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Executive Dysfunction in High Functioning Autism

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Submitted in part fulfilment of the degree of doctorate in Clinical Psychology at the University of Edinburgh

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Thesis Abstract

Background: There is presently a lack of consistency in research designed to measure executive functioning (EF) in autism that may be attributable to lack of homogeneity or comorbid conditions (i.e. learning disability or additional diagnosis) in test samples.

Aim: A systematic review focused on a subset of EF (verbal fluency: VF) was conducted, using only studies of high-functioning individuals with autism (HFA) without an additional diagnosis or learning disability. An empirical study was conducted comparing the executive functioning profile of individuals with HFA and typically developed (TD) individuals.

Method: For the systematic review, 16 studies met the specified inclusion criteria, depicting 15 semantic (category), 14 phonological (letter), and 6 switching (categories) VF tasks. In order to assess potential bias, the available VF information of the included papers was scrutinised by the author and an independent clinical practitioner. For the empirical paper, 22 HFA and 22 TD participants (mean age = 28, range = 17-73, 52% male) without a comorbid condition, learning disability or brain injury completed three subtests from the WAIS-IV (vocabulary, block design and digit span) and all subtests of the Delis–Kaplan Executive Functioning System (D-KEFS).

Results: For the systematic review, a minority of semantic and phonological VF studies reported a significant difference between typically developed and HFA populations. Five of the six semantic switching studies reported a significant difference between groups. All papers included were of good or adequate quality and inter-rater reliability was high. For the empirical paper, the HFA group performed significantly poorer on the switching condition of the design fluency task, semantic conditions of the verbal fluency task and on the word context task overall. No other significant differences were observed.

Summary: Although the systematic review concluded that there was insufficient evidence to support that disfluency can be attributed to autistic symptomology, the empirical study found that the HFA group performed poorer than TD in semantic VF and other subtests designed to measure generating novel 'imaginative' ideas, without visual cues to aid performance. The deficit on these subtests was increased when there was the added condition requiring the participant to switch between newly formed concepts.

Conclusions: Although in VF, results are mixed, the empirical study demonstrates that even in a group of high-functioning individuals there are still measurable differences in EF between TD and HFA samples that may not be apparent through more general cognitive testing. Implications for using a neuropsychological profile for adults with HFA are discussed.

Systematic Review

Title:

A Systematic Review of Verbal Fluency in High Functioning Autism

Running title:

Verbal Fluency in HFA

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Author contributions:

Hollie Burnett designed and conducted the review under the academic supervision of Dr Jill Cossar and Dr Suzanne O'Rourke, who assisted with the design and research question and provided comments on the final manuscript.

Word count (excluding tables, figures and references): 4493

This systematic review was prepared in accordance with the author specifications for the journal 'Autism Research'.

Abstract

Aim: There is lack of consistency found in research designed to measure verbal fluency (VF) in autism that may be attributable to lack of homogeneity in testing samples. The aim of this review is to investigate VF studies using only high-functioning individuals with autism (HFA), without an additional diagnosis or learning disability to determine whether there is a VF deficit in ASD independent of a comorbid condition. *Method:* 16 studies met the specified inclusion criteria, depicting 15 semantic (category), 14 phonological (letter), and six switching (categories) VF tasks. In order to assess potential bias, the available VF information of the included papers was scrutinised by the author and an independent clinical practitioner. *Results:* Only five semantic and five phonological VF studies reported a significant difference between typically developed and HFA populations. Five of the six semantic switching studies reported a significant difference between groups. All papers included were of good or adequate quality and inter-rater reliability was high.

Summary: When studies measuring VF in HFA populations without other covariates (i.e. IQ below 70, additional diagnosis) were investigated, very little evidence for disfluency was found. Although switching between categories produced the most consistent deficit, only a small number of studies include this condition and the result is confounded by the demand it places on other executive skills (i.e. set shifting). It is concluded that there is insufficient evidence to support that disfluency can be attributed to autistic symptomology.

Key words

Autism, Asperger, verbal fluency, semantic, phonological, category fluency, letter fluency, word generation, generativity, executive functioning, language

Introduction

It is proposed that many of the social, communication and flexibility of thought difficulties experienced by individuals with autism spectrum disorder (ASD) can be attributed to higher-level cognitive difficulties, such as planning, fluency, inhibition, attention, problem-solving and monitoring (e.g. Stuss & Benson, 1986; Russell, Jarold & Hood, 1999; Hill, 2004; Pellicano, 2006), using the umbrella term 'executive dysfunction' (Ozonoff et al., 1991).

Executive dysfunction can be seen to underlie many of the key characteristics of autism, both in the social and non-social domains. The behaviour problems addressed by this theory are rigidity and perseveration, being explained by a poverty in the

initiation of new non-routine actions and the tendency to become stuck in a given task set (Hill, 2004).

Support for this theory comes from tasks designed to measure executive functioning, which have yielded significant deficits for ASD, for example, the Wisconsin card sorting task (Ozonoff, Pennington & Rogers, 1991), in which the subject is required to form/switch concepts, and the tower of Hanoi or London (Shallice, 1982), in which the subject has to solve problems by planning before acting (Hughes, Russell & Robbins, 1994).

As well as psychometric assessments, neuro-imaging studies have found deficits in prefrontal and subcortical brain areas in relation to executive functioning tasks in ASD (Philip et al., 2012). Abnormalities in the prefrontal cortex and its connections to other brain structures such the middle frontal gyrus, basal ganglia, striatum, and cerebellum have been found in individuals with autism (Duncan & Owen, 2000; Philip et al., 2012).

Whilst much research attention has being paid to the areas of executive functioning, which have more consistently found to be lacking in those with ASD, such as set-shifting and planning, verbal fluency (VF) has being somewhat neglected and has produced more variations in performance, with some studies reporting a profound deficit (e.g. Verté et al., 2005) and other studies showing no significant difficulties in those with ASD (e.g. Robinson et al., 2009). VF, often called 'generativity', is an important subset of executive functioning. VF tasks are often included in

neuropsychological assessment, in clinical practice, and in research (Oriá et al., 2013). VF is often used as one of the most sensitive tests for identifying cerebral dysfunction (Benton, 1968; Ruff et al., 1997) and although there is little consensus with ASD, the clinical utility has being demonstrated in various other clinical populations, such as individuals with schizophrenia (Joyce et al., 1996), attention-deficit hyperactivity disorder (ADHD; Andreou & Trott, 2013) and those with neurodegenerative diseases, such Alzheimer's (Gomez & White, 2004), Parkinson's (Henry & Crawford, 2006) and Huntington's (Ho et al., 2002) disease.

VF refers to the ability of being able to verbally access and recall words that relate to a specific subset of information. For this individuals must first recognise a concept, then draw links between that and related demographic items. Typical VF tasks involve a participant being asked to produce as many unique words within a specific category (semantic fluency) or that begin with a specific letter (phonological fluency) within one minute (e.g. Benton, 1968; Turner, 2009). The most common exemplar categories include animal species, boys' names, fruits/vegetables, food, countries and words beginning with specific letters 'F, A, S' (e.g. Turner, 1999, Delis et al., 2001). Some batteries include a semantic switching task, which involves the participant producing as many words that belong to two separate categories alternately, such as fruit and furniture (e.g. Delis et al., 2001). The participant's score in each task is the number of unique correct words. Some VF measures convert overall mean scores into scaled scores for the purpose of comparing tasks within a battery of executive functioning assessments (e.g. Delis-Kaplan Executive Functioning System, Delis et al., 2001).

To measure VF, studies tend to use either a letter or a category fluency task, combine both measures to make one overall performance score, or use the switching condition. However, combining or comparing scores from phonological, semantic and switching fluencies may prove problematic, as it is argued that they have different mechanisms for retrieving verbal knowledge, and are possibly mediated by different regions of the prefrontal cortex (Szatkowska et al., 2000). It is therefore possible that where one aspect of VF may be impaired, the other may be relatively intact (Henry & Crawford, 2006). It may also be that this accounts for the variation in reported performance in those with ASD as one area of VF may be impaired, whereas other areas may not.

Similar to concept formation, semantic VF involves the ability to put words into categories, by grouping targets according to meaningful and conceptual features, which has being shown to be a deficit in those with ASD (Burnett & Jellema, 2013). Temple Grandin (1995) described the difficulties individuals with ASD have in linking together previously learned concepts based on a specified commonality. Phonological deficits may also be explained by the semantic pragmatic difficulties (such as word order errors, word category errors, verb tense errors) experienced by individuals with ASD **(**Adams & Bishop, 1989; Bishop & Norbury, 2002).

Lack of consensus may be also be explained by the wide variety of communication and cognitive deficits individuals with ASD have across the spectrum. As many studies include data with confounding participant variables, such as very low

cognitive ability or other comorbid condition(s) such as schizophrenia (e.g. Joyce et al., 1996; Barnard et al., 2008; Kover & Abbeduto, 2010), task understanding, verbal ability or a comorbid diagnosis which may falsely identify a pattern of disfluency associated with autistic symptomology. A lack of reported consistency has meant that most executive functioning tests, including VF, cannot be included in ASD assessments.

There is a need for more reliable ASD screening tools in clinical assessment to distinguish ASD from other conditions, particularly in high-functioning individuals who are able to develop coping strategies to mask presenting difficulties. It is also important to better understand the processes that individuals use to retrieve verbal information, particularly in those with high-functioning autism (HFA) and Asperger Syndrome (AS), where there is similar vocabulary skills to TD individuals. There may be different mechanisms for abstracting semantic and phonological information as they require the individual to not just establish the learned word definition, but to construct either 'meaning' or 'sound'. It is therefore important to conduct a review detailing what clear evidence there is for disfluency in HFA/AS independently of other conditions, by comparing performance on semantic, phonological and switching tasks separately.

In summary, there is a lack of clear evidence that individuals with ASD, without a comorbid condition (i.e. learning disability, traumatic brain injury, neurological disorder) have disfluency, and if they do, what aspects of VF (semantic/phonological/switching) underlies the impairment. Hence, the aim of this

review is to investigate available literature with the performance of non-learning disabled ASD populations without a comorbid condition on VF tasks. This will contribute to our understanding of the mechanisms that underlie ASD, particularly regarding executive functioning, language and communication.

Methodology and selection criteria

Inclusion criteria

1) All studies included in this review stated that individuals tested had a diagnosis of ASD, high-functioning autism or Asperger syndrome. For the purposes of this review, all participants will collectively be referred to as having 'high-functioning autism' (HFA). HFA is not a term used by the DSM-IV or ICD-10, but as a majority of published papers do not take a detailed developmental history, including early language delay, it is difficult to use the DSM-V criteria for Asperger syndrome. Asperger syndrome is a controversially applied diagnostic term and has since been removed from the DSM-V (2013) criteria. Some argue that it is unhelpful to have two separate diagnostic terms and ASD should be used as an umbrella term to describe all individuals with the same areas of presenting difficulties. 'High-functioning' is a term used in this review for the purposes of describing higher cognitively able individuals with ASD who do not have a learning disability.
2) It was also important that all studies included a cognitive screening measure to confirm that participants did not have an intellectual impairment (i.e. they must

have an IQ of above 70), to ensure task understanding and to ensure language ability sufficient to complete the VF task.

3) As ASD is a lifelong condition and VF is a measure used with individuals as young eight, studies using participants of all ages (8 years +) were included.
4) Only tasks recording verbal responses to a given letter or category were used in the analysis. This was important to establish VF from other fluencies and skills, such as reading ability, working memory and visual scanning.
5) All papers that included a VF measure (irrespective of whether this was the primary research question or what design was imposed) were considered for inclusion.
6) Studies of all sample sizes were considered for inclusion.

Exclusion criteria

1) Studies were excluded if the participants had an intellectual impairment (IQ below 70, confirmed by cognitive assessment, or if no cognitive assessment was completed to determine this) or comorbid condition (e.g. ADHD, dyspraxia, schizophrenia, and Tourette's syndrome). This was to ensure that any potential clinical difference could not be attributable to another diagnosis.
2) As the purpose of this review is to examine VF as an executive function, other fluencies such as reading fluency were excluded from the analysis.
3) All papers not translated into English were also excluded from the analysis.
4) All unpublished papers were excluded from the analysis.

Search strategy for identification of studies

Literature was initially scoped and search terms developed with reference to common key words denoting ASD (including exact terms: Asperger, autis*) AND words denoting VF (including exact terms: word generation, generatively, verbal, semantic, category, animal, phonological, word fluency). Systematic searches of the following electronic databases were conducted: EMBASE, Medline, PsychINFO, PubMed, Web of Science, SciElo and Ovid. Reference lists of included studies were also searched. No restriction on publication date was imposed. Following search completion, titles and abstracts were screened to remove studies not meeting the inclusion criteria. The remaining studies were read in full.

Methodological quality

The quality of the selected papers was assessed by evaluating the available VF information. For the purposes of this review, the important information to assess for bias was the participant sample, examination process, tests used and statistical design. An assessment tool was adapted to focus specifically on these areas, using the questions relating to methodology and statistics from the following standardised checklists: the QUADAS tool (Whiting et al., 2003), the Scottish Intercollegiate Guidelines Network Methodology Checklist (SIGN, 2007) and STARD guidelines for reporting of diagnostic studies (Bossuyt et al., 2003). Information about the abstract, reflections in the discussion or the paper title was decided to be irrelevant to the aims of this review.

The final assessment tool (see Appendix 1) has a total score of eighteen derived from the following domains: participant and recruitment (8 points); measures (4 points); power and analysis (6 points). A three-choice rating scale was applied where each item was coded according to the following ratings: well addressed =2, adequately addressed =1, poorly/not addressed =0. Quality ratings for all papers were made by the author of this review (HB). The papers were then alphabetically ordered based on the first author's surname into two piles, then an independent clinical practitioner (AS) was asked to choose one half to assess for quality (the latter half, consisting of nine papers).

Results

Outcome of search process

Electronic databases were searched on 1st March 2015, with 3019 studies retrieved. A further seven studies were identified from the internet search engine 'Google'. This process is summarised in Figure 1.

