Abnormal Sensory Behaviours within the Autism Spectrum

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Declaration

This thesis is submitted for the Doctorate in Clinical Psychology at the University of Sheffield. It has not been submitted for any other degree or to any other academic institution.

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Overall Abstract

Literature Review

The current systematic literature review examined the relationship between sensory and repetitive and restricted behaviours in individuals with autism. Fifteen studies were selected according to relevant search terms and inclusion/exclusion criteria. Although results showed significant correlations between sensory and repetitive behaviours, there was much variability. The relationship was also likely to be artificially inflated due to the weaknesses of the measures used to assess these constructs. Methodological weaknesses of included studies are discussed as well as clinical implications and recommendations for future research.

Research Report

The research report attempted to reconcile two competing (neuronal inhibition verses excitation) theories of autism, by examining the impact of epilepsy (a disorder caused by increased excitation) on visual orientation discrimination abilities (whereby superior orientation abilities thought to be an index of increased inhibition). In line with the inhibition theory it was hypothesised that the ASD would show significantly better orientation discrimination abilities, whereas the epilepsy group would perform significantly poorer. Orientation discrimination abilities were compared in three groups of children; those with ASD, epilepsy or neuro-typical controls. Results found no superior discrimination abilities in the ASD group which may suggest that visual discrimination abilities are not a reliable marker for increased inhibition. However, the epilepsy group showed significantly poorer discrimination abilities compared to neuro-typical controls. This would be expected by both inhibition and excitation theories. Methodological weaknesses, theoretical implications and suggestions for further research are considered.

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Section 1: Literature Review

The Relationship between Repetitive and Restrictive Behaviours and Abnormal Patterns of Sensory Behaviours: A Systematic Literature Review

Abstract

Objectives. The DSM-V Repetitive and restricted behaviours (RRBs) category for diagnosing autism now includes abnormal sensory behaviours. Though, there appears to be little empirical evidence to suggest the two are inter-related. The aim of the review was to examine whether there is a relationship between repetitive and sensory behaviours in individuals with autism, and what factors influence this relationship.

Methods. Four electronic databases (Web of Science, Science Direct, PsycInfo and PubMed) were searched (November 2016 –January 2017). Search terms included; repetitive OR stereotyp* OR restrict* AND autism OR ASD OR Asperger* AND sensory OR processing OR auditory OR tactile OR visual OR vision OR percep* OR integration OR seeking OR avoiding OR hyper* OR hypo* OR pattern.

Results. Studies reviewed (n = 15) found significant relationships between different sensory and repetitive behaviours. Anxiety and intolerance of uncertainty influenced this relationship. The association is likely to be artificially inflated due to RRB measures measuring sensory behaviours and vice versa.

Conclusions. At best the review suggests a moderate, artificially inflated relationship between sensory and repetitive behaviours. There seems to be little evidence to indicate that sensory behaviours should be merged with RRBs in the DSM-V classification system.

Practitioner points

- When developing interventions to reduce RRBs and sensory behaviours it may be useful to address influencing factors of anxiety and intolerance of uncertainty.
- Researchers/clinicians should focus upon developing and validating a small number of sensory and RRB measures which do not include overlapping items
- Few longitudinal and experimental studies within this area exist. No studies have examined the relationship in adults diagnosed with autism.

Autism (ASD) is a pervasive developmental disorder recently characterised by the DSM-V (Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition American Psychiatric Association, 2013) as encompassing two categories of impairments, namely social and communication difficulties and restricted and repetitive behaviours (RRBs). The current review aims to investigate RRBs, which will be discussed in further detail below.

The DSM-V (APA, 2013) has categorised RRBs into four symptom types; 'B1) stereotypical/repetitive speech, motor movement or object use, B2) abnormal adherence to routines and excessive resistance to change, B3) highly restricted, fixated interests that are abnormal in intensity/focus and B4) hypo or hyper-responsiveness to sensory input or unusual interest in sensory aspects of the environment (p. 15). Researchers have split RRBs into at least two levels; lower order stereotypical motor behaviours (DSM-V B1 symptoms) and higher level cognitive behaviours relating to restricted interests, insistence on sameness, compulsions and rituals (Turner, 1999).

