/she struggles to learn a different way of doing it	RI
12	My child has meltdowns/extreme emotional outbursts	ME
13	My child has difficulty noticing his/her own mistakes when doing schoolwork/homework	SA
14	My child finds it difficult to adjust his/her behaviour when plans change (e.g. if being picked up from school in new location would still go to usual location)	C
15	My child finds instructions with three or more parts difficult to follow (e.g. "go upstairs and get x, y and z")	M
16	My child loses things or leaves things at school all the time (e.g. PE kit)	M
17	When my child loses something, he/she struggles to independently think of places it could be	GG

18	My child takes a long time to get started on everyday tasks (e.g. washing, getting dressed)	GG
19	My child misunderstands tasks (e.g. in class, homework) as focuses on specific details rather than the overall picture	FA
20	My child has difficulty focusing on important tasks he/she is not interested in	FA
21	My child finds non-specific instructions (e.g. “go and get ready”) hard to put into action	GG
22	My child does everyday activities in a set way (e.g. getting dressed in a particular order)	C
23	My child finds it difficult to have all the things he/she needs for a lesson at school	P
24	My child finds it difficult to switch between tasks and activities	RI
25	My child goes to another room and forgets what he/she went there for	M
26	If something unexpected happens, my child has trouble coming up with ideas about how to react	G
27	My child finds it difficult to adjust his/her behaviour based on feedback	RI
28	My child finds it difficult to get to lessons on time	P
29	My child has difficulty deciding what to do first when doing his/her homework	P
30	When given instructions for a task, my child goes ahead with the first part without paying attention to the rest of the instructions	FA
31	My child finds it difficult to change his/her plan or point of view as a result of new information	RI
32	When my child wants to do something, he/she needs to do it immediately	FA
33	My child finds it difficult to independently get started on tasks or activities	GG
34	My child interrupts others or blurts things out	FA
35	When my child has free time, he/she finds it difficult to come up with new ideas for what to do	GG
36	My child has to follow routines in daily life, in order to remember what he/she needs to do	C
37	My child has difficulty multi-tasking (e.g. listening to teacher’s instructions and writing them down)	FA
38	My child forgets to check whether he/she has items that they need every day (e.g. lunch card, planner)	M
39	My child has difficulty sustaining his/her attention (e.g. in class)	FA

40	My child needs many prompts when getting ready for school in the morning	C
41	My child finds it difficult to do a task on his/her way to doing something else (e.g. if asked to take out the bins on his/her way to get the bus)	RI
42	Changes to my child's routine can throw his/her whole day off course	C
43	My child has difficulty mentally putting something on hold (e.g. something that happened earlier) so that he/she can focus on something else instead	FA
44	My child sticks with one way of doing something even when it is not working	RI
45	My child seems to get upset or angry very quickly when things do not go his/her way	ME
46	My child relies on routines rather than trying new ways, as he/she struggles to come up with new ideas (e.g. always goes the same route around school even though this is not the most efficient)	C
47	After starting a task, my child continues without pausing to check he/she is doing it right	SA
48	My child finds it difficult to plan for an overnight trip (e.g. pack clothes that are appropriate, consider how many changes of clothes are needed)	P

Appendix I

Online Piloting Recruitment Advert

Developing new ways of understanding planning and organisational abilities ('executive function') in children with Autism Spectrum Disorder

- *Are you a parent of a school-aged child with Autism Spectrum Disorder (ASD)?*
- *Are you interested in participating in online research to further our understanding of executive functioning in children with ASD?*

Executive functioning includes things like planning, organisation and working memory and research shows that children with ASD often find these areas difficult. This can have a big impact on their lives at home and school. We have designed a new questionnaire to measure executive functioning in children with ASD – we hope that this will help us better understand these difficulties to improve the help available to children with ASD.

We are looking for parents to complete our new online questionnaire. We hope this new questionnaire will give us valuable information about how children with ASD plan and organise their behaviour day-to-day.