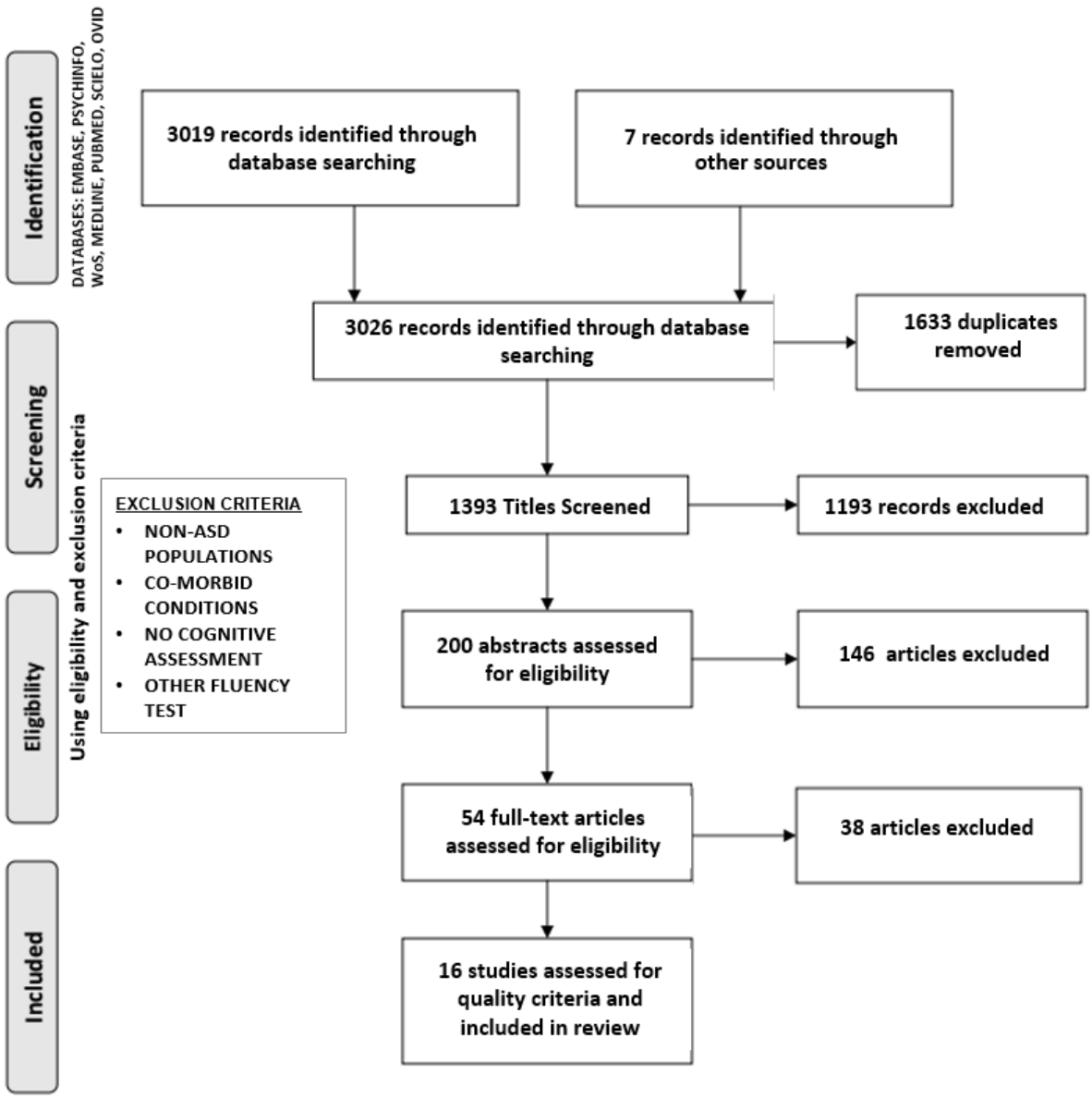

Figure 1. Flow chart of systematic search and study selection process

Included study characteristics

Study demographic information is presented in Table 1. This systematic review included data from a total of 530 participants, from six countries, using nine different measures of semantic, phonological and semantic switching VF. Clinical sample sizes varied from 12 to 63. Most of the studies used a comparison group, with the

only exception being Kleinhans et al. (2005). For a majority of articles the primary aim was not to investigate VF; the main aim of Ditcher et al. (2009) was to investigate repetitive behaviours and both Goddard et al. (2014) and Weismuller et al. (2015) investigated physiological changes in boys, whereas Lind and Bower (2010) aimed to investigate subsets of memory. Most of the studies investigated VF as part of a wider assessment of neurological or executive functioning. Studies varied on the method of participant recruitment, from advertising on notice boards to using previous patients that had accessed treatment.

Methodological quality of included studies

Overall all papers were adequate or good in quality, scoring seven or above (out of 18) on the quality criteria and thus were included in this review (see Table 2). For the nine papers additionally rated by an independent practitioner, inter-rater reliability was 82%. Where discrepancies were identified, these were resolved through discussion and rechecking of papers. Ratings never differed more than one point between raters. An inter-rater reliability analysis using the kappa statistic was performed to determine consistency among raters, which was found to be high (kappa = 0.68, $p < .0.001$).

More consistently, papers failed to include information out about how they recruited their sample, who administered test instructions and how they handled indeterminate results, missing responses and outliers. However, all studies reviewed past diagnostic information or completed further ASD screening assessments to ensure the sample

they were testing was authentic. In nearly all cases the profession and qualification of this assessor was stated. It was therefore accepted that the available information was sufficient to determine that every study contained VF information for ASD. Most papers did not explicitly state whether data was excluded but stated an inclusion and exclusion criteria (excl. Bramham et al., 2009).

All papers used samples of participants with no reported additional diagnosis and a cognitive ability within the average range (using a cognitive assessment). The only exception to this was Mister et al. (2013), which used a sample with an IQ that was below the average range (although not impaired). On further investigation, the methodological and statistical quality of this paper was adequate, since although participants were not controlled for IQ, no significant difference between VF scores was observed. Despite the ASD group having significantly poorer cognitive skills, a potentially 'false significant' finding for VF was not found between groups.

Statistical analysis

All papers were included in this review (see Table 3). Studies varied in how they reported scores, with some reporting an overall mean, and others reporting scaled test scores. It is not possible to convert scaled scores back to raw scores without access to the data, which meant that it was not possible to complete a meta-analysis. This was mainly due to VF not being the primary focus of the included articles. However, an analysis to determine effect size was conducted using the mean and standard deviation scores to obtain a statistical effect size on the studies that reported a

significant and non-significant test result between HFA and TD individuals. This was established by performing a Cohen's *d* analysis (Cohen, 1988), which is used to indicate the standardised difference between two mean and standard deviation scores.

Semantic fluency

Of the fifteen studies that included a semantic measure, only five (33%: Dichter et al., 2009; Semrud-Clikeman et al., 2013; Spek et al., 2009; Verte et al., 2005; Weismuller et al., 2015) found a deficit for HFA (see Table 3). Spek et al. (2013) found semantic fluency tasks to be stimuli- (categories: animals vs professions) and group-dependent (HFA vs Asperger syndrome). For the five significant semantic VF findings (including Spek et al., 2013), the effect size was found to be between medium to large (d =-0.5 to -1.1, M= -0.72). For the eleven non-significant semantic VF findings (including Spek et al., 2013), the effect size was found to range between small and large (d =-0.1 to -0.9, M=-1.01).

Phonological fluency

Of the twelve studies that included a phonological measure, only five studies (42%: Papers Bramham et al., 2009; Kleinhans et al., 2005; Spek et al., 2009; Verte et al., 2005; Weismuller et al., 2015) found a deficit for HFA (see Table 3). Spek et al. (2013) found phonological fluency tasks to be stimuli- (letters: M vs L) and group-dependent (HFA vs Asperger syndrome). The effect size for significant findings was found to range between small to large (d =-0.3 to -1.0, M=-0.7). For the eight non-significant phonological VF findings (including Spek et al., 2013), the effect size was found to be between small to large (d =-0.009 to -0.7, M= -0.27).

Semantic switching fluency

Of the six studies that used a semantic switching measure, a majority of five (83% Papers Begeer et al., 2014; Corbett et al., 2009; Kleinhans et al., 2005; Semrud-Clikeman et al., 2013; Weismuller et al., 2015) found a deficit for HFA (see Table 3). The effect size for significant findings ranged between a small to large effect (d =-0.2 to -13, M=-0.93). For the one non-significant semantic VF finding the effect size was found to be medium (d =-0.7).

Reference	Clinical *N*	Diagnosis	*Male N*	Age (sd)	IQ (sd)	TD *N*	*Male N*	Age (sd)	IQ (sd)	Cognitive Measure
Anderson-Day (2014)	30	15 ASD, 15 AS*	28	14.7 (2.5), 12.9 (2.1)*	102.7 (13.7), 107.4 (14.5)*	15	10	14.05 (2.72)	110.4 (10.5)	WASI
Begeer et al. (2014)	26	26 ASD	23	13.8 (6.1)	109 (12.2)	26	22	11.8 (5.1)	109 (9.5)	PPVT
Bramham et al. (2009)	45	45 ASD	38	32.8 (12.5)	107.0 (16.4)	31	28	32.81 (9.02)	109.8 (16.8)	WAIS-III
Corbett et al. (2009)	18	18 ASD	17	9.4 (1.96)	94.2 (17.9)	18	12	9.56 (1.81)	112.2 (14.8)	WASI
Dichter et al. (2009)	39	39 ASD	38	9.7 (2.7)	101.7 (17.5)	39	38	10.57 (3.35)	111.7 (16.1)	Leiter-R
Goddard et al. (2014)	63	63 ASD	51	12.6 (2.8)	103.6 (13.1)	63	51	12.10 (2.26)	104.8 (11.8)	WASI
Kleinhans et al. (2005)	12	6 AS, 6 HFA	12	26.4 (7.7)	101.9 (10)	-	-	-	-	WASI
Lind & Bowler (2010)	14	14 HFA	11	41.4 (12.7)	105.9 (14.5)	14	11	43.83(10.39)	108.6 (18.2)	WAIS-III
Lopez et al. (2005)	17	17 ASD	14	29.1 (8.0)	77 (15)	17	11	29.4 (11.4)	89.0 (13.0)	WAIS-III
Maister et al. (2013)	14	14 ASD	14	12.2 (0.6)	109.5 (13.7)	14	13	12.1 (0.2)	120.6 (19.7)	BPVS
McKnight &Culotta (2012)	23	23 AS	0	12.5 (8.5)	106.0 (13.0)	50	0	12.4 (10.6)	106.0 (11.4)	WISC-IV
Robinson et al. (2009)	54	54 ASD	42	12.5 (2.9)	103.5 (10.5)	54	42	12.08 (2.34)	104.96 (28.0)	WASI-II
Semrud-Clikeman et al. (2013)	37	37 ASD	30	12.8 (2.6)	102.9 (16.0)	40	25	13.1 (2.6)	113.0 (10.9)	WASI
Spek et al. (2009)	62	31 HFA, 31 AS*	67	40.8 (10.9), 38.6 (11.8)*	114.8 (9.5), 111.8 (9.7)*	30	28	39.89 (11.45)	116.8 (11.3)	WAIS-III
Verté et al. (2005)	61	61 HFA	57	9.1 (1.9)	99.2 (17.1)	47	40	9.4 (1.6)	112.1 (9.7)	WISC-R
Weismuller et al. (2015)	15	15 ASD	15	9.4 (2.4)	99.3 (18.4)	12	12	10.6 (3.25)	118.3 (15.1)	WASI-IV

Table 1 Study demographics: Study Main author (date published), *N* number of (HFA) High Functioning Autism, (AS) Asperger Syndrome, (AD) Autism Spectrum Disorder and (TD) Typically Developed participants. The *N* number of males in the study, age in years and Total IQ Score (Standard deviation: sd). The cognitive measure used: (WASI) Wechsler Abbreviated Scale of Intelligence (Wechsler, 1999, 2003), (WISC) Wechsler Intelligence Scale for Children (Wechsler, 1992, 2003), (WAIS) Wechsler Adult Intelligence Scale (Wechsler, 1999, 2003), (BPVS) British Picture Vocabulary Scale (Dunn et al., 1987), Leiter-R (Roid & Miller, 1997), (PPVT) Peabody Picture Vocabulary Test (Dunn, 2007)

Author	1	2	3	4	5	6	7	8	9	Total
1.Anderson-Day (2014)	++	+	++	+	+	-	++	++	+	+
2.Begeer et al. (2014)	-	++	+	-	+	-	-	+	++	+
3.Bramham et al. (2009)	+	++	-	+	+	-	++	++	-	+
4.Corbett et al. (2009)	-	++	+	++	+	-	++	-	++	+
5.Dichter et al. (2009)	+	++	++	++	+	-	++	+	-	+
6.Goddard et al. (2014)	++	++	++	+	-	-	+	++	-	+
7.Kleinhans et al. (2005)	-	++	+	+	+	-	+	+	-	+
8.Lind & Bowler (2010)	-	++	+	+	+	-	+	+	+	+
9.Lopez et al. (2005)	-	++	+	-	+	+	+	+	-	+
10.Maister et al. (2013)	++	+	++	++	+	-	++	-	-	+
11.McKnight & Culotta (2012)	+	+	++	-	+	++	-	-	-	+
12.Robinson et al. (2009)	++	++	+	-	-	-	++	-	-	+
13.Semrud-Clikeman et al. (2013)	++	++	++	++	+	++	++	+	+	++
14.Spek et al. (2009)	-	+	++	+	+	-	+	++	-	+
15.Verté et al. (2005)	-	++	+	++	+	-	++	+	++	+
16.Weismuller et al. (2015)	+	+	+	++	+	-	-	+	-	+

Table 2. Quality Criteria: The overall agreed scores between the 1st author and co-reviewer. ++ well addressed (score of 2), + adequately addressed (score of 1), - poorly/ not addressed (score of 0). The total score ++ 12> (good quality), + 7-12 (adequate quality), - 0-6 (poor quality).

Reference	HFA *N*	TD *N:*	Measures Cognitive, VF	Description	ASD impaired SVF	*d*	*r*	ASD impaired PVF	*d*	*r*	ASD impaired Switch	*d*	*r*	Overall Quality Score
1.Anderson-Day (2014)	30	15	ACE-R	(S) Animals (P) P	N	-0.8,-0.7*	-0.4, -0.3*	N	-0.6,-0.7*	-0.3, 0.3*	-	-	-	-
2.Begeer et al. (2014)	26	26	Spreen & Strauss, 1991	(S) Animals	N	0.05	0.02	-	-	-	Y	-0.2	-0.1	17
3.Bramham et al. (2009)	45	31	Strauss et al, 2006	(P) F A S	-	-	-	Y	-0.8	-0.4	-	-		-
4.Corbett et al. (2009)	18	18	D-KEFS	standard	N	-1.6	-0.6	N	-0.5	-0.3	Y	-1.3	-0.5	21
5.Dichter et al. (2009)	39	39	Lezak, 1995	(S) Animals	Y	-0.8	-0.5	-	-	-	-	-	-	14
6.Goddard et al. (2014)	63	63	No Ref	(S) Animals, fruit/veg, clothes	N	0.01	0.004	-	-	-	-	-	-	16
7.Kleinhans et al. (2005)	12	-	D-KEFS	standard	N	0.1	0.03	Y	-0.7	-0.3	Y	-0.9	-0.4	16
8.Lind & Bowler (2010)	14	14	COWA, Turner, 1999	(S) Animals, Countries, food (P) F,A,S	N	-0.01	-0.005	N	0.3	0.2	-	-	-	14
9.Lopez et al. (2005)	17	17	D-KEFS	standard	N	-0.5	-0.2	N	-0.5	-0.2	N	-0.7	-0.3	16
10.Maister et al. (2013)	14	14	WAB, Benton, 1968	(S) Animals (P) B	N	-0.4	-0.2	N	-0.4	-0.2	-	-	-	21
11.McKnight &Culotta (2012)	23*	50	COWA, CFT	(S) Animals, Countries, food (P) K,M	N	0.5*	0.2*	N	-0.009*	-0.005*	-	-	-	9
12.Robinson et al. (2009)	54	54	No ref	(S) Animals, fruit/veg, clothes	N	-0.1	-0.1	-	-	-	-	-	-	17
13. Semrud-Clikeman et al. (2013)	37	40	D-KEFS	standard	Y	nr		N	nr	nr	Y	nr	nr	18
14. Spek et al. (2009)	62	30	GIT, Benton, 1968	S) Animals, Professions (P) K,M	Y N	-1.0,-0.5* -0.3*	-0.4, -0.3* -0.1*	Y N	-0.7, -0.3* -0.5, -0.3*	-0.3,-0.1* -0.2,-0.1*	- -	- -	- -	16
15.Verté et al. (2005)	61	47	COWA	(S) Animals, Food (P) K,M	Y	-0.9	-0.4	Y	-0.7	-0.3	-	-	-	21
16.Weismuller et al. (2015)	15	12	D-KEFS	standard	Y	-1.1	-0.5	Y	-1.0	-0.5	Y	-1.3	-0.6	11

Table 3. Findings: Study main author (data published), (N) number of (HFA) high functioning autism and (TD) typically developed participants. The measure/reference text for verbal fluency and description of the (S) semantic, (P) phonological tasks. (D-KEFS) Delis-Kaplan Executive Functioning System (Delis et al., 2001) s=boys names/animals, p=FAS, switch=fruit/furniture, (CFT) Category Fluency Task (Benton & Hamsher, 1989), (WAB) Western Aphasia Battery (Kertesz, 1982), (GIT) Groinger Intelligence Scale (Luteijn & Burelds, 2004), (COWAT) Controlled Word Association Task (Lezak, 1995), (ACE-R) Addenbrooke's cognitive Examination (Moioshi et al., 2006). Autism significantly impaired (Y) yes, (N) no, d = Cohen's effect size, r = Pearson's r. *Identified as sample of individuals with Asperger Syndrome, nr=no means (sd) reported in text.