Whilst stereotypical behaviours are defined as a lack of goal oriented behaviours, insistence on sameness, restricted interests and rituals represent a ridged thinking style, often dictated by rule driven behaviour (Turner, 1999). Most interventions have focused on treating lower order stereotypical behaviours, using a range of behavioural interventions. In a review of the literature, Boyd, McDonough and Bodfish (2012) highlighted the need for more interventions to treat higher order RRBs. Chronological age has been shown to moderate the presence and severity of RRBs. Over time higher order RRBs increase, (Richler et al., 2010) whereas lower order RRBs either stay the same or decline (Kim & Lord, 2010).

The DSM-V RRB category now includes three patterns of abnormal sensory behaviours. Hyper-responsivity refers to an overly adverse reaction or avoidance of environmental stimuli (for example negative behavioural reactions to certain textures,

sounds or foods), whereas hypo-responsivity can be defined as a diminished, under-reaction to environmental stimuli (for example high pain threshold, indifference to sudden loud noises). Sensory-seeking is the third sensory pattern defined as excessively seeking out sensory input, resulting in highly focused interests (e.g. overly preoccupied with lights, movement, smelling or touching objects).

Sensory features were first noted in Kanner's (1943) descriptions of autism. However, the inclusion of abnormal patterns of sensory behaviours as a core feature of autism has long been disputed. Consequently, although sensory difficulties were included within the DSM-III (American Psychiatric Association, 1980), they have been absent in preceding editions until the DSM-V. The recent readmission coincides with an increasing amount of evidence suggesting abnormal sensory behaviours are prevalent in 60-95% of individuals with autism (Crane et al., 2009; Kern et al., 2007).

In a meta-analysis comparing between group sensory patterns, Ben-Sasson et al. (2009) found the ASD group showed higher numbers of sensory behaviours relative to the neurotypical group. Between group differences were highest for hypo-responsivity and lowest for sensory-seeking. They found chronological age moderated the development of sensory abnormalities (larger effect for those under 9 years).

Alternative conceptualisations of sensory processing patterns

In addition to DSM-V's categorisation, there are several other theoretical approaches to conceptualising abnormal sensory patterns. Dunn (1997) proposed a sensory processing model which suggests there are four sensory behaviour quadrants, falling along two dimensions, namely neurological threshold (the amount of input required for the nervous system to respond) and behavioural response. Sensory seeking is explained by a high neurological threshold combined with an active behavioural response whereas a high neurological threshold combined with a passive behavioural

response is referred as low registration (comparable to DSM-V's concept off hypo-responsivity). A low neurological threshold with active and passive behavioural responses are described as sensory avoiding and sensory sensitivity respectively. The latter two quadrants would be subsumed under the DSM-V's categorisation of hypersensitivity. Moreover, Miller et al. (2007) provides an alternative sensory processing model. They used the term sensory modulation disorder to define an abnormal response to sensory input. They propose three subtypes of sensory modulation disorder; Over-responsivity occurs when sensory information is processed more quickly or for a longer duration in either specific modalities or across sensory modalities. Under-responsivity suggests a lack of stimulation, resulting in behaviours aiming to increase stimulation. Finally, sensory-seeking refers to an intense craving for sensory input. These subtypes are similar to descriptions of hyper-responsivity, hypo-responsivity and sensory-seeking defined with the DSM-V.