We are looking to recruit parents of school-aged children with ASD to participate in our online research. ASD incudes diagnoses such as 'autism', 'autism spectrum condition', 'Asperger's syndrome', 'Asperger's disorder' and 'Pervasive Developmental Disorder – Not Otherwise Specified' The online questionnaire will take about 25 minutes to complete. Everyone who completes the questionnaire will be entered into a prize draw to win one of three £30 Amazon vouchers.

Furthermore, for every completed questionnaire from the first 120 respondents, a donation of £2 will be made to the National Autistic Society.

If you are interested in taking part, please click on the following link which will take you to further information about the study: *Insert link to study.*

I am a trainee Clinical Psychologist at UCL carrying out research into ASD as part of my doctorate. If you have any questions, please feel free to contact me on [redacted].

Thank you for taking the time to read this.

Please note, all data will be collected and stored in accordance with the Data Protection Act (1998).

Appendix J

Endorsement Frequencies

Table J1

Endorsement Frequencies for 48-item Questionnaire

		strongly disagree (score 1)		slightly disagree (score 2)		neither agree nor disagree (score 3)		slightly agree (score 4)		strongly agree (score 5)	
Item	Sub-scale	Frequency N	Frequency %	Frequency N	Frequency %	Frequency N	Frequency %	Frequency N	Frequency %	Frequency N	Frequency %
Item 10	C	30	30.3030303	17	17.17171717	2	2.02020202	17	17.17171717	33	33.33333333
Item 14	C	31	31.31313131	17	17.17171717	8	8.080808081	23	23.23232323	20	20.2020202
Item 22	C	14	14.14141414	15	15.15151515	12	12.12121212	23	23.23232323	35	35.35353535
Item 36	C	22	22.22222222	20	20.2020202	10	10.1010101	24	24.24242424	23	23.23232323
Item 40	C	18	18.18181818	11	11.11111111	7	7.070707071	29	29.29292929	34	34.34343434
Item 42	C	33	33.33333333	17	17.17171717	7	7.070707071	20	20.2020202	22	22.22222222
Item 46	C	22	22.22222222	21	21.21212121	9	9.090909091	23	23.23232323	24	24.24242424
Item 3	FA	19	19.19191919	21	21.21212121	6	6.060606061	25	25.25252525	28	28.28282828
Item 19	FA	25	25.25252525	21	21.21212121	12	12.12121212	9	9.090909091	32	32.32323232
Item 20	FA	7	7.070707071	13	13.13131313	9	9.090909091	29	29.29292929	41	41.41414141
Item 30	FA	10	10.1010101	22	22.22222222	19	19.19191919	29	29.29292929	19	19.19191919
Item 32	FA	14	14.14141414	17	17.17171717	12	12.12121212	23	23.23232323	33	33.33333333
Item 34	FA	13	13.13131313	20	20.2020202	9	9.090909091	31	31.31313131	26	26.26262626
Item 37	FA	29	29.29292929	15	15.15151515	9	9.090909091	17	17.17171717	29	29.29292929
Item 39	FA	30	30.3030303	19	19.19191919	8	8.080808081	21	21.21212121	21	21.21212121