Discussion

Findings from the current review

Sixteen studies reporting findings for VF with individuals with HFA without a comorbid condition or intellectual impairment were included in this review. Methodological and statistical information from included papers were extracted and subjected to a quality assessment. Overall quality was found to be 'adequate' or 'good' for each paper, and inter-rater reliability was high. VF had being significantly under-investigated previously in comparison to other executive skills in the literature, and had yielded inconsistent findings. Findings of this review indicated that a majority of semantic and phonological tasks with HFA yielded non-significant results, when compared to TD individuals. Although semantic switching deficits were more consistently reported for HFA, this is confounded by the demand it places on other cognitive skills (i.e. set shifting). Set shifting has more consistently yielded deficits for ASD samples (e.g. Courchesne et al., 1994).

Whilst it is possible to conclude from this review that there is little evidence for disfluency in HFA, it is important to address other considerations first.

The limitations of the papers included in the review

There are significant methodological variabilities in the included articles that require further discussion. Although in order to meet the specified inclusion criteria, all

studies clearly presented the method of statistical analysis and participant data, some studies did not complete a statistical analysis for semantic independent of phonological fluency or other task scores (e.g. design fluency, novel fluency tasks, episodic memory assessments).

Overall, studies tended not to include participants over the age of sixty or below the age of five, which may limit applicability of the findings of this review to older adults or young children. It was also the case that not all studies used an analysis that adjusted for participant's age, or assessed whether the matched sample differed in terms of sex, level of education or cognitive ability. The cognitive assessment method used also varied from more current, established measures, such as the Wechsler Intelligence Scale for Children (WISC-III, 1997) to older, less reported measures, such as the Leiter International Performance Scale (Leiter, 1980).

Areas of possible bias

Few papers reported when and where the study was completed. Some did not report how the participants were recruited/excluded, a rationale for choosing the measures used, and the training and sufficient expertise of the individuals carrying out the test instructions. Whilst much of the unreported information may have simply being left out of the published paper, under-reporting also limits conclusions from the data, since possible bias and measurement or methodological error is potentially hidden. Not all of the studies used populations considered representative of an ASD clinical population. Some studies used individuals that had being diagnosed by the authors,

that were inpatients or had being treated by a clinician previously – but not currently. Many studies did not fully report the results of statistical analyses.

Also, only published papers were included in the review. Whilst the aim of this review was to include papers of the highest quality, i.e. peer-reviewed studies, it is recognised that publication bias may mean that there is a bias towards finding a majority of studies with a significant effect – as non-significant studies are less likely to be submitted for publication. However, this was not the finding in this review as a majority of studies for all fluencies were not statistically significant.

Methodological variabilities that limit comparisons between studies

Studies differed by country of origin, entailing variations in translation of fluency task instructions, and potential culturally-dependent factors. In addition, studies varied in context, with some adopting more widely used categories and letters, such as 'F,A,S', animals and foods (Benton, 1968; Lind & Bowler, 2010) and others using less frequently used and more unusual categories and letters, such as, 'K, M' countries and vehicles (McKnight & Culotta, 2012) which has potential implications for bias. The standard text used also differed between studies, with some adopting standardised tests with given instructions (e.g. D-KEFS, Delis et al., 2001) while others did not, meaning that the procedure each included study used may have varied significantly and may have impacted on the results obtained. Studies also varied in how they reported scores, with some reporting an overall mean, and others reporting scaled test scores.

It was also not possible to complete a meta-analysis due to a mixture of raw and scaled scores reported in the papers selected. A further review may involve contacting authors for raw data. Although this may be considered a limitation of the review, the main aim of the review was to scope available literature and time restraints made it difficult to complete further assessment.

Due to a relatively small number of papers containing VF information for high-functioning individuals in the literature, it was also not possible to sub-divide participants based on other demographic information, such as age. It is recognised that this is a limitation of this review as it would be helpful to analyse child and adult data separately due to continuing brain development throughout adolescence. A repeat review may include only samples of adults or children in years to come, when there are more published studies containing VF information.

The quality of included papers

The quality assessment used for this review was adapted from other standard texts (e.g. STARD) to improve the relevance of the reporting criteria. However, this may also skew the quality assessment findings as it was designed by the first author and therefore has the potential to be biased in itself. On discussion and reflection with the co-rater it was recognised that some of the questions are too long and have multiple factors attached, which may negatively skew quality findings. These items needed more discussion in order to agree a quality rating. If a future review is conducted it

may be helpful to agree a quality rating tool with a co-reviewer before the selection of papers and pilot it accordingly.

In the quality review, most papers scored poorly on item six 'the qualifications of the person administering the test' and item nine 'how the missing data and outliers were handled'. Although these two items are important as both enhance the quality the data represent, it may be less common in practice that publications contain this information. It is possible that a future review could remove this from a quality rating and contact authors for this information as part of the exclusion criteria.

Other limitations of the review

Only quantitative responses for VF were extracted for analysis. Previous findings indicate that individuals with ASD may produce different qualitative responses to TD individuals, for example, Begeer et al. (2014) reported a clinical difference in the types of clusters (or categories) individuals use in the animal fluency task. It would also have been useful to compare category differences, as some studies report a significant difference between ASD and TD individuals on some variations of tasks (e.g. animals) and not others (e.g. professions: Spek et al., 2013). 'Animals' is the most commonly used exemplar category, and requires individuals to access animate information. Grandin (1995) and Burnett and Jellema (2013) have found that individuals with ASD have difficulty accessing animate concepts, but less difficulty forming and switching between inanimate concepts.

Summary

Previous studies testing VF in ASD populations have yielded inconsistent findings. The aim of this review was to investigate available literature with the performance of non-learning disabled HFA populations, without a comorbid condition on VF tasks. A further aim was to compare findings of phonological, semantic and semantic switching VF. A literature search was conducted which extracted 16 studies meeting the specified inclusion criteria, which found very few studies reporting a significant difference between typical and clinical populations on phonological and semantic tasks. Only a very small number of studies reporting semantic switching were found; although most of these were found to be significant this may be due to the high demand of other executive skills, such as set shifting.

In summary, it is concluded that there is insufficient evidence to support the hypothesis that disfluency can be attributed to autistic symptomology. However, the current methodological variabilities in reported articles requires further investigation.

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Disclosure statement

No potential conflicts of interest were disclosed.

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Empirical Paper

Title:

Do high-functioning adults with autism display executive functioning deficits on a neuropsychological battery of assessments?

Running Title:

Executive Dysfunction in HFA

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Hollie Burnett designed and conducted the study under the academic supervision of Dr Jill Cossar and Dr Suzanne O'Rourke, who assisted with the design and research question and provided comments on the final manuscript.

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Keywords: autism, high-functioning, Asperger syndrome, executive functioning, Delis-Kaplan Executive Functioning System, D-KEFS, neuropsychological profile, set shifting.

Abstract

Aim: To compare the executive functioning profile of individuals with high-functioning autism (HFA) and typically developed (TD) individuals. *Method:* 22 HFA and 22 TD participants (mean age = 28, range = 17-73, 52% male) without a comorbid condition, learning disability or brain injury completed three subtests from the WAIS-IV (vocabulary, block design and digit span) and all subtests of the Delis–Kaplan Executive Functioning System (D-KEFS). *Results:* The HFA group performed significantly poorer on the switching condition of the design fluency task, semantic conditions of the verbal fluency task and on the word context task overall.

No other significant differences were observed. *Summary:* The HFA group performed poorer than the TD group on subtests designed to measure generating novel 'imaginative' ideas, without visual cues to aid performance. The deficit on these subtests was increased when there was the added condition requiring the participant to switch between newly-formed concepts. The results suggest that even in a group of high-functioning individuals there are still measurable differences in executive functioning (EF) between TD and HFA samples that may not be apparent through more general cognitive testing. Implications for using a neuropsychological profile for adults with HFA are discussed.

Introduction

Executive functioning (EF) refers to higher-level cognitive skills such as anticipation, goal selection, planning, fluency, inhibition, attention, problem-solving and monitoring (e.g. Stuss & Benson, 1986; Russell, Jarrold & Hood, 1999; Hill, 2004; Pellicano, 2006). These mechanisms allow a typical person to shift attention flexibly, inhibit proponent responses, generate verbal responses and goal-directed behaviour, and solve problems in a planned, strategic way (Baron-Cohen & Swettenham, 1997). Animal and human neuro-imaging studies have found that some components of EF can be localised to specific areas of the brain, such as the pre-frontal cortex and the cerebellum (e.g. D'Esposito et al., 1995; Tulving et al., 1996; Pellis & Pellis, 2006) and damage to these areas has rendered individuals impaired in specific tasks designed to measure EF (e.g. Shallice, 1982).

In order to inform clinical practice, EF tasks have being used to demonstrate patterns of dysfunction in acquired brain injury (Echemendia et al., 2001), neurodegenerative conditions such as Alzheimer's disease (Welsch et al., 1991), Huntington's disease (Beglinger et al., 2010), and Parkinson's disease (Jacobs et al., 1995). Patterns of executive dysfunction have also being found in other conditions, such as schizophrenia (Saykin et al., 1994), attention deficit hyperactivity disorder (Wodka et al., 2008), and individuals who have had a stroke (Sashdev et al., 2004).

Some studies show that autism spectrum disorder (ASD) and cerebellum damaged individuals show similar deficits in the ability to plan and initiate activities and switch attention (Luria, 1980; Duncan, 1986; Shallice, 1982; Courchesne et al., 1994). Ozonoff at al. (1991) name these executive difficulties as the 'theory of executive dysfunction' and uses this to explain the social, communication and flexibility of thought difficulties experienced by individuals with ASD. Although findings are mixed, some research indicates that individuals with ASD are impaired in tasks designed to measure EF, such as the Wisconsin card sorting task (WCST: Ozonoff et al., 1991), in which the individual has to form/switch concepts; the tower of Hanoi/London (e.g. Shallice, 1982), where the individual has to solve problems by planning before acting (e.g. Hughes, Russell & Robbins, 1994); and the verbal fluency task (e.g. Verte et al., 2005) where the subject has to generate novel examples of words beginning with a given letter or category, in a fixed time period. However, using similar tasks, other studies have found no such difference (e.g. Minshew et al., 1992; Robinson et al., 2009). Some research has found cognitively intact individuals with HFA to perform equally to TD individuals on psychometric

tests, but exhibit impaired executive processes in real-life situations (i.e. Channon et al., 2001; Gnanathusharan et al., 2011).

Some studies have modified EF tasks for individuals with ASD so that they do not contain as many demanding social characteristics (e.g. Russell et al., 1999). Ozonoff (1995) and Tsuchiya et al. (2004) have found that individuals with ASD performed better, for example, using a computerised version of the WCST, which involves no social or verbal demands. Others suggest that broader cognitive skills, such as working memory (Lehto, 2003), vocabulary and perceptual reasoning (Salthouse, 2005) are closely linked to EF, which suggests that higher-functioning individuals with ASD may not show the same pattern of dysfunction on EF tests as their less able counterparts.

When measuring EF in people with ASD, many studies include data with confounding participant variables, such as very young or advanced age, very low cognitive ability or other comorbid condition(s) such as other developmental disorders, dyslexia, schizophrenia or Tourette syndrome (e.g. Kover & Abbeduto, 2010; Verté et al., 2005). It is therefore possible that task understanding, underdeveloped frontal lobes, or a comorbid diagnosis may falsely associate executive dysfunction with autistic symptomology. Also, for most studies, two to three EF tests are typically chosen as a measure of EF (e.g. verbal fluency, card sorting and trail making). This makes it difficult to compare within sample performance and establish specific strengths and weaknesses for an ASD population in EF. It is not currently known how individuals with ASD perform on a battery of

assessments measuring a wider variety of EF skills. It would be useful to establish whether an EF battery gives a clear pattern of dysfunction for ASD, as it is unclear from the literature. A clear profile for ASD may in the future aid clinical decision-making for ASD diagnosis, management and rehabilitation or support goals.

In summary, there is a lack of clear evidence that individuals with high-functioning ASD, without a comorbid condition (i.e. learning disability, traumatic brain injury, neurological disorder) are impaired in tasks designed to measure EF. It is possible that a lack of homogeneity in testing samples and lack of consistency in EF measures used contributes to the apparent absence of clear evidence for executive dysfunction in ASD. There is a need for more reliable ASD screening tools in clinical assessment to distinguish ASD from other conditions, particularly in high-functioning individuals, who are able to develop coping strategies to mask presenting difficulties.

The aim, therefore, of this study is to compare the neuropsychological profile of executive functioning in adults with high-functioning autism (HFA), without a comorbid condition, learning disability or brain injury, to TD adults.

Method

Participants

A total of 22 adults with HFA (13 males, 9 females) and 22 TD adults (11 males, 11 females) participated in the study by responding to e-mails or flyers distributed on

behalf of the examiner by autism support (Scottish Autism) and education services (four universities) across the UK (see Table 1). It was requested that universities contact all students with a diagnosis of ASD, HFA and Asperger syndrome. Scottish Autism contacted all clients that were known to have studied at higher level (A-level) or above. This was to exclude the possibility of participants volunteering who potentially had a learning disability. The examiner had no prior knowledge of participants or their details. The participant could simply reply to the researcher via the information circulated to them if they were interested in participating in the study. Twenty-two participants was estimated sufficient for the number of variables measured in this study, determined by an *a priori* power analysis (Gpower = 0.95: Faul & Erfelder, 1992).

The HFA participants had a diagnosis of ASD, high-functioning autism or Asperger syndrome previously made by a clinical psychologist, psychiatrist or multi-disciplinary team. For the purposes of this paper, all participants will collectively be referred to as having 'high-functioning autism' (HFA). HFA is not a term used by the DSM-IV or ICD-10, but as it was not possible to take a detailed developmental history, including early language delay, it is difficult to use the DSM-V criteria of 'Asperger syndrome'. Asperger syndrome is a controversially applied diagnostic term and has since been removed from the DSM-V (2013) criteria. Some argue that it is unhelpful to have two separate diagnostic terms and ASD should be used as an umbrella term to describe all individuals with the same areas of presenting difficulties. 'High-functioning' is a term used in this paper for the purposes of

describing higher cognitively able individuals with ASD, without a learning disability.

All participants' intellectual ability was within the average range or above, using three subtests from the WAIS-IV-UK (Wechsler, 2010, see Table 1 and below for details) and did not have a mental or neurological illness or previous brain injury. Participants took part in the research voluntarily and were given feedback at the end about their relative strengths and weaknesses. All TD and HFA participants were asked if they could see the materials, spoke English as their first language, and provided written consent prior to the experiment. Data collection was made by a specialist psychological practitioner who was trained to administer and interpret psychometric tests. The study was approved by the ethics committee of the host academic institution.