Links between repetitive and sensory behaviours

Some authors have suggested that increased arousal causes hyper-responsivity. Subsequently, individuals develop compensatory strategies in the form of RRBs, to re-establish and regulate optimal arousal levels and soothe individuals (Liss, Saulnier, Fein & Kinsbourne, 2006; Zentall & Zentall, 1983). This is in accordance with the over-arousal theory of ASD first described by Hutt, Hutt, Lee & Ounsted (1964), who found a significant correlation between EEG activation and stereotypical behaviours. They argued individuals with ASD exhibited a reticular formation dysfunction (a brain area thought to maintain arousal levels), resulting in significantly high arousal levels. To prevent any more over-arousal, individuals with ASD exhibit an avoidance of new situations, instead presenting with an insistence on sameness. Liss et al. (2006) suggested sensory-seeking is an attempt to avoid unpleasant environmental input,

instead transforming it into a more controllable and pleasant form. This strategy is then repeated over and over. Additionally, Baker, Lane, Angley & Young (2007) suggest repetitive behaviours may be a strategy employed to increase stimulation in individuals with ASD who present with hypo-responsive and sensory-seeking behaviour.

Nonetheless Rimland (1964) and DesLauriers & Carlson (1969), proposed an under-arousal hypothesis. They suggested reticular formation dysfunctions can also be under-functioning, resulting in chronically low levels of arousal. They argued regardless of whether arousal levels are initially abnormally low or high, both dysfunctions result in limbic system suppression, leading to 'sensory deprivation'. Sensory deprivation (i.e. inability to receive sensory communications) negatively impacts upon the development of reward and affective circuits, resulting in behaviour which is repetitive and not goal oriented. Despite these latter arousal theories, empirical evidence and models to support these are lacking.

Rationale for current review

Given the lack of empirical evidence to support theories linking RRBs and sensory behaviours, it is surprising that they have been combined into the DSM-V category, implying the two are inter-related. This is some despite research suggesting RRBs and sensory behaviours are distinct, and have poor inter-correlations with one another (Tadevosyan-Leyfar et al., 2003). There has not been an extensive review of the literature examining studies which explore whether there is a relationship between the two. This is important because it has implications upon the current diagnostic framework and how sensory and repetitive behaviours are defined. If they are highly correlated, this may suggest that the current DSM-V framework is appropriate. Though, a lack of relationship would suggest that the two are distinct, and therefore should not be combined within the same DSM category. The relationship between sensory and

repetitive behaviours also has treatment implications. Current interventions to treat repetitive and sensory behaviours are relatively distinct from one another. Sensory Integration Therapy is a widely used treatment for sensory abnormalities, despite its lack of empirically supported evidence and lack of impact in reducing RRBs (Sniezyk & Zane, 2014). In contrast, there is some evidence for the effectiveness of behavioural interventions in treating RRBs, though this empirical base only relates to lower order stereotypical behaviours (Boyd et al., 2012). If repetitive and sensory behaviours are related, interventions may need to focus on both elements. Moreover, there could be cognitive processes mediating the relationship between sensory and repetitive behaviours, which may prove useful in developing future interventions.

Glod, Riby, Honey & Rodgers (2015) examined the relationship between sensory and repetitive behaviours. However, this exploration was part of a wider review, and only included four studies. They found stereotypy and insistence on sameness were associated with higher abnormalities in all three sensory patterns in individuals with ASD. Thus, the aim of the current systematic review was to expand upon Glod et al.'s (2015) findings to answer the following research questions;

1. Is there a relationship between sensory and repetitive behaviours in individuals diagnosed with ASD?
2. What other factors influence the relationship between sensory and repetitive behaviours?

Method

Search Method

Four electronic databases (Web of Science, Science Direct, PsycInfo and PubMed) were searched from November 2016–January 2017 to identify relevant studies. All years were included. The following search terms were used in combination;

repetitive OR stereotyp* OR restrict* AND autism OR ASD OR Asperger* AND sensory OR processing OR auditory OR tactile OR visual OR vision OR percep* OR integration OR seeking OR avoiding OR hyper* OR hypo* OR pattern. This identified 4,857 papers, of which 3,684 were duplicates and subsequently removed. Eight papers were identified through ancestry and citation searches, based upon all the relevant papers included in the current review. Citations and bibliographies from previous reviews by Glod et al. (2015) and Ben Sasson et al. (2009) were also searched. The remaining 1,181 papers titles and abstracts were screened based upon of their relevance to the research question. Seventy-seven full text papers were subsequently screened (title and abstracts) according to inclusion and exclusion criteria. The process of identifying and screening of papers is illustrated in Figure 1.