		strongly disagree (score 1)		slightly disagree (score 2)		neither agree nor disagree (score 3)		slightly agree (score 4)		strongly agree (score 5)	
Item	Sub-scale	Frequency N	Frequency %	Frequency N	Frequency %	Frequency N	Frequency %	Frequency N	Frequency %	Frequency N	Frequency %
Item 43	FA	16	16.16161616	20	20.2020202	12	12.12121212	20	20.2020202	31	31.31313131
Item 5	GG	20	20.2020202	12	12.12121212	8	8.080808081	25	25.25252525	34	34.34343434
Item 17	GG	20	20.2020202	19	19.19191919	8	8.080808081	27	27.27272727	25	25.25252525
Item 18	GG	25	25.25252525	18	18.18181818	5	5.050505051	20	20.2020202	31	31.31313131
Item 21	GG	24	24.24242424	19	19.19191919	6	6.060606061	24	24.24242424	26	26.26262626
Item 26	GG	23	23.23232323	19	19.19191919	10	10.1010101	27	27.27272727	20	20.2020202
Item 33	GG	25	25.25252525	17	17.17171717	7	7.070707071	22	22.22222222	28	28.28282828
Item 35	GG	17	17.17171717	15	15.15151515	10	10.1010101	24	24.24242424	33	33.33333333
Item 2	M	30	30.3030303	15	15.15151515	2	2.02020202	19	19.19191919	33	33.33333333
Item 4	M	25	25.25252525	15	15.15151515	7	7.070707071	22	22.22222222	30	30.3030303
Item 9	M	15	15.15151515	18	18.18181818	6	6.060606061	26	26.26262626	34	34.34343434
Item 15	M	25	25.25252525	19	19.19191919	8	8.080808081	15	15.15151515	32	32.32323232
Item 16	M	20	20.2020202	21	21.21212121	6	6.060606061	21	21.21212121	31	31.31313131
Item 25	M	29	29.29292929	16	16.16161616	18	18.18181818	21	21.21212121	15	15.15151515
Item 38	M	26	26.26262626	17	17.17171717	8	8.080808081	20	20.2020202	28	28.28282828
Item 12	ME	23	23.23232323	20	20.2020202	4	4.04040404	22	22.22222222	30	30.3030303
Item 45	ME	16	16.16161616	20	20.2020202	9	9.090909091	17	17.17171717	37	37.37373737
Item 1	P	37	37.37373737	12	12.12121212	6	6.060606061	16	16.16161616	28	28.28282828
Item 6	P	32	32.32323232	15	15.15151515	4	4.04040404	20	20.2020202	28	28.28282828
Item 23	P	30	30.3030303	20	20.2020202	13	13.13131313	15	15.15151515	21	21.21212121

		strongly disagree (score 1)		slightly disagree (score 2)		neither agree nor disagree (score 3)		slightly agree (score 4)		strongly agree (score 5)	
Item	Sub-scale	Frequency N	Frequency %	Frequency N	Frequency %	Frequency N	Frequency %	Frequency N	Frequency %	Frequency N	Frequency %
Item 28	P	38	38.38383838	16	16.16161616	19	19.19191919	15	15.15151515	11	11.11111111
Item 29	P	28	28.28282828	22	22.22222222	12	12.12121212	17	17.17171717	20	20.2020202
Item 48	P	24	24.24242424	19	19.19191919	14	14.14141414	12	12.12121212	30	30.3030303
Item 11	RI	11	11.11111111	19	19.19191919	13	13.13131313	25	25.25252525	31	31.31313131
Item 24	RI	22	22.22222222	25	25.25252525	5	5.050505051	23	23.23232323	24	24.24242424
Item 27	RI	20	20.2020202	21	21.21212121	11	11.11111111	27	27.27272727	20	20.2020202
Item 31	RI	18	18.18181818	21	21.21212121	6	6.060606061	31	31.31313131	23	23.23232323
Item 41	RI	18	18.18181818	19	19.19191919	13	13.13131313	24	24.24242424	25	25.25252525
Item 44	RI	17	17.17171717	23	23.23232323	13	13.13131313	26	26.26262626	20	20.2020202
Item 7	SA	15	15.15151515	18	18.18181818	9	9.090909091	23	23.23232323	34	34.34343434
Item 8	SA	16	16.16161616	19	19.19191919	7	7.070707071	23	23.23232323	34	34.34343434
Item 13	SA	15	15.15151515	26	26.26262626	12	12.12121212	27	27.27272727	19	19.19191919
Item 47	SA	16	16.16161616	18	18.18181818	14	14.14141414	29	29.29292929	22	22.22222222