	N	Sex	Age	AQ	IQ-T	IQ-V	IQ-P	IQ-WM
TD	22	11M, 11F	24.0 (7.8; 17-55)	16.2 (5.8)	12.2 (2.4; 8.7-16.7) 110.8 (12.0)	11.7 (2.5; 6-17) 108.4 (12.3)	12.4 (2.8; 6-19) 112.0 (13.9)	12.4 (3.6; 7-19) 111.8 (18.2)
HFA	22	12M, 9F	32.0 (15.9; 17-73)	34.1 (5.9)	12.9 (3.1; 7.3-18.0) 114.6 (15.6)	14.0 (3.5; 8-19) 120.0 (17.5)	12.3 (4.0; 7-17) 111.6 (19.8)	12.4 (3.8; 6-19) 112.1 (19.3

Table 1: HFA: High Functioning Autism; TD: Typically developed; N: Number of participants; AQ: Autism Spectrum Quotient; M: Male; F: Female; Mean (standard deviation) for subscale and converted index scores: IQ-T: Total Mean Intelligence Score, IQ-P: Performance Mean Intelligence Score; IQ-V: Verbal Mean Intelligence Score, IQ-WM: Working Memory Intelligence Score.

Measures and procedure

The Wechsler Adult Intelligence Scale – Fourth Edition – UK Version (WAIS-IV-UK, Wechsler, 2010) is a measure of cognitive functioning validated in a typically developed sample. The WAIS-IV (Wechsler, 2008) has previously been used with ASD samples, with some studies finding deficits in working memory and processing speed (e.g. Bucaille et al., 2016) and others in verbal comprehension (Holdnack et al., 2011). All participants completed three subtests of the WAIS-IV-UK (the vocabulary, block design and digit span tests) in order to give an estimate of verbal, perceptual reasoning and working memory ability, a method often employed to restrict participant fatigue (e.g. Best et al., 2008; Wechsler Abbreviated Scale of Intelligence: WASI, Wechsler, 1999). The subtests were chosen in line with previous studies and WAIS-IV abbreviations (e.g. WASI). The purpose was primarily as a control for EF, as successful performance on EF tests would require some combination of broader cognitive skills, such as language, memory and attention, and also to ensure that the groups did not differ significantly. One participant with HFA was excluded from the analysis due to having an IQ-working memory subscale score that was below the average range.

The Autism Quotient (AQ, Baron-Cohen et al., 2001), is a fifty-statement, self-administered questionnaire designed to measure the degree to which an adult with normal intelligence has traits associated with ASD. Although the AQ is not a diagnostic tool, a score of 32 or higher (out of 50) has been shown to correlate with ASD (Baron-Cohen et al., 2001; Wheelwright et al., 2006; Hoekstra et al., 2008).

Others suggest that a more conservative threshold score of 26 would ensure that false negatives are limited (Woodbury-Smith et al., 2005). The AQ was used mainly as a screening tool to ensure that TD participants did not possess a high number of autistic-like traits (32+) that met the HFA threshold, which may confound any possible findings. The AQ was also used as an exclusion criterion for the HFA sample, if they reported a low number of autistic-like traits (lower than 26). No participants were excluded based on the AQ, as no TD participants scored above 32 (range: 5-27). Only one HFA participant (range: 22-47) scored below the conservative cut off of 26 (scoring 22), however, this individual had brought along written reports detailing their diagnosis, including the Autism Diagnostic Observation Schedule (ADOS, Lord et al., 1999), and was thus included in the study.

The Delis-Kaplan Executive Functioning System (D-KEFS, Delis, Kaplan & Kramer, 2001) The D-KEFS (Delis et al., 2001) was chosen for the purposes of the current study. It comprises more subtests (nine) than most batteries of EF and measures a wider variety of EF skills (see below). It is standardised on typically developed (TD) individuals from 8-89 years. Each task of the D-KEFS undoubtedly requires several higher level cognitive abilities, but tasks are designed to predominantly measure key components of EF more specifically and convert them into scaled scores, which are easily comparable to other cognitive assessments (e.g. WAIS-IV-UK, Wechsler, 2010).

Although no published studies give an overall profile for ASD on the D-KEFS, lower scaled scores have being found on subtests designed to measure verbal fluency,

visual scanning, flexibility of thought, planning and inhibition, with significant deficits found only on the switching and inhibition conditions (Kleinhans, Akshoomoff & Delis, 2005; Lopez et al., 2005). Other studies have found no significant difference between inhibition and verbal fluency (e.g. Corbett et al., 2009). In some studies, authors have attempted to use specific subtests of the D-KEFS to compare other skills or deficits, such as repetitive behaviours, with mixed findings (e.g. Lopez et al., 2005; Kenworthy et al., 2009). Ridley, Homewood and Walters (2011) found a non-clinical sample of adults to be significantly poorer at motor function and verbal set shifting ability with higher degrees of autistic-like traits, using the autism quotient (AQ).

All nine subtests were administered for the purpose of this study and twelve of the main achievement scores were analysed. These include:

-The *trail making test* measures flexibility of thinking on a visual-motor sequencing task. It comprises five conditions, involving a visual cancellation task, a series of 'connect the circle' tasks and a sequence switching condition, where a participant has to alternate between connecting letters and numbers in numerical and alphabetical order.

-The *verbal fluency test* measures letter fluency (generating words that begin with a specific letter, e.g. 'F, A, S'), category fluency (generating words within a specific category e.g. animals) and category switching (generating as many words by

alternating between two categories, e.g. furniture and fruits). There is a 60 second time limit for each task.

-The *design fluency test* measures initiation of problem-solving behaviour, fluency in generating visual patterns, creativity, drawing the designs whilst keeping to the rules and restrictions of the task, and inhibiting previously drawn designs. There are three conditions, all requiring the participant to draw designs using four lines to connect five dots. The second condition involves ignoring competing black-dotted stimuli, and the third task involves alternating between black and empty dots in a sequence.

-The *colour-word interference test* measures the ability to inhibit a dominant and automatic verbal response. It is based on the original Stroop task (Stroop, 1935). There are two baseline conditions involving identifying colours and reading words across a page. The next two conditions involve stating the colour of the ink in which the word is printed, not what the word actually reads. The final condition involves multiple rule switching.

-The *sorting test* measures concept-formation skills, modality-specific problem-solving skills, and the ability to explain sorting concepts abstractly. The sorting test is based on the Wisconsin card sorting test (WCST, Berg, 1948). The participant is given two card sets, comprising six cards which they must sort into two groups of three. The participant must then describe what each group has in common.

-The *twenty questions test* measures the ability to categorise, formulate abstract questions, and incorporate the examiner's feedback to give more efficient questions. The participant is shown 30 pictures on a page and must guess in as few questions as possible which image the examiner has chosen. They may ask any question they like, so long as the examiner can answer only yes or no. How the participant initially abstracts the information, the number of questions and an achievement score is recorded.

-The *word context test* measures verbal modality, deductive reasoning, integration of information, hypothesis testing and flexibility of thinking. For each item the participant attempts to discover the meaning of a made-up word, using clue sentences. They must adjust their response if the word no longer fits with the next clue sentence.

-The *tower test* measures spatial planning, rule learning and inhibition of impulsivity. The task is to move the discs of various size across three pegs to build a designated tower in the fewest possible moves. The participant may only move one piece at a time and cannot put a larger piece on top of a smaller piece.

-The *proverb test* measures the ability to form novel, verbal abstractions. Eight sayings (four common, four more unusual) are read to the participant and they are required to explain the meaning. The participant is then given a multiple choice where they can identify which sentence best explains the proverb.

Results

Statistical design

A 2×12 Analysis of covariance (ANCOVA) was performed, in which the between-participants factor was GROUP (HFA vs TD) and the within-subjects factor was TASK (12 primary subscale scores from the D-KEFS, see Figure 1). An analysis was not completed on every primary score as the purpose of some subtests was only to act as a baseline or practice condition for the tasks included in the analysis, to assess language or reading ability. The covariates included were the IQ, age and sex. Age and sex were considered important factors as underdeveloped/advanced brain development related to age or specific strengths (e.g. verbal reasoning) may be related to gender differences that may impact on performance on neuropsychometric testing.

Results for EF assessment

The analysis showed a statistically significant main effect for Group ($F(12,26) = 3.58$, $p < .01$, $\eta^2 = .602$), reflecting that the HFA sample performed poorer (M = 11.6, SD = 3.1) than the TD sample (M = 12.5, SD = 2.7) overall on the 12 sub-scores

analysed on the D-KEFS. However, of the included subscales, there was no significant difference between groups on 6/9 tasks: the trail making, inhibition, card sorting, twenty questions, tower and proverb tasks (all *P*s > 0.5).For only the following three tasks was a significant difference found:

Verbal fluency task: There was no significant difference for phonological fluency (*p*> 0.10). Semantic fluency (*p*< .05) was significant, reflecting that the number of generated category words was higher for TD (M=15.8, SD=3.4) than HFA individuals (M=14.3, SD=3.8), and switching between categories was highly significant (*p*< .001), again reflecting that the TD group generated significantly more words (M=14.9, SD=3.4) than the HFA group (M=12.0, SD=3.8).

Design fluency task: The design fluency switching condition was highly significant (*p*< .001), reflecting that the number of generated designs was higher for TD (M=14.41, SD=2.98) than HFA individuals (M=11.13, SD=3.41).

Word context test: There was a significant difference between groups in word identification (*p*< .05), reflecting that the TD group were able to identify the correct word with fewer clue sentences (M=11.95, SD=1.7) than the HFA group (M=10.68, SD=2.99).

Post-hoc analysis: As semantic fluency conditions were found to be statistically significant, an additional analysis was conducted to identify if the HFA group made a similar number of accurate switches between categories. The HFA group (M = 11.7,

SD = 3.1) were found to make significantly fewer correct switches than the TD group (M = 15.0, SD = 2.9) ($p < .001$). An additional analysis was also conducted to examine whether baseline conditions of the design fluency task were also significant, although the HFA group performed poorer on both conditions, no significant finding was made $p > .05$.

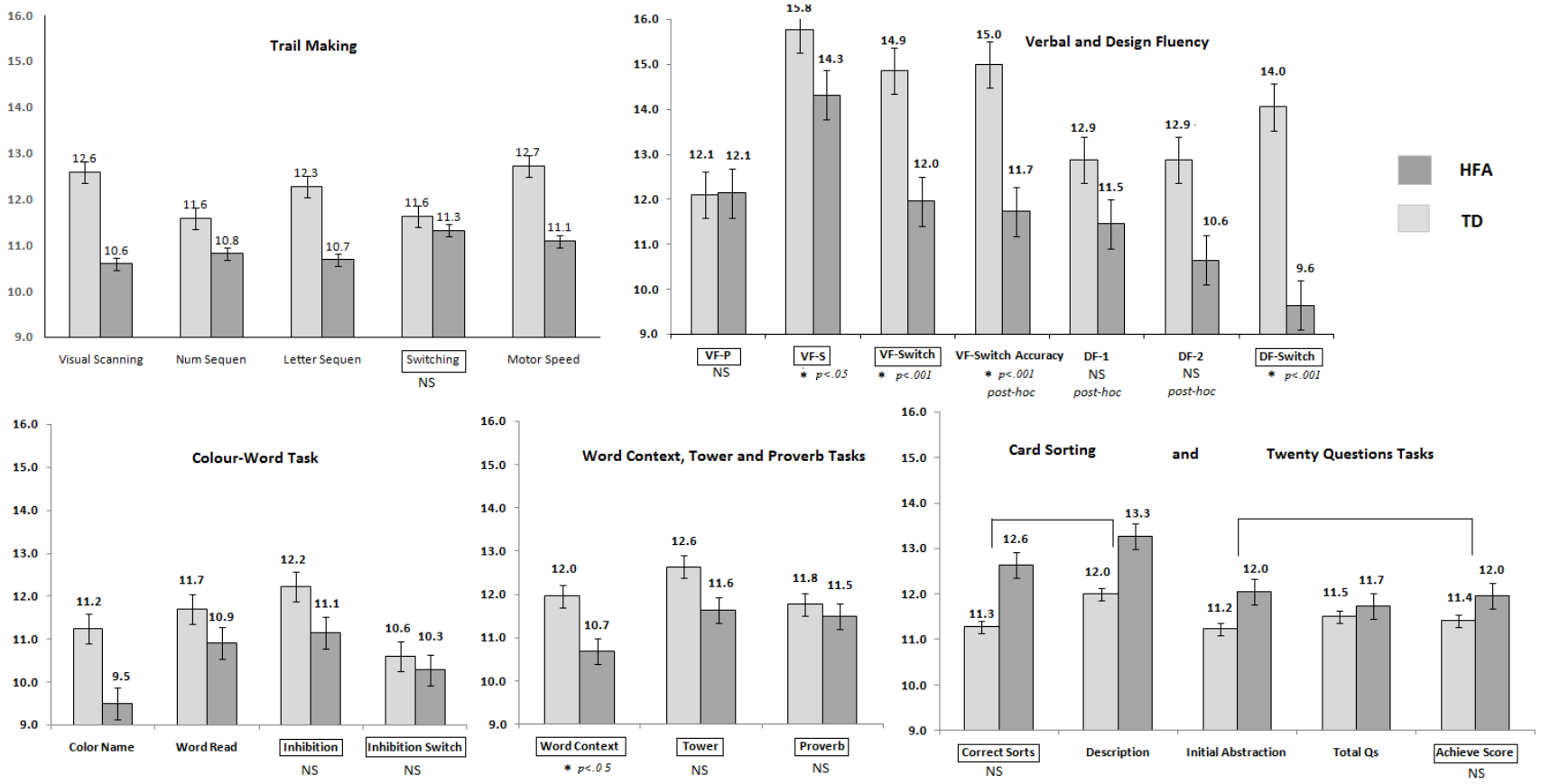

Figure 1. All subtest mean scaled scores (standard error) for high-functioning autism (HFA) and typically developed (TD) participants on the Delis-Kaplan Executive Functioning System. A square box around the subtest name indicates that test was included in the overall analysis, post-hoc indicates the test was included in the post-hoc analysis. If a significant difference was found, the value is indicated. No significant difference (NS).

Covariate findings

IQ (the three combined subtest scaled scores from the WAIS-IV-UK,Wechsler, 2010) was found to be significant when compared to the subtests of the D-KEFS. However, IQ did not differ significantly (*P*> 0.10) between the HFA (M = 12.9, SD = 3.1) or TD group (M = 12.2, SD = 2.4), nor did an analysis of the block design and digit span subtests separately (*Ps*> 0.05). There was a significant difference between the two groups for vocabulary (t (42) = -2.54, p<.05), with the HFA group displaying better knowledge of word meaning (M = 14.0, SD = 3.5) than TD individuals (M = 11.7, SD = 2.5, p=0.015). As there were multiple comparisons, a Bonferroni correction of .021 was used to reduce the possibility of producing false positives.

Age was significant between groups (F (12, 26) = 2.56, *p*< .05, η^2 = .996) with the TD individuals (M=24.0, SD=7.8) being significantly younger than HFA individuals (M=32.0, SD=15.9). However, on all subtests where a significant effect was found, age was not significant (*Ps*> 0.10).

Similarly, gender was not significant on all subtests of the D-KEFS where a significant finding was indicated (*p* > .05).