Inclusion criteria

Studies examining the relationship between sensory behaviours and RRBs were included if 1) individuals (either adults or children) had a diagnosis of an ASD 2) they included a measure of RRBs, either in the form of parent/caregiver questionnaires, standardised clinical assessment tools or clinician/researcher observations 3) they included a parent/caregiver reported measure of sensory behaviours and/or a paradigm measuring sensory behaviours 4) They explicitly reported an association between RRBs and sensory behaviours within the ASD group 5) were published in a peer reviewed journal and 6) were written in English.

Exclusion criteria

1) No inferential statistics used.

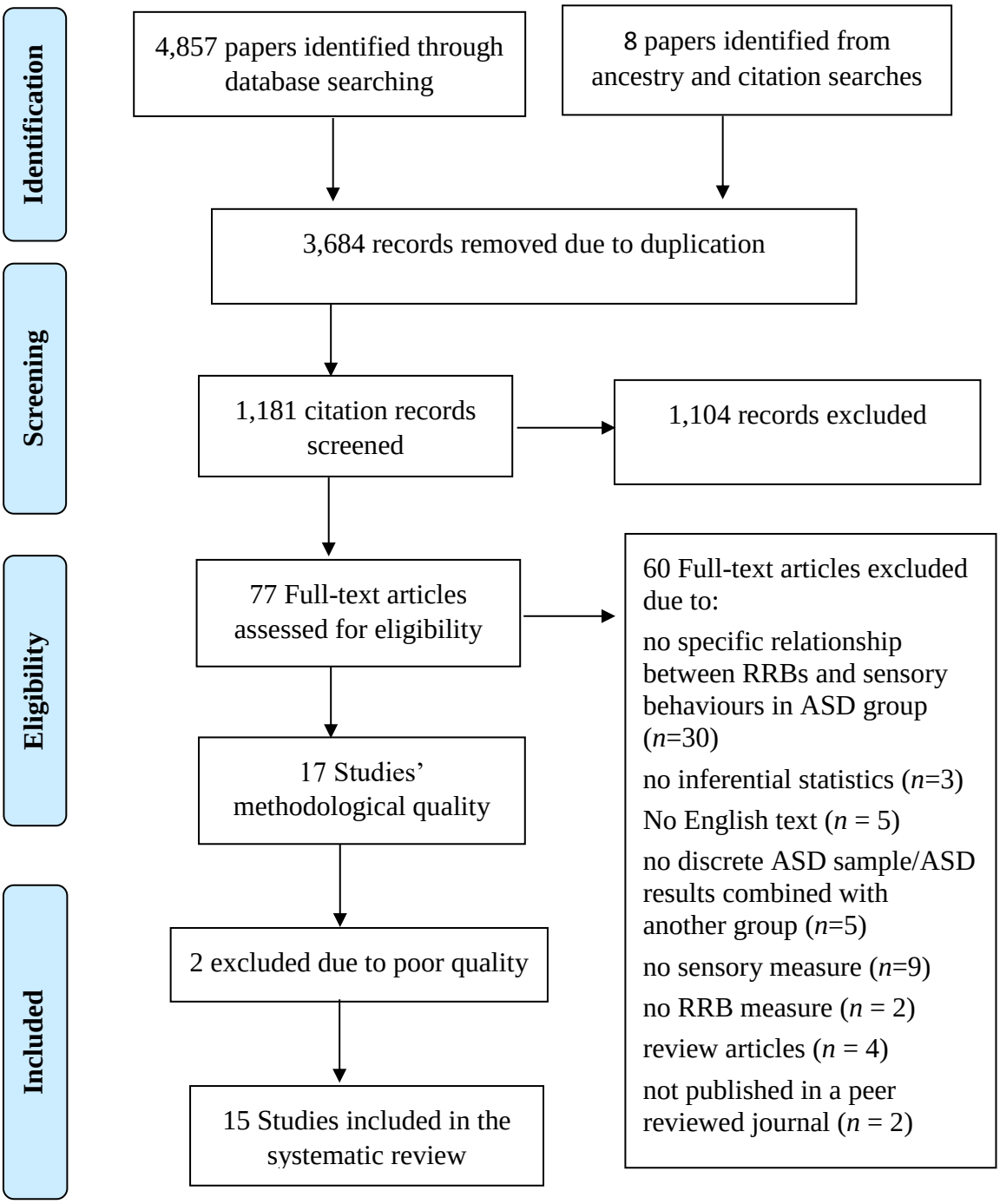

Figure 1. *Prisma flow diagram (adapted from Moher et al. 2010)*

Assessment of methodological quality

The methodological quality of each study initially selected for the review (*n*=17) was assessed using an adapted version of Downs and Blacks (1998) quality appraisal tool. To take into account cross-sectional and correlational studies several items were omitted. It was felt that even with items omitted it assessed a broad range of areas concerning methodological quality.

Correlational studies were assessed out of a possible 15 items and studies comparing two groups were assessed out of a possible 17 items (see Appendix A for items included). The scoring criteria for the final question relating to power was simplified according to Samoocha et al's., (2010) paper, awarding 1 point if the paper reported a sample size power calculation and 0 if not. All items were awarded either 1 point or a 0, except for item 5 which gave 1 point if they partially met or 2 if they fully met the criterion. To ensure studies included within the review were of acceptable methodological quality, the total score was converted into a percentage and those scoring <50% were rated as 'poor' (O'Connor et al., 2015). Two studies (Schauder et al., 2015; Mccormick et al., 2014) did not meet this threshold and were omitted from the review (see Appendix A for their quality appraisal scores). The remaining (*n*=15) studies met between 53%-82% of criteria (Table 1). A third of papers were scored by another researcher. An inter-rater Kappa reliability analysis showed very good agreement across raters, k=.84 (95% CI, .69, .99).

Methodological characteristics

Table 1 depicts the methodological and demographical characteristics of the studies included within the review. Thirteen (87%) of the studies were published within the last 10 years. All used a cross-sectional design. The overall sample included 1,744 participants, of which 1,225 had a diagnosis of ASD. The remaining 519 participants