Discussion

The aim of this study was to compare the neuropsychological profile of EF for TD and HFA adults without a comorbid condition, learning disability or brain injury. All

nine subtests of the D-KEFS (Delis et al., 2001) were completed and twelve primary subscale scores were analysed. The results indicated that the HFA group performed poorer on the switching condition of the design fluency task, the semantic conditions of the verbal fluency task, and overall on the word context task. No other significant differences were found. The lowest mean subscale scores for HFA on the D-KEFS was still within the average range, which only suggests this being a relative deficit for this HFA sample. Possible explanations of why HFA participants may have found the three tasks more demanding than the TD sample are further discussed.

The verbal fluency test measures letter fluency (phonological), category fluency (semantic) and category switching fluency (semantic switching). No significant difference was found for phonological fluency, but semantic and semantic switching fluency significantly differed between groups, with the HFA group generating fewer words than TD individuals. When the HFA group made switches between categories, they were also significantly less accurate than the TD group. It is not uncommon for one area of verbal fluency to be found intact and another impaired (Henry & Crawford, 2004), as it is argued that phonological and semantic fluencies may have different mechanisms for retrieving verbal knowledge, and are possibly mediated by different regions of the prefrontal cortex (Szatkowska et al., 2000). In a systematic review of verbal fluency in HFA, Burnett et al. (2015) found very few studies able to corroborate a deficit in generating letters or categories. More consistently a deficit was found in a very small number of studies investigating switching between semantic categories.

Although overall IQ (three subtests of WAIS-IV-UK) was found to significantly correlate with performance on the D-KEFS, there was no significant difference between the HFA and TD groups, and means were adjusted using a Bonferroni corrected design. Furthermore, the vocabulary subtest of the WAIS-IV-UK was significantly stronger for the HFA than the TD group, reflecting that underperformance on the two sub-sets of verbal fluency was not due to unfamiliarity with general words. It is possible that, similar to concept formation, semantic verbal fluency involves the ability to put words into categories by grouping targets according to meaningful and conceptual features, which has being shown to be a deficit in ASD (Temple Grandin, 1995; Burnett & Jellema, 2013). Hence, the retrieval of 'abstract meaning' rather than 'sound' or 'definition' may explain the deficit found for semantic and semantic switching fluency in HFA.

The design fluency task measures initiation of problem-solving behaviour, fluency in generating visual patterns, creativity, producing designs whilst keeping to the rules and restrictions of the task, and inhibiting previously drawn designs. The HFA individuals generated significantly fewer designs on the switching condition, but a further analysis found no significant difference on the two baseline conditions, although the results approached significance. It is therefore likely that all conditions of the design fluency task were demanding for the HFA group, but the added complexity of set shifting increased the deficit to be clinically significant.

Similar to the verbal fluency task, for the design fluency task, the participant was asked to generate ideas from scratch. Although the participant was not required to

abstract meaning, they had to keep to three different rules in order to create designs: draw four straight lines connecting dots, alternating between black and white dots, and remembering not to repeat previously drawn designs. It could be argued that this task places a heavy demand on working memory, which has being found in some studies to be a difficulty in individuals with ASD (e.g. Barendse et al., 2013). However, no significant difference was found between HFA and TD for working memory on the WAIS-IV-UK or on other subtests on the D-KEFS that are argued to be demanding on working memory, for example the tower task (Ozonoff & Strayer, 2001).

The word context test measures verbal modality, deductive reasoning, integration of information, hypothesis testing, and flexibility of thinking. The HFA individuals were delayed or produced fewer correct responses to each clue sentences. All participants were able to generate an initial response, but it was more difficult for participants with HFA to adjust their given response if the word no longer fitted with the next clue sentence. Similar to the word context task, individuals with ASD have been found to be poorer at other tasks designed to measure response selection and monitoring (Happe et al., 2006). Again, difficulties in this subtest cannot be attributed to poor vocabulary or set shifting difficulties, as performance on other tasks (i.e. vocabulary, trail making) does not support this. However, similar to the fluency tasks, the participant is asked to generate concepts from scratch, without visual cues to aid performance, for example, visual features on cards, letters and numbers to switch between or a multiple choice option. Concept formation and flexibility of thinking have been shown to be a deficit in ASD individuals (e.g.

Ozonoff, 1995; Burnett & Jellema, 2013). Individuals with ASD are often described as 'rigid' and find it difficult to switch and change between ideas once formed (Grandin, 1995), which may account for the difficulties found for those with HFA on the three D-KEFS tasks.

Initially the results may seem surprising, as understanding tasks measuring complex abstract sayings (i.e. proverb task) and forming and switching between ideas (i.e. card sorting and twenty questions task), which are widely reported to be difficulties experienced by individuals with ASD were not found in this study. However, individuals with HFA did underperform when compared to TD individuals on most conditions, although this did not reach statistical significance. A conservative Bonferroni corrected *p* value of .021 was enforced to ensure reliability of test findings as there were multiple dependent factors. Although this adds to the reliability of the significant findings, it is also possible that this decreased the chance of finding significant group differences on other tasks. Also, to limit the number of dependent variables, the baseline and confounding subtest scores were excluded from the overall analysis, which limits how the findings may be used to generate a profile for EF in HFA individuals. It is also possible that on some subtests the difference in performance may be accounted for by the TD group overperforming or by the sampling method employed.

It is possible that the participants that volunteered in the study did not experience significant difficulties in any further areas of EF and were therefore more likely to take part. Although this is possible, it is unlikely as deficits were found within the

HFA sample that related to forming novel ideas and switching attention, and individuals with HFA also indicated difficulties relating to flexibility of thinking on the AQ.

It is also possible that the D-KEFS or other EF assessments are not sensitive enough to measure deficits in HFA. It has been argued that many of the current conventional EF tests used by clinicians and neuropsychologists tend to be crude and underspecified in terms of the cognitive processes that they engage and are not sensitive enough to detect executive dysfunction in different clinical groups (Burgess, 1998). As mentioned, some research has found cognitively intact individuals with HFA to perform equally to TD individuals on psychometric tests, but be impaired in executive processes in real-life situations (i.e. Channon et al., 2001; Gnanathusharan et al., 2011). An example of this is the virtual errands task, where a participant is asked to plan what they need when they go shopping.

It is important to note that although cognitive testing is useful to profile strengths and weaknesses, the skills do not always translate into practical, real-life situations. It is argued that neuropsychological assessments should be designed with ecological validity in mind (the verisimilitude approach – as close to everyday life tasks as possible), as opposed to veridically comparing traditional test scores as a more effective way of predicting functioning (Cheytor & Schmitter, 2003; Spooner & Panchana, 2006). Some progress has been made on EF measures, such as the 'test of everyday attention' (Robertson et al., 1994); however, it is possible that the skills measured by the assessments in this study (i.e. D-KEFS, WAIS-IV) do not tap into

the deficits experienced by those with HFA in everyday life, nor do they contain the same social demands.

The lack of significant findings could also reflect the fact that many higher level cognitive skills are in fact intact in individuals with HFA. Many previous studies that have found executive dysfunction in autism have included populations with confounding variables (e.g. very young or advanced age, neuropsychological insult, comorbid condition, or low intellect) which may have falsified a deficit for EF (e.g. Begeer et al., 2013; Kover & Abbeduto, 2010). Also, many previous tasks do not use individuals with average to above average IQ who may also be better at compensating for the social demands in EF tasks that have hampered the performance of lower functioning individuals (e.g. Russell et al., 1999; Ozonoff, 1995; Tsuchiya et al., 2004).

Similar to Kleinhans, Akshoomoff and Delis (2001), lower subscale scores on the D-KEFS were found on most subtests, with few significant findings. Previous studies of executive functioning in ASD differ significantly with some studies reporting a profound deficit (e.g. Weismuller et al., 2015) and others no significant difficulties (e.g. Corbett et al., 2009; Lopez et al., 2005). In the present study, individual subtest differed, with no significant difference found on some tasks (e.g. inhibition task), and other tasks significantly impaired (e.g. semantic verbal fluency test).

The current study must be viewed in context of some other limitations. The D-KEFS (Delis et al., 2001) was the assessment tool chosen for this study due to the variety of

executive skills each subtest tapped into. However, it is not known how samples of HFA perform on any other battery of executive functioning and no comparisons were made. The D-KEFS is also a lengthy assessment tool, taking approximately two and a half hours to administer, which limits the ability to generalise the findings of this study.

A general limitation of the study is that other cognitive skills such as language ability or reading level were not addressed. It is possible that these skills may have provided a more coherent baseline score with which to compare some of the functions measured by the D-KEFS, for example, verbal fluency. However, as the WAIS batteries are the most widely used cognitive assessments available and participant test fatigue was considered a significant factor other cognitive measures were excluded from testing. Also, many of the D-KEFS baseline measures (for example, parts 1-3, 5 of the trail making, inhibition parts 1-2) provide a baseline condition for the cognitive skill measured (e.g. visual scanning, reading speed).

In summary, the present study provides insight into the neuropsychological profile of executive functioning in high-functioning adults with autism on a battery of assessments. Although the HFA group performed poorer that the TD group on most subtests, this only reached statistical significance for three tasks; the design fluency-switching condition, the verbal fluency-semantic conditions and the word context task. It was recognised that each of these tasks required the generation of concepts from scratch, flexibility of thinking and imagination. Unlike many other conditions, the three tests did not provide visual cues to aid performance, for example, visual

features on cards, letters and numbers to switch between, or a multiple choice option. However, the lowest mean subscale scores for HFA on the D-KEFS were still within the average range, which only provides a relative deficit for HFA. This highlights the importance of establishing cognitive baseline scores and a full battery of assessments with high-functioning individuals in order to establish relative strengths and weaknesses of performance. Measures of set shifting, imagination and generativity may help to distinguish HFA from TD individuals; however, neuropsychological assessments require further investigation in order to establish reliability and validity in HFA samples. Further research is also required to investigate how testing EF skills empirically can translate into real-life situations.

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Appendix A1. Systematic Review: Quality assessment tool

Main Author Name/Date:

_____

A) PARTICPANTS / RECRUITMENT	**Well Addressed**	**Adequately Addressed**	**Poorly/Not Addressed**
1) The selection criteria. How the ASD sample were selected/recruited.	Good representative sample of ASD individuals in everyday life. Not known to researcher prior to task.	Adequate sample but less representative, e.g. through adult diagnostic service.	Not reported or not representative e.g. Sample of inpatient ASD. Known to researcher, e.g. gave participant diagnosis.
2) Diagnostic information of ASD participants. How they received diagnosis of ASD. Demographics available.	ASD screening assessment completed. Demographics evident.	Previous ASD diagnostic information reviewed. Demographics available.	Information not available, not clear.
3) The Inclusion/exclusion criteria. Who was included in the study.	States inclusion criteria or if there were participants excluded and why.	Inclusion criteria specified. But information about excluded participants, how many excluded, not fully available or transparent why.	Information not available, not apparent. Did not explain excluded participants.
4) Any confounding participant variables e.g. if they take	States how many participants have additional	Gives rough idea of comorbid conditions or what types of medication	Information not available, not apparent. Many co-

medication, or have comorbid condition.	diagnosis, what that is. If participants take medication and what medication.	participants take but not explicit.	variables that will interfere with test validity.
TOTAL SECTION A.	**/8**		
B) MEASURES	**Well Addressed**	**Adequately Addressed**	**Poorly/Not Addressed**
5) The executive functioning or VF assessment used. If this is referenced what was the validity of using the assessment with the ASD group.	Good Psychometric properties, validated on sample used. Fully referenced.	Tool chosen referenced but not identified as validated with ASD sample.	New measure, or measure not referenced. Or unclear as to what assessment was used.
6) The qualification of the persons executing and reading the test instructions.	Qualified staff, e.g. DClinPsy. Or staff with sufficient training and expertise to use assessment tools.	PhD orDClinPsy students or reasonable expectation that qualification or training would be sufficient to administer test, without fully acknowledging so.	Undergraduates, not stated or those without sufficient expertise to administer and interpret assessments. Or training or qualifications not explained.
TOTAL SECTION B.	**/4**		

C) POWER AND ANALYSIS	**Well Addressed**	**Adequately Addressed**	**Poorly/Not Addressed**
7) Clinical and demographic characteristics of the study population. Any differences accounted for in statistical analysis	Analysis conducted on covariates. If different, accounted for in statistical analysis.	Analysis conducted on covariates or states not significant, but not as clear or fully accounted for in overall analysis	Not explained or accounted for. Missing or there is a significant covariate but this is not accounted for or adjusted for analysis.
8) Overall data. Power analysis completed, *p* value adjusted to account for number of means/simultaneous inference (e.g. Bonferroni corrected	Overall quality of results good. Additional analysis completed to account for bias and power.	Statistical analysis completed. Power analysis completed. Post-hoc analysis or	Statistical test not appropriate or not completed. No power or post-hoc analysis.

analysis).		design not fully explained. Less clear.	
9) How indeterminate results, missing responses and outliers of the index tests were handled.	Described in detail. Explicitly states if participants excluded or data missing. Or no data excluded. Reason is justified and does not interfere with findings.	States data excluded, but reason or justification not given fully. Assumed but not stated all data included in analysis.	Not explained or accounted for. Missing. Excluded without justified explanation.
TOTAL SECTION C.	**/6**		
TOTAL QUALITY SCORE	**/18**		

Appendix A2: Systematic Review and Empirical Paper: Author Guidelines

Author Guidelines

AUTISM RESEARCH will cover research related to Autism Spectrum Disorder (ASD) and closely related neurodevelopmental disorders. The Journal focuses on basic genetic, neurobiological and psychological mechanisms and how these influence developmental processes in ASD. The Journal encourages the submission of research articles that take a developmental approach to the biology and psychology of autism, with a particular emphasis on identifying underlying mechanisms and integrating across different levels of analysis. Individuals included in research studies can span the full spectrum of ASD, including the broader phenotype, and there are no restrictions on study participants in terms of age or

intellectual ability. The Journal also encourages papers reporting work on animals or cell or other model systems that are directly relevant to a greater understanding of ASD. The journal will also publish reports of carefully conducted clinical trials of treatments for the core symptoms or one of the common comorbid symptoms of ASD. Papers reporting clinical trials will be judged, in part, on whether there is an empirical justification for using the treatment that is reported.

Research Article

Submissions of original research articles of broad interest and potential for high impact are encouraged. The text of these articles should include an Abstract, Introduction, Methods, Results and a Discussion. Research articles should be a maximum of 5,000 words in length. If there are extenuating circumstances that would require an author to extend the length of an article, please contact the editorial office with specific details and rationale for the Editor-in-Chief's consideration.

Conflict of Interest Disclosure

AUTISM RESEARCH requires that all authors disclose any potential sources of conflict of interest. Any interest or relationship, financial or otherwise, that might be perceived as influencing an author's objectivity is considered a potential source of conflict of interest. These must be disclosed when directly relevant or indirectly related to the work that the authors describe in their manuscript. Potential sources of conflict of interest include, but are not limited to, patent or stock ownership,

membership on a company board of directors, membership on an advisory board or committee for a company, and consultancy for, or receipt of speaker's fees from, a company. The existence of a conflict of interest does not preclude publication in this journal. If the authors have no conflict of interest to declare, they must also state this within the submitted manuscript.

It is the responsibility of the corresponding author to review this policy with all authors and to collectively list in the cover letter to the Editor-in-Chief, in the manuscript (under the Acknowledgment section), and in the online submission system ALL pertinent commercial and other relationships.

Format

The manuscript should have uniform style and be submitted exactly as the author wishes it to appear in print. It should be as concise as possible without omitting relevant results. All manuscript text in each of the sections described below must be double spaced. The manuscript should be subdivided into the following sequence with each section beginning on a new page:

Title page : The first page of the manuscript should include:

Title of paper, full name of author(s), institutional affiliation and complete address of all authors, running title not to exceed 45 letters and spaces, number of text pages, number of tables, number of figures.

Telephone and facsimile numbers and e-mail address of the corresponding author

Individual and address to whom correspondence concerning manuscript should be sent. All grant information in the following format: Grant sponsor _____; Grant number: _____.

Lay Abstract : Submit a brief (max 250 words) description of the paper that is understandable by the general public and which avoids technical jargon. This abstract will appear on the publicly accessible part of the Society's website.

Scientific Abstract : Submit an abstract of 250 words or less. The abstract must be written in complete sentences. It should concisely state the questions addressed, the methods used, the main results and their significance.

Key Words:

Append three to eight key words at the end of the abstract for the purposes of citing your work by the secondary services.

Text : This material should be divided into sections appropriate for the type of manuscript being submitted.

Abbreviation and Units: Use standard abbreviations. Spell out all nonstandard abbreviations the first time used. Abbreviations are contained in the current edition of the CBE style manual (sixth edition, 1994, Council of Biology Editors, Inc. Suite 230 N. Michigan Ave., Chicago, IL 60601). Spelling reference is to the current

edition of Webster's International Dictionary. In items of human gene nomenclature, this journal adheres to the principles specified in HUGO's HGNC guidelineshttp://www.genenames.org/guidelines.html where appropriate.

Figures and Legends: At acceptance of the manuscript, the authors must submit the final revised version of an accepted manuscript (text, tables, and illustrations) online. Text files must be submitted as .doc or .rtf files. Tables must be submitted as .doc or .rtf files (which can be embedded in the text file) or as separate .xls files. Figures must be submitted as .tif or .eps files. Do not submit PDFs, jpegs, or PowerPoint files. Please select LWZ compression (an option in the "save" process of programs such as Photoshop) when saving your figures. This is a lossless compression routine that reduces the size of figures without compromising their quality.

Figures should be submitted as electronic images to fit either one (55 mm, 2 3/16", 13 picas), two (115 mm, 4 1/2", 27 picas), or three (175 mm, 6 7/8", 41 picas) columns. The length of an illustration cannot exceed 227 mm (9"). Journal quality reproduction requires grey scale and color files at resolutions of 300 dpi. Bitmapped line art should be submitted at resolutions of 600-1200 dpi.

Helvetica typeface is preferred for lettering of illustrations. All letters, numbers and symbols must be at least 2 mm high. Courier typeface should be used for sequence figures. Number figures in one consecutive series with Arabic numerals, and key them into the text. Submit a brief descriptive legend with each illustration, and do not repeat results in figure legends. Legends for each figure should not exceed 200

words. Abbreviations used in figures and legends must match exactly those used in the text.

Color Figures: Color figures, when deemed necessary, are always published online free of charge.

Tables: Each table must have a self-explanatory title, be numbered in order of appearance with Arabic numerals and be cited at an appropriate point in the text. Tables should present comparisons of data that are too cumbersome to describe in the text; they should not merely repeat text information.

References

Reference should be made only to articles that are published or in press. Unpublished results and personal communications should be cited parenthetically in the text, not in the reference list. Authors are responsible for the accuracy of the references. References in the text should be made by author's name followed by the year of publication, arranged chronologically, then alphabetically. When there are more than two authors, use the first author's name and et al.

When references are made to more than one paper by the same author, published in the same year, designate them as a, b, c, etc. In the final list, arrange references alphabetically listing the first six authors, followed by et. al. where applicable, then year of publication. Spell out journal names in roman style, following these examples:

For Journals : Pinter, R., Hogge, W.A., & McPherson, E. (2004). Infant with severe penicillamine embryopathy born to a woman with Wilson disease. American Journal of Medical Genetics, Part A, 128A, 294–298.

Books: Reece, R.J. (2004). Analysis of genes and genomes. New York: Wiley-Liss. P.469.

Chapter in Book: Hunter, A.G.W. (2005). Down syndrome. In: Cassidy, S.B., Allanson, J.E., editors. Management of genetic syndromes, 2e. New York: Wiley-Liss, pp 191–210.

Web Citation : U.S. Census Bureau. (2004). America's families and living arrangements: 2003 (Table C3). Retrieved November 24, 2004, from http://www.census.gov/population/www/socdemo/hh-fam/cps2003.html

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Appendix B1: Ethics Proposal, Amendments and acceptance

LEVEL 1 SELF AUDIT FORM

SA1**Primary Research Question:**

Please tick	What type of research are you planning to do?
	Study of a novel intervention or randomised clinical trial to compare interventions in clinical Practice
√	Study utilising questionnaires, interviews or measures, including auto-ethnographic.
	Study limited to working with routinely collected clinical data
	Meta-analysis or systematic review
	Research database containing non-identifiable information

SA2**Please provide a brief summary of your proposed study. Our interest is in areas of your methodology where ethical issues may arise so please focus your detail on areas such as recruitment, consent, describing your participants and the nature of their involvement and data handling.**

Executive Dysfunction in Non-Learning Disabled Adults with Autism Spectrum Disorder (ASD)

The aim of the project is to investigate how adults with a diagnosis of ASD, without a cognitive impairment, perform on executive functioning tasks. I am hoping to use the results of self-reported questionnaires and neuropsychological assessments in order to bridge this literature gap and better aid clinical decision-making.

Research Questions / Objectives:

To investigate whether a sample of individuals with a diagnosis of ASD differ in performance on existing executive functioning task (D-KEFS) compared with typically developed (TD) controls who do not differ in sex, age and cognitive ability (Wechsler Adult Intelligence Scale-Forth Edition (WAIS-IV, Wechsler, 2008).

Potential Methodological and Ethical Issues

Recruitment; All participants will have gained higher grades (A Levels) or entry to university. Faculty administrators or disability coordinators at participating university institutions will contact individuals who meet this criteria. Individuals contacted from Disability Services will already have an established diagnosis of ASD and disclosed this to the university/support service that they attend.
If individuals are interested in taking part, they will contact the researcher. No confidential data will be given to the researcher regarding the participant without written consent.

Consent; As the study involves handling confidential data, lengthy confidential assessments and potentially sensitive feedback, every effort will be made to verify participant understanding of what the study will entail. It will be required that all TD and ASD participants provide written consent prior to the experiment. The purpose of the study and what it will entail will be made explicit both verbally and in writing. It is unlikely that individuals will have a learning disability, as they will have to obtained higher grades (A levels) or gained access to university.

Testing; The researcher will be testing participants alone in university/support service settings. They will make staff on the premises and a work colleague aware of the location and start/finishing time of the testing session. As some of the assessments are lengthy, lasting up to 1 hour 30 min, it may there is an effect of fatigue. Every effort will be made to ensure frequent breaks between subtests. Participants will also be reminded of the right to withdraw throughout testing, which will be made explicit at the time of consent.

Feedback; If participants request feedback on their performance it will be given. However, to avoid distress when feeding back the results of the assessments, an adapted feedback form for the WAIS-III will be used (see attached). Feedback for all assessments will be given sensitively, stressing individual strengths and omitting numerical scores.

Data handling; Data will be kept both electronically and in written form.

However, all electronic and written data will be anonymously stored, using participant numbers, separately from consent forms to preserve confidentiality. Only the researcher will be able to identify participants by their assigned number

Please circle your answer as appropriate:

	ETHICAL ISSUES		
SA 3	Bringing the University into disrepute **Is there any aspect of the proposed research which might bring the University into disrepute? For example, could any aspect of the research be considered controversial, prejudiced, critical of a minority group or religion etc.?**	**NO**	
SA 4	**Protection of research subject confidentiality** *Will you make every effort to protect research subject confidentiality by conforming to the University of Edinburgh's guidance on data security, protection and confidentiality as specified in:* http://www.ed.ac.uk/schools-departments/information-services/services/research-support/data-library/research-data-mgmt/data-security *For example, there are mutually understood agreements about:* (a) non-attribution of individual responses; (b) Individuals, and organisations where necessary, being anonymised in stored data, publications and presentations; (c) publication and feedback to participants and collaborators; (d) With respect to auto-ethnographic work it is recognised that the subject's anonymity cannot be maintained but the confidentiality of significant others must be addressed.		**YES**

SA5	**Data protection and consent** *Will you make every effort to ensure the confidentiality of any data arising from the project by complying with* the University of Edinburgh's Data Protection procedures (see www.recordsmanagement.ed.ac.uk); For example (a) Ensuring any participants recruited give consent regarding data collection, storage, archiving and destruction as appropriate; (b) Identifying information[1], (e.g. consent forms) is held separately from data and is only accessible by the chief investigator and their supervisors; (c) There are no other special issues arising regarding confidentiality/consent. (d) That where NHS data is being accessed Caldicott Guardian approval has been obtained.		**YES**
SA6	Duty to disseminate research findings Are there issues which will prevent all participants and relevant stakeholders having access to a clear, understandable and **accurate summary** of the research findings?	**NO**	
SA7	**Moral issues and Researcher/Institutional Conflicts of Interest** *Are there any SPECIAL MORAL ISSUES/CONFLICTS OF INTEREST?* Examples include, but are not limited to: • Where the purposes of research are concealed; • Where respondents are unable to provide informed consent • Where there is financial or non-financial benefit for *anyone* involved in the research, or for their relative or friend. • Where research findings could impinge negatively or differentially upon participants or stakeholders (for example	**NO**	

	when selecting an unrepresentative sample of a larger population). • Where there is a dual relationship between the researcher and subject? E.g. Where the researcher is also the subject's practitioner or clinician.		
SA8	***Potential physical or psychological harm, discomfort or stress*** Is there any foreseeable potential for: a) significant psychological harm or stress for participants? b) significant physical harm or discomfort for participants? c) significant risk to the researcher? Examples of issues/ topics that have the potential to cause psychological harm, discomfort or distress and should lead you to answer 'yes' to this question include, but are not limited to: *Relationship breakdown; bullying; bereavement; mental health difficulties; trauma / PTSD; Violence or sexual violence; physical, sexual or emotional abuse in either children or adults; feedback of results from the project's assessments.*		**YES**
SA9	**Vulnerable participants** Will you be ***recruiting*** any participants or interviewees who could be considered vulnerable? Examples of vulnerable groups, the inclusion of which should lead you to answer yes to this question include, but are not limited to: Clients or patients of either the researcher OR the person recruiting subjects; Children & young people; people		**YES**

	who are in custody or care for example, offenders, looked after children or nursing home resident; persons with mental health difficulties including those accessing self-help groups; auto-ethnographic researchers examining distressing topics.		

Assessment outcome:

SA10 **Have you circled any answers in <u>BOLD</u> typescript?** Please tick as appropriate

No ☐

(i) Your responses on the completed self-audit confirm the ABSENCE OF REASONABLY FORESEEABLE ETHICAL RISKS.
(ii) Please now read the guidance below and provide the required signatures.
(iii) You are NOT REQUIRED to complete a level 2/3 application form.
(iv) Please submit the UoE HSS Ethics Application Form electronic document (in its entirety) along with ALL additional required documentation, failure to do so will mean that your form is returned to you.

Yes ☑

(i) Your responses on the completed self-audit indicate that we require further information to consider your application.
(ii) Read the Guidance below and provide the required signatures.
(ii) You **ARE REQUIRED** to complete a level 2/3 application form.

(III) Please continue to page x of this document where you will find the level 2/3 form

Subsequent to submission of this form, <u>any</u> alterations in the proposed methodology of the project should be reviewed by both the applicant and their supervisor. If the change to methodology results in a change to any answer on the form, then a resubmission to the Ethics subgroup is required.

The principal investigator is responsible for ensuring compliance with any additional ethical requirements that might apply, and/or for compliance with any additional requirements for review by external bodies.

Hollie Burnett 24/01/2015

Student Name Date signed

Suzanne O'Rourke 30.08.2012

Academic Supervisor (2): Name Signature Date signed

LEVEL 2 / 3 ETHICAL REVIEW

Complete only if indicated in the conclusion of your level 1 form.

RISKS TO, AND SAFETY OF, RESEARCHERS NAMED IN THIS APPLICATION

ER1**Do any of those conducting the research named above need appropriate training to enable them to conduct the proposed research safely and in accordance with the ethical principles set out by the College?**

YES: It is a requirement by NHS Code of Practice that researchers working with potentially vulnerable groups (e.g. children) have a Disclosure Scotland (CRB), which the researcher has, as part of their clinical doctorate training. The study will also require the researcher to have extensive knowledge of complex psychometric tests and have had training to administer them. This has being provided as part of the clinical doctorate training programme and through clinical placements. Also, supervisors will provide further support for the researcher to use and interpret the clinical tools throughout supervision.

ER2**Are any of the researchers likely to be sent or go to any areas where their safety may be compromised, or they may need support to deal with difficult issues?**

YES: Personal safety will also be addressed. Testing alone with individuals unknown to NHS, in university settings should be done with caution. The researcher will make staff on the premises and a work colleague aware of location and start/finishing time of the testing session.

[ER3]**Could researchers have any conflicts of interest?**

NO

RISKS TO, AND SAFETY OF, PARTICIPANTS

[ER4]**Are any of your participants children or protected adults (protected adults are those in receipt of registered care, health, community care or welfare services – please refer to?**

NO

Do any of the researchers taking part in this study require Disclosure Scotland approval?

NO

[ER5]**Could the research induce any psychological stress or discomfort?**

YES: As some of the assessments are lengthy, lasting up to 1 hour 30 minutes, it may be that there is an effect of fatigue. Every effort will be made to ensure frequent breaks between subtests. Participants will also be reminded of the right to withdraw throughout testing, which will be made explicit at the time of consent.

To avoid distress when feeding back the results of the assessments, a standardised feedback form for the WAIS-III will be adapted and used (see attached). Feedback for all assessments will be given sensitively, stressing individual strengths and weaknesses.

[ER6]**Does the research involve any physically invasive or potentially physically harmful procedures?**

NO

[ER7]**Could this research adversely affect participants in any other way?**

Other than those mentioned in ER5 there are no further risks.

RESEARCH DESIGN

ER8**Does the research involves living human subjects specifically recruited for this research project**

YES

ER9 How many participants will be involved in the study?

The clinical group will consist in total of 22 adults with autism spectrum disorder (ASD). Individuals contacted from Disability Services will already have an established diagnosis of ASD and disclosed this to the university/Support Service that they attend. The control group will consist of approximately 22 typically developed (TD) individuals that attend/have attended/are about to attend university. Twenty-two TD students would be a sufficient control sample for the clinical group, when age, sex and cognitive ability are accounted for, determined by *an a priori* power analysis (Gpower: Faul & Erfelder, 1992), *Power = 0.95*. However, depending on time constraints, more participants will be tested so that a correlational analysis may be performed, comparing the performance on executive functioning measures (D-KEFS) with autistic-like traits (AQ). A total sample of 64 participants will be required to perform a correlation analysis, as determined by *a priori* power analysis (Gpower: Faul & Erfelder, 1992), *Power = 0.8.*

ER10**What criteria will be used in deciding on inclusion/exclusion of participants?**

It will be required that all TD and ASD participants have normal or corrected-to-normal vision and hearing, and provide written consent prior to the experiment. Participants will be excluded if they disclose that they have previously had a traumatic brain injury or disclose that they have a comorbid psychiatric condition (e.g. psychosis), this will be further explained during consent. English will be required to be the participants' first language. The study will include participants over the age of 17. Participants that are outliers in terms of age may be excluded (i.e. mature students) due to not being able to find controls of a similar age in the TD group. If an individual scores less than 70 on the abbreviated intelligence test (the cut-off for learning disability) then their data will be excluded from analysis.