were used as a comparison groups in 7 studies. The mean ages of participants were between 19 months and 15 years old. The proportion of males to females allocated to ASD groups was high in 13/15 studies (mean = 86% male). The mean number of males in comparison groups was 70%. Only 4 studies reported the ethnicity of participants. Of these, 3 included a high number of Caucasian participants (82-94%). Eight studies were conducted in the USA, 3 in the UK, 1 in Australia, 1 in Israel and 1 in Japan. One study recruited both USA and UK participants.

The sensory and RRB measures used within the current review are depicted in Table 2. Such a wide range of measures makes it difficult to interpret and compare findings across studies. Only 3 studies incorporated objective measures, namely the Tactile Defensiveness and Discrimination Test (TDDT, Baranek, 1993) and the Habituation to Tactile Stimuli Applied the Face (FACE-HAB, Baranek & Berkson, 1994), which both measured tactile hyper-responsiveness. One study (Joosten & Bundy, 2010) did not use a standardised measure, instead grouping participants into high RRBs based upon clinician and referrer observations, which is prone to subjectivity. Four studies used multiple measures to examine sensory behaviours.

Most studies used parent/caregiver reported measures of behaviour. Though, the questionnaires measure different constructs. For example, whilst the Repetitive Behaviour Scale-Revised (RBS-R, Bodfish & lewis, 2002) and the Repetitive Behaviour Questionnaire (RBQ, Turner, 1995) examined 6 and 4 RRB domains respectively, The Stereotyped and Self-Injurious Movement Interview (SSIMI, Turner, 1999b) only examined lower level stereotypical motor behaviours, whereas the Childhood Routines Inventory (CRI, Evans et al. 1997) examined higher level RRBs such as rituals and insistence on sameness. Similarly, whilst the Sensory Profile (SP, Dunn, 1999) included a range of subdomains, with researchers able to calculate hyper/hypo sensitivity and sensory seeking scores, the Touch Inventory for pre-

schoolers (TIP, Roveen, 1987) only examines hyper-responsivity in one domain, and the sensory questionnaire (SQ, Boyd & Baranek, 2005) only provides an overall score based upon 6 items. The Sensory Experiences Questionnaire (SEQ, Baranek et al., 2006) also examines a wide range of sensory modalities, but only calculates hypo/hyper responsivity scores, not sensory-seeking.