Individuals with ASD must previously have received a diagnosis of ASD that is congruent to the current DSM-V (2013) classification. The AQ will be used as a screening tool to exclude TD participants who score 32 or more from the TD group, but their data may be included in a correlation analysis.

ER11**How will the sample be recruited? (E.g. posters, letters, a direct approach- specify by whom.)**

All participants will be contacted by faculty administrators or disability coordinators at participating support services/ university institutions. At Support Services, the administrator will specify in the distributed information the academic criteria (higher level grade/degree). This will be done by giving an information leaflet about the study by e-mail or by hand. If individuals are interested in taking part, they will contact the researcher by e-mail. No confidential data will be given to the researcher regarding the participant without written consent, as the potential participant will be responsible for approaching the researcher.

ER12**Will the study involve groups or individuals who are in custody or care, such as students at school, self-help groups, residents of nursing home?**

No

ER13**Will there be a control group?**

The control group will consist of approximately 22 students recruited from a range of university departments in the arts, natural sciences, humanities and social sciences. All students in participating subject areas will be sent an email by their faculty administrators containing information about the study and given the contact details of the researcher to contact if they are interested in taking part. It may be that some students take part in the study for a course credit.

ER14**What information will be provided to participants prior to their consent? (e.g. information leaflet, briefing session)**

An information sheet will be given to the faculty administrators or disability coordinators at participating university institutions. They will give this to individuals with a diagnosis of ASD and TD individuals, by hand or e-mail. If individuals are interested in taking part, they will contact the researcher by e-mail. Then written consent will be obtained, prior to completing any of the assessments described.

The participants will also be informed how long the study will be expected to last. This is based on testing manuals' recommendations and a short pilot, involving an assistant psychologist.

ER15**Participants have a right to withdraw from the study at any time. Please tick to confirm that participants will be advised of their rights, including the right to continue receiving services if they withdraw from the study.**

√ **YES**

ER16**Will it be necessary for participants to take part in the study without their knowledge and consent? (e.g. covert observation of people in non-public places)**

NO

ER17**Where consent is obtained, what steps will be taken to ensure that a written record is maintained?**

Written consent will be obtained and stored securely on NHS property, for the duration of the study (until May 2016). All other electronic and written data will be anonymously stored, using participant numbers, separately from consent forms to preserve confidentiality. Only the researcher will be able to identify participants by their assigned number.

ER18**In the case of participants whose first language is not English, what arrangements are being made to ensure informed consent?**

The majority of assessments are extremely cognitive and verbally demanding, therefore the information sheet will clearly state that only individuals who are native English speakers can participate in the study.

ER19**Will participants receive any financial or other benefit from their participation?**

Participants will receive no financial benefit from participation. However, some faculties require students to participate in research for course credits, which may be made available for the purposes of this study. The participant will be made aware of the potential benefits the study may have in terms of improving health and care through new knowledge. The participant will also have the opportunity to receive feedback should they wish, which will be sensitively given, stressing strengths and weaknesses.

ER20**Are any of the participants likely to be particularly vulnerable, such as elderly or disabled people, adults with incapacity, your own students, members of ethnic minorities, or in a professional or client relationship with the researcher?**

The clinical sample will have a diagnosis of ASD, and it is unlikely that they will have a learning disability, having gained higher grades or entry to university and they are over the age of 17. Every effort will be made to ensure that individuals understand the costs/benefits of participation and right to withdraw at the time of consent according to the *Good Practice Guidelines* (2011). Information and the consent forms will be presented both visually and verbally.

ER21**Will any of the participants be under 16 years of age?**

No

ER22**Will any of the participants be interviewed in situations which will compromise their ability to give informed consent, such as in prison, residential care, or the care of the local authority?**

No

DATA PROTECTION

ER23 Will any part of the research involve audio, film or video recording of individuals?

No

ER24**Will the research require collection of personal information from any persons without their direct consent?**

No

ER25**How will the confidentiality of data, including the identity of participants (whether specifically recruited for the research or not) be ensured?**

NHS Code of Practice on Protecting Patient Confidentiality (2003) states that researchers must inform participants of the purpose of the study, who will manage the data, how it will be stored and the limitations of confidentiality. *Good Practice Guidelines* (2011) will be followed to ensure participants are given clear, accurate information.

In accordance to the Data Protection Act (1998), consent forms will have an identifying number, which will then be assigned to questionnaires. Consent forms will be stored in a locked cabinet, separate from questionnaires, in order to maintain confidentiality. Questionnaires, forms and electronic data will not contain identifying information (e.g. name, address, DoB or combination of potentially identifying information), just the participants assigned number. Following completion of the study (May 2016), consent forms will be destroyed.

ER26**Who will be entitled to have access to the raw data?**

Only the principal researcher and supervisors will have access to any data.

ER27**How and where will the data be stored, in what format, and for how long?**

The consent forms will be stored in a locked cabinet on an NHS site, which only the researcher will have access to. The electronic data, forms and questionnaires will have no potentially identifying information, just the participant's assigned number. It is predicted that the data will be retained until the study is completed in May 2016, and then destroyed securely, following NHS guidelines.

ER28**What steps have been taken to ensure that only entitled persons will have access to the data?**

Identifiable data will be stored securely in a filing cabinet on an NHS site which will only be accessible by the principal researcher.

ER29**How will the data be disposed of?**

Data will be disposed of in accordance with NHS Fife policies for confidential information by May 2016.

ER30**How will the results of the research be used?**

As it is a requirement of the doctorate in clinical psychology, the project will be written as part of a thesis, conference poster, and will be submitted to a peer-reviewed journal. Feedback of results will also be given to the support services/ universities and the participants that took part and the clinical psychology department where the researcher is based.

ER31**What feedback of findings will be given to participants?**

NHS code of practice states that participants should be given formal feedback in no more than 40 days of request. Professional standards (HPC, 2012) require researchers to share feedback with participants sensitively, demonstrating strengths and weaknesses, rather than 'whole test scores' to avoid any distress or disappointment. A standardised client feedback summary, provided for the WAIS-III will be adapted.

ER32**Is any information likely to be passed on to external companies or organisations in the course of the research?**

No

ER33**Will the project involve the transfer of personal data to countries outside the European Economic Area?**

No

ER34**An application at this level is likely to require additional documentation, for example consent forms or participant information sheets. Please return to the Documentation Checklist on page 2 to list your supporting documentation.**

REFERENCES

NHS Code of Practice on Protecting Patient Confidentiality (2003): www.show.scot.nhs.uk/confidentiality

Data Protection Act (1998): http://www.legislation.gov.uk/ukpga/1998/29/contents

Good Practice Guidelines:http://www.psy.ed.ac.uk/psy_research/documents/

PARTICIPANT INFORMATION SHEET: ASD

STUDY TITLE: Executive Functioning in Non-Learning Disabled Adults with Autism Spectrum Disorder (ASD)

You are being invited to take part in a research study. Before you decide if you would like to take part, it is important for you to understand why the research is being done and what it would involve for you. Please take time to read the following information carefully, and talk to others about the study if you wish. If there is anything that is unclear, or if you have any questions, please do not hesitate to ask the principal researcher. You can also contact them using the details at the end of this sheet.

WHAT IS THE RESEARCH ABOUT?

The aim of the project is to investigate how adults with ASD, without a learning disability, perform on cognitive tasks. We are hoping to use the results of self-reported questionnaires and assessments in order to help clinicians be better informed when working with patients.

WHY HAVE I BEEN INVITED?

We are inviting individuals that have higher level grads (A levels)/attend/attended university with a diagnosis of ASD to take part in the study. This brief study will involve up to sixty-four participants.

DO I HAVE TO TAKE PART?

No. It is up to you to decide whether or not to join this study. If you do decide to take part, you will be asked to sign a consent form, and you will be able to keep this information sheet for reference. If you decide to take part you will still be able to withdraw from the study at any point and you do not have to give a reason.

WHO CANNOT TAKE PART?

It will be required that all participants have normal or corrected to normal vision and hearing. Participants must not have had a traumatic brain injury in the past or presently have any mental health problems (such as severe depression). This is so the researcher is clear that your performance is to the best of your ability and not impacted by how you are presently feeling or due to an injury you have sustained. A participant's first language must be English, due to the high level of verbal demands in the assessments which may be more difficult for non-native speakers.

WHAT WILL HAPPEN TO ME IF I TAKE PART?

If you choose to take part in this study, you will be required to complete two questionnaires which may take up to 15 minutes of your time. Additionally you will be asked to complete two cognitive assessments and a semi-structured interview which may take up to three hours. The assessments are designed to investigate different cognitive skills (e.g. working memory, attention switching) and the semi-structured interview is designed to measure autistic traits.

WHAT ARE THE POSSIBLE RISKS AND DISADVANTAGES OF TAKING PART?

There are no identified risks in taking part in this study. However, some tasks may take a long time to complete. Although the researcher will offer frequent breaks throughout, you can request additional breaks or ask to stop the assessment at any time. If you request feedback from your performance on the assessments this will be sensitively given. However, it is possible that you may feel dissatisfied or that the feedback may cause some distress; if this should occur you can inform the researcher and or follow the complaints procedure (e-mail addresses below).

WHAT ARE THE POSSIBLE BENEFITS OF TAKING PART?

It is likely that there will not be an immediate benefit to you, but your participation will aid in the development of a research project. This larger project aims to improve our understanding of cognitive strengths and weaknesses in individuals with ASD. Individual feedback on performance for the cognitive assessment will be available upon request.

WHAT HAPPENS IF THERE IS A PROBLEM?

Any issues or complaints that you might have will be addressed by the research team. Full details of how you can contact us are outlined at the end of this sheet. If you should wish to make a formal complaint, you can do so by using the University of Edinburgh Complaints procedure. Full details are provided at the end of the information sheet.

WILL MY TAKING PART IN THE STUDY BE KEPT CONFIDENTIAL?

All the information we collect from you will be kept confidential. We will keep your data anonymous during the research by assigning it a unique reference number. This reference number will be the only link between your data and your consent form. Once the study has been completed, your consent form and any identifiable data will be destroyed but your anonymous data may be held for future authorised research. During the study your data will be securely stored on an NHS site and will be kept separately from consent forms.

WHAT WILL HAPPEN TO THE RESULTS OF THE RESEARCH STUDY?

This study will be written up and reported in a thesis format in order to satisfy the requirements of the doctorate in clinical psychology. We also hope to submit this thesis project to relevant academic journals for publication. Your name or any identifiable date will not appear in any reports or publications. Should you wish to enquire about the progress of the research, you can contact the principal researcher, Hollie Burnett.

WHO IS ORGANISING AND FUNDING THE RESEARCH?

The study is being organised by Dr Hollie Burnett, specialist psychological practitioner, under the supervision of Dr Jill Cossar and Dr Suzanne O'Rourke (University of Edinburgh). The study is being supported by both NHS Fife and the University of Edinburgh.

WHO HAS REVIEWED THE STUDY?

This project has reviewed by the Section of Clinical Psychology Ethics Research Panel at the University of Edinburgh.

CONTACT INFORMATION

If you have any complaints or concerns, please contact the principal researcher: Hollie Burnett Email: **s1163678@sms.ed.ac.uk**

If you should wish to make a formal complaint, you can do so by writing to the University of Edinburgh: *The Investigations Manager, SASG Business Unit, The University of Edinburgh, Old College, South Bridge, Edinburgh, EH8 9YL. Email: complaints@ed.ac.uk*or to the project supervisor *Email: Suzanne.O'Rourke@ed.ac.uk*

INFORMED CONSENT FORM: ASD

STUDY TITLE: Executive Functioning in Non-Learning Disabled Adults with Autism Spectrum Disorder (ASD)

PROJECT SUMMARY

The aim of the project is to investigate how adults with ASD, without a cognitive impairment perform on cognitive assessments. We are hoping to use the results of self-reported questionnaires and assessments in order to help clinicians be better informed when working with patients.

By signing below, you are agreeing that (please tick each statement):

(1) You have read and understood the Participant Information Sheet. ☐

(2) Questions about your participation in this study have been answered to your satisfaction. ☐

(3) You are aware of the potential risks (if any). ☐

(4) You are taking part in this research study voluntarily. ☐

(5) You are aware that you may withdraw from the study at any point and that this will not affect support you may receive in the future. ☐

_____

Participant's Name (Printed)

_____ _____

Participant's signature **Date**

_____ _____

Name of person obtaining consent (Printed) **Signature of person obtaining consent**

PARTICIPANT INFORMATION SHEET: TD

STUDY TITLE: Executive Functioning in Non-Learning Disabled Adults with Autism Spectrum Disorder (ASD)

You are being invited to take part in a research study. Before you decide if you would like to take part, it is important for you to understand why the research is being done and what it would involve for you. Please take time to read the following information carefully, and talk to others about the study if you wish. If there is anything that is unclear, or if you have any questions, please do not hesitate to ask the principal researcher. You can also contact them using the details at the end of this sheet.

WHAT IS THE RESEARCH ABOUT?

The aim of the project is to investigate how adults with ASD, without a cognitive impairment, perform on tasks. We are hoping to use the results of self-reported questionnaires and assessments in order to help clinicians be better informed when working with patients.

WHY HAVE I BEEN INVITED?

You are been invited to take part because you **DO NOT** have a diagnosis of ASD, and you will be in a control group for participants who do have a diagnosis of ASD. This brief study will involve up to sixty-four participants.

DO I HAVE TO TAKE PART?

No. It is up to you to decide whether or not to join this study. If you do decide to take part, you will be asked to sign a consent form, and you will be able to keep this information sheet for reference. If you decide to take part you will still be able to withdraw from the study at any point and you do not have to give a reason.

WHO CANNOT TAKE PART?

It will be required that all participants have normal or corrected to normal vision and hearing. Participants must not have had a traumatic brain injury in the past or presently have any mental health problems (such as severe depression). This is so the researcher is clear that your performance is to the best of your ability and not impacted by how you are presently feeling or due to an injury you have sustained. A participant's first language must be English, due to the high level of verbal demands in the assessments which may be more difficult for non-native speakers.

WHAT WILL HAPPEN TO ME IF I TAKE PART?

If you choose to take part in this study, you will be required to complete two questionnaires which may take up to 15 minutes of your time. Additionally you will be asked to complete two cognitive assessments and a semi-structured interview which may take up to three hours. The assessments are designed to investigate different cognitive skills (e.g. working memory, attention switching) and the semi-structured interview is designed to measure autistic traits.

WHAT ARE THE POSSIBLE RISKS AND DISADVANTAGES OF TAKING PART?

There are no identified risks in taking part in this study. However, some tasks may take a long time to complete. Although the researcher will offer frequent breaks throughout, you can request additional breaks or ask to stop the assessment at any time. If you request feedback from your performance on the assessments this will be sensitively given. However, it is possible that you may feel dissatisfied or that the feedback may cause some distress; if this should occur you can inform the researcher and or follow the complaints procedure (e-mail addresses below).

WHAT ARE THE POSSIBLE BENEFITS OF TAKING PART?

It is likely that there will not be an immediate benefit to you, but your participation will aid in the development of a research project. This larger project aims to improve our understanding of cognitive strengths and weaknesses in individuals with ASD. Individual feedback on performance for the cognitive assessment will be available upon request.

WHAT HAPPENS IF THERE IS A PROBLEM?

Any issues or complaints that you might have will be addressed by the research team. Full details of how you can contact us are outlined at the end of this sheet. If you should wish to make a formal complaint, you can do so by using the University of Edinburgh Complaints procedure. Full details are provided at the end of the information sheet.

WILL MY TAKING PART IN THE STUDY BE KEPT CONFIDENTIAL?

All the information we collect from you will be kept confidential. We will keep your data anonymous during the research by assigning it a unique reference number. This reference number will be the only link between your data and your consent form. Once the study has been completed, your consent form and any identifiable data will be destroyed but your anonymous data may be held for future authorised research. During the study your data will be securely stored on an NHS site and will be kept separately from consent forms.

WHAT WILL HAPPEN TO THE RESULTS OF THE RESEARCH STUDY?

This study will be written up and reported in a thesis format in order to satisfy the requirements of the doctorate in clinical psychology. We also hope to submit this thesis project to relevant academic journals for publication. Your name or any identifiable date will not appear in any reports or publications. Should you wish to enquire about the progress of the research, you can contact the principal researcher, Hollie Burnett.

WHO IS ORGANISING AND FUNDING THE RESEARCH?

The study is being organised by Dr Hollie Burnett, specialist psychological practitioner, under the supervision of Dr Jill Cossar and Dr Suzanne O'Rourke (University of Edinburgh). The study is being supported by both NHS Fife and the University of Edinburgh.

WHO HAS REVIEWED THE STUDY?

This project has reviewed by the Section of Clinical Psychology Ethics Research Panel at the University of Edinburgh.

CONTACT INFORMATION

If you have any complaints or concerns, please contact the principal researcher: Hollie Burnett Email: **s1163678@sms.ed.ac.uk**

If you should wish to make a formal complaint, you can do so by writing to the University of Edinburgh: *The Investigations Manager, SASG Business Unit, The University of Edinburgh, Old College, South Bridge, Edinburgh, EH8 9YL. Email: complaints@ed.ac.uk* or to the project supervisor *Email: Suzanne.O'Rourke@ed.ac.uk*

INFORMED CONSENT FORM:

TD

STUDY TITLE: Executive Functioning in Non-Learning Disabled Adults with Autism Spectrum Disorder (ASD)

PROJECT SUMMARY

The aim of the project is to investigate how adults with ASD, without a cognitive impairment perform on tasks. We are hoping to use the results of self-reported questionnaires and assessments in order to help clinicians be better informed when working with patients. You are been invited to take part because you **DO NOT** have a diagnosis of ASD/Asperger syndrome, and you will be in a control group for participants who do have a diagnosis of ASD/Asperger syndrome.

By signing below, you are agreeing that (please tick each statement):

(1) You have read and understood the Participant Information Sheet. ☐

(2) Questions about your participation in this study have been answered to your satisfaction. ☐

(3) You are aware of the potential risks (if any). ☐

(4) You are taking part in this research study voluntarily. ☐

(5) You are aware that you may withdraw from the study at any point and that this will not affect support that you may receive in the future. ☐

_____

Participant's Name (Printed)

_____ _____

Participant's signature **Date**

_____ _____

Name of person obtaining consent (Printed) **Signature of person obtaining consent**

Appendix B3: Measures

The Autism Quotient (AQ)

The AQ is a fifty-statement, self-administered questionnaire designed to measure the degree to which an adult with normal intelligence has traits associated with ASD. Participants rate their own behaviour in terms of social skills, attention switching, attention to detail, communication and imagination on a four-point scale (definitely agree, slightly agree, slightly disagree, and definitely disagree). Although the AQ is not a diagnostic tool, a score of 32 or higher (out of 50) has been shown to correlate with a diagnosis of ASD (Baron-Cohen et al., 2001; Wheelwright et al., 2006; Wakabyashi et al., 2006; Bartels, Cath, & Boomsma, 2007). Internal consistency is reported between .71-.81. The internal consistency of items in each of the five domains was also calculated, and Cronbach's Alpha coefficients were all moderate to high (communication = 0.65; social, = 0.77; imagination = 0.65; local details = 0.63; attention Switching = 0.67).

The Adult Autism Spectrum Quotient (AQ)

Ages 16+

For full details, please see: S. Baron-Cohen, S. Wheelwright, R. Skinner, J. Martin and E. Clubley, (2001) The Autism Spectrum Quotient (AQ) : Evidence from Asperger Syndrome/High Functioning Autism, Males and Females, Scientists and Mathematicians Journal of Autism and Developmental Disorders 31:5-17

Name:..... Sex:.....

Date of birth:..... Today's Date.....

How to fill out the questionnaire

Below are a list of statements. Please read each statement very carefully and rate how strongly you agree or disagree with it by circling your answer. **DO NOT MISS ANY STATEMENT OUT.**

Examples

E1. I am willing to take risks.	definitely agree	slightly agree	slightly disagree	definitely disagree
E2. I like playing board games.	definitely agree	slightly agree	slightly disagree	definitely disagree
E3. I find learning to play musical instruments easy.	definitely agree	slightly agree	slightly disagree	definitely disagree
E4. I am fascinated by other cultures.	definitely agree	slightly agree	slightly disagree	definitely disagree

1. I prefer to do things with others rather than on my own.	definitely agree	slightly agree	slightly disagree	definitely disagree
2. I prefer to do things the same way over and over again.	definitely agree	slightly agree	slightly disagree	definitely disagree
3. If I try to imagine something, I find it very easy to create a picture in my mind.	definitely agree	slightly agree	slightly disagree	definitely disagree
4. I frequently get so strongly absorbed in one thing that I lose sight of other things.	definitely agree	slightly agree	slightly disagree	definitely disagree
5. I often notice small sounds when others do not.	definitely agree	slightly agree	slightly disagree	definitely disagree
6. I usually notice car number plates or similar strings of information.	definitely agree	slightly agree	slightly disagree	definitely disagree
7. Other people frequently tell me that what	definitely	slightly	slightly	definitely

I've said is impolite, even though I think it is polite.	agree	agree	disagree	disagree
8. When I'm reading a story, I can easily imagine what the characters might look like.	definitely agree	slightly agree	slightly disagree	definitely disagree
9. I am fascinated by dates.	definitely agree	slightly agree	slightly disagree	definitely disagree
10. In a social group, I can easily keep track of several different people's conversations.	definitely agree	slightly agree	slightly disagree	definitely disagree
11. I find social situations easy.	definitely agree	slightly agree	slightly disagree	definitely disagree
12. I tend to notice details that others do not.	definitely agree	slightly agree	slightly disagree	definitely disagree
13. I would rather go to a library than a party.	definitely agree	slightly agree	slightly disagree	definitely disagree
14. I find making up stories easy.	definitely agree	slightly agree	slightly disagree	definitely disagree
15. I find myself drawn more strongly to people than to things.	definitely agree	slightly agree	slightly disagree	definitely disagree
16. I tend to have very strong interests which I get upset about if I can't pursue.	definitely agree	slightly agree	slightly disagree	definitely disagree
17. I enjoy social chit-chat.	definitely agree	slightly agree	slightly disagree	definitely disagree
18. When I talk, it isn't always easy for others to get a word in edgeways.	definitely agree	slightly agree	slightly disagree	definitely disagree
19. I am fascinated by numbers.	definitely agree	slightly agree	slightly disagree	definitely disagree
20. When I'm reading a story, I find it difficult to work out the characters' intentions.	definitely agree	slightly agree	slightly disagree	definitely disagree
21. I don't particularly enjoy reading fiction.	definitely agree	slightly agree	slightly disagree	definitely disagree
22. I find it hard to make new friends.	definitely agree	slightly agree	slightly disagree	definitely disagree

23. I notice patterns in things all the time.	definitely agree	slightly agree	slightly disagree	definitely disagree
24. I would rather go to the theatre than a museum.	definitely agree	slightly agree	slightly disagree	definitely disagree
25. It does not upset me if my daily routine is disturbed.	definitely agree	slightly agree	slightly disagree	definitely disagree
26. I frequently find that I don't know how to keep a conversation going.	definitely agree	slightly agree	slightly disagree	definitely disagree
27. I find it easy to "read between the lines" when someone is talking to me.	definitely agree	slightly agree	slightly disagree	definitely disagree
28. I usually concentrate more on the whole picture, rather than the small details.	definitely agree	slightly agree	slightly disagree	definitely disagree
29. I am not very good at remembering phone numbers.	definitely agree	slightly agree	slightly disagree	definitely disagree
30. I don't usually notice small changes in a situation, or a person's appearance.	definitely agree	slightly agree	slightly disagree	definitely disagree
31. I know how to tell if someone listening to me is getting bored.	definitely agree	slightly agree	slightly disagree	definitely disagree
32. I find it easy to do more than one thing at once.	definitely agree	slightly agree	slightly disagree	definitely disagree
33. When I talk on the phone, I'm not sure when it's my turn to speak.	definitely agree	slightly agree	slightly disagree	definitely disagree
34. I enjoy doing things spontaneously.	definitely agree	slightly agree	slightly disagree	definitely disagree
35. I am often the last to understand the point of a joke.	definitely agree	slightly agree	slightly disagree	definitely disagree
36. I find it easy to work out what someone is thinking or feeling just by looking at their face.	definitely agree	slightly agree	slightly disagree	definitely disagree
37. If there is an interruption, I can switch back to what I was doing very quickly.	definitely agree	slightly agree	slightly disagree	definitely disagree
38. I am good at social chit-chat.	definitely agree	slightly agree	slightly disagree	definitely disagree
39. People often tell me that I keep going on	definitely	slightly	slightly	definitely

and on about the same thing.	agree	agree	disagree	disagree
40. When I was young, I used to enjoy playing games involving pretending with other children.	definitely agree	slightly agree	slightly disagree	definitely disagree
41. I like to collect information about categories of things (e.g. types of car, types of bird, types of train, types of plant, etc.).	definitely agree	slightly agree	slightly disagree	definitely disagree
42. I find it difficult to imagine what it would be like to be someone else.	definitely agree	slightly agree	slightly disagree	definitely disagree
43. I like to plan any activities I participate in carefully.	definitely agree	slightly agree	slightly disagree	definitely disagree
44. I enjoy social occasions.	definitely agree	slightly agree	slightly disagree	definitely disagree
45. I find it difficult to work out people's intentions.	definitely agree	slightly agree	slightly disagree	definitely disagree
46. New situations make me anxious.	definitely agree	slightly agree	slightly disagree	definitely disagree
47. I enjoy meeting new people.	definitely agree	slightly agree	slightly disagree	definitely disagree
48. I am a good diplomat.	definitely agree	slightly agree	slightly disagree	definitely disagree
49. I am not very good at remembering people's date of birth.	definitely agree	slightly agree	slightly disagree	definitely disagree
50. I find it very easy to play games with children that involve pretending.	definitely agree	slightly agree	slightly disagree	definitely disagree

Developed by:

The Autism Research Centre

University of Cambridge

The Delis-Kaplan Executive Functioning System (D-KEFS)

The D-KEFS (Delis et al., 2001) is a battery consisting of nine individually administered tests, designed to measure cognitive abilities such as attention, perception and language. Administration should take approximately 15 minutes per subtest and 90 minutes to complete the full battery. The D-KEFS is designed for use with children as young as 8 years and adults in their upper 80s. Normative data has been standardised for 8/9 subtests, on a sample of 1,700 individuals from 8-89 years old. The 'proverb' test is standardised for 16-89 years old. The domains include: concept formation, problem solving, creativity, impulse control, inhibition, flexibility and planning. Each subtest of the D-KEFS is a standalone measure of executive functioning and there are no aggregate indexes or composite scores. Internal consistency is reported to range from .80 to .98.

The Wechsler Adult Intelligence Scale (WAIS-IV-UK)

The Wechsler batteries are the most common cognitive assessment used for assessing the intelligence of TD adults and children. The WAIS-IV-UK (2010) provides scores for verbal IQ, non-verbal (performance) IQ, and full scale IQ, along with four secondary indices (verbal comprehension, working memory, perceptual organization, and processing speed). There are ten subtests and five supplemental tests. The Wechsler tests have yielded a fairly consistent profile for individuals with ASD (Frith, 1989), with superior performance on the block design and extremely poor performance on the picture arrangement and comprehension tasks. Results of the performance tests are usually better than those of verbal tests. However, varying results have being found within an AS population, with some studies reporting little

difference compared with TD individuals. The WAIS-IV-UK takes around 60-90 minutes to complete.

The WAIS–IV was standardised on a normative sample of 2,200 individuals and divided into 13 age bands spanning ages 16:0 to 90:11. The sample was stratified to match demographic variables: age, sex, education level, race/ethnicity, and geographic region; studies include a sample of individuals with ASD. The technical manual provides extensive reliability data for individual subtests using coefficient alpha. Coefficients are good to excellent in all cases, ranging from the low .80s to upper .90s.

Only three subtests of the WAIS-IV will be used to give an indication of working memory abilities (digit span test), verbal intelligence (vocabulary test) and non-verbal intelligence (block design test).

Appendix B4: Participant Feedback

PARTICIPANT FEEDBACK SHEET

PRIVATE AND CONFIDENTIAL

Dear X,

As requested, the feedback for performance on the Wechsler Abbreviated Scale of Intelligence (WASI; Wechsler- Two, 2011) and the subtest from the Wechsler Adult Intelligence Scale – Fourth Edition (WAIS-IV: Wechsler, 2008) is described below.

The WASI-II and WAIS-IV are individually administered tests of an adult's intellectual ability and cognitive profile. You completed some subtests that come under indices measuring: verbal comprehension, perceptual reasoning and working memory. An individual may have scores that fall into categories 'extremely low' to 'very superior'. Most individuals, however, perform within the 'average range'. The WASI-II and WAIS-IV scores should be interpreted with some caution because any individual may score slightly higher or lower if tested again on a different day.

The results indicate that overall, your general cognitive ability lies within the **high average range** of intellectual abilities. However, this is only an indication of your

cognitive ability as the full battery was not completed. Also, significant differences within your performance indicated certain strengths and weaknesses, so it is more meaningful to consider your performance on each index separately:

Verbal Comprehension

The verbal comprehension index examines verbal conceptualisation, knowledge and expression. This includes answering oral questions that measure factual knowledge, word meanings, reasoning and the ability to express ideas in words. It can also be thought of assessing 'school learned' knowledge or skills. Your performance on the verbal comprehension index fell into the **high average range**.

Perceptual Reasoning

Your perceptual reasoning index (a measure of perceptual and fluid reasoning, spatial processing and visuo-motor integration) was made up from subtests examining the integration of information presented visually with non-verbal reasoning to solve problems (i.e. making blocks into patterns, choosing patterns that successfully make up bigger patterns and seeing themes/patterns among visual diagrams). Your performance fell into the **high average range**.

Working Memory

Working memory is the ability to manipulate and hold information in mind. The subtests involve handling numbers and letters in a step-by-step, sequential fashion and letter-number sequencing. Your performance on this index fell within the **low average range**.

Summary

In short, based on the results there is a suggestion that although you demonstrate stronger verbal and perceptual reasoning, your performance on the assessment is indicative of some difficulty with working memory. However, as mentioned your performance was still within the average range. It is normal for individuals to do better at some assessments than others: we all have strengths and weaknesses. It may benefit you to rehearse things that you need to learn, to write them down or to have visual prompts.

Thank you again for taking part in this study. Please do not hesitate to contact me if you have any queries.

Yours sincerely

Hollie Burnett

(Trainee Clinical Psychologist)