Moreover, using discrete measures of RRB and sensory behaviours to measure the relationship between the two is confounded by the fact that RRB measures also tend to measure sensory behaviours and vice versa. For example, The Autism Diagnostic Observation Schedule - RRB algorithm (ADOS, Lord, Rutter, DiLavore & Risi, 2001) incorporates unusual sensory interests and The Stereotyped Behaviour Checklist (SBC, Berkson, Gurermurh, & Baranek, 1995) found two unnamed factors which one could argue are hyper-responsiveness behaviours, such as experiencing washing and brushing teeth as uncomfortable and avoiding certain foods. Additionally, Challman et al. (2012) found a sensory factor deriving from the CRI. A sensory-repetitive motor behaviour factor was also identified on the RBQ. It could be argued this RBQ factor comprises both sensory-seeking and stereotypical behaviours.

Furthermore, some of the measures used have not been evaluated for reliability or validity (FACE-HAB, SQ, SBC). Others have found good psychometric properties but these have been developed using a non-ASD sample (CRI, Sensory Profile). Those with excellent or good psychometric properties include the ADOS-RRB, the RBQ, the RBS-R and the SEQ (See Appendix B for further critiques of studies included within the review).

Table 1. *Methodological and demographic characteristics of studies included within the review*

Study and Country	Sample size	Mean age (S.D)	Study design	Main Outcome measures	Quality rating (%)
Baranek, Foster & Berkson (1997) USA	29	10 years (1.9)	Cross-sectional design, within group (ASD) correlational study	RRB; SBQ Sensory; TIP FACE-HAB & TDDT	8/15 (53%)
Boyd, McBee, Holtzclaw, Baranek & Bodfish (2009) USA	ASD group; 61 Control; 64	*ASD*; 123 months (33.4) *Control*; 141 months (40)	Cross-sectional, mixed methods design. 2 groups – ASD and control	RRB; RBS-R Sensory; SQ Executive functioning; BRIEF[1]	11/17 (65%)
Boyd et al. (2010) USA	ASD*;* 67 Developmental Delay (DD); 42	*ASD:* 52 months (17.1) *DD:* 49 months (24.2)	Cross sectional design. 2 groups; ASD and DD	RRB; RBS-R Sensory; SEQ, Sensory profile & TDDT-R	12/17 (71%)
Chen, Rodgers & McConachie (2009) U.K	29	11 years 11 months (25.6)	Cross-sectional design, within group (ASD) correlational study	RRB; CRI Sensory; Short sensory profile	11/15 (73%)
Foss-Feig, Heacock & Cascio (2012) USA	34 (81.9 months; 10 months)	81.9 months (10)	Cross-sectional design, within group (ASD) correlational study	RRB: ADOS Sensory: SEQ, sensory profile and TDDT-R	10/15 (67%)
Gabriels et al. (2008) USA	70	10.8 years (4)	Cross-sectional design, within group (ASD) correlational study	RRB: RBS-R Sensory: Sensory profile	10/15 (67%)
Gal, Dyck & Passmore (2010) Israel	ASD; 98 Control; 44 Visual loss*;* 75 hearing loss*; 87*	ASD; 9.7 years (1.8) Control; 8.8 (1.6) Visual; 9.3 (1.7) Hearing; 9 (1.3)	Cross sectional 2 factor (group x DD) between group study. Within group differences measured	RRB: SSIMI Sensory: Short sensory profile (SSP)	9/15 (53%)
Green et al., (2016) U.K	ASD; 116 SEN; 72	ASD; 11.6 years (0.9) SEN; 12.7 (0.9)	Cross-sectional study. Between and within group differences examined	RRB; ICD-10 Sensory; SSP and ADI-R	11/17 (65%)

Table 1 (continued)

Study and Country	Sample size	Mean age (S.D)	Study design	Main Outcome measures	Quality rating (%)
Inada et al. (2015) Japan	ASD; 274 Atypical; 36	ASD; 15 years (6.3) Atypical; 13 (7.3)	Cross-sectional study. Between and within group differences examined	RRB; RBS-R Sensory: Sensory profile (SP)	11/17 (65%)
Johnson et al. (2014) USA	256	5.4 years (2.4)	Cross-sectional within group (ASD) correlational study	RRB; RBS-R sensory; SSP Eating; HEI[2]	11/15 (73%)
Joosten & Bundy (2010) Australia	ASD & ID; 29 ID; 23	ASD & ID; 9.5 years (unknown) ID; 9.7 (unknown)	Cross-sectional between group study	RRB; Groups selected due to high RRBs. Sensory; SP	11/17 (65%)
Lidstone et al. (2014) study 2 UK	49	10.7 years (3.1)	Cross-sectional within group correlational study	RRB; RBQ-2 Sensory; SP Anxiety; SCAS-P[3]	11/15 (73%)
Rodgers, Hepburn &Wehner (2003) USA	ASD; 26 Fragile X; 20 DD; 32 Control; 24	ASD, 33.3 months (3.6) Fragile X; 36.1 (8.1) DD; 33.3 (6.7) Control; 19.4 (4.8)	Cross-sectional study Between and within group differences examined	RRB; ADOS restrictive activities score Sensory; SSP	10/17 (59%)
Wiggins, Robins, Bakeman & Adamson (2009) USA	34 (34 months; unknown)	34 months (unknown)	Cross-sectional within group correlational study	RRB; ADOS Sensory; SSP	14/17 (82%)
Wigham, Rodgers, South, McConachie & Freeston (2015) UK and USA	53	12.5 years (2.3)	Cross-sectional correlational study	RRB; RBQ Sensory; SSP Anxiety; SCAS-P[3]	11/15 (73%)

Note: [1] The Behavior Rating Inventory of Executive Function (BRIEF, Giola et al., 2000); [2] The Healthy Eating Index (HEI, Guenther et al., 2006); [3]The Spence Anxiety Scales (SCAS-P, Spence, 1998).

Table 2. *Measures used within the review to assess RRBs and sensory behaviour*

Name (Author)	Type	Behaviours	Reliability and validity	*N* studies within current review used
Sensory behaviours				
TDDT (Baranek, 1993)	Objective assessment	Tactile hyper-responsivity - Light touch to different areas of body).	Acceptable inter-rater and test-re-test reliability Validated on ASD sample. No norms	3
The FACE-HAB, (Baranek & Berkson, 1994)	Objective assessment	Tactile hyper-responsivity	None reported	1
The TIP (Roveen, 1987)	Self-report	Tactile hyper-responsivity	Good internal consistency (.90) but validity questionable (Baranek & Berkson, 1994). No norms derived	1
Sensory Profile (SP, Dunn, 1999)	125-item Parent-report	Seven domains; tactile, movement, visual/auditory, taste/smell sensitivity, low energy, Auditory filtering and under-responsiveness/ seeks sensation.	Good convergent and discriminative validity Inter-rater validity varied between .47-.91 depending upon subscale.	6
Short Sensory Profile (Dunn, 1999)	caregiver-report	Same domains as SP but 38 items	Adequate internal consistency ranging from .68-.92 depending on subscale Good discriminative and convergent validity	6
The SQ (Boyd & Baranek, 2005)	Caregiver-report	6 items Provided a total sensory score.	None reported	1
The SEQ (Baranek et al. (2006)	Caregiver-report	Hyper and hyposensitivity in 5 domains; tactile, auditory, visual, vestibular-proprioceptive and gustatory-olfactory	Internal consistency .80 and excellent test-retest reliability (Little et al., (2011)	2

Table 2 (continued)

	Type	Behaviours Assessed	Reliability and validity	*N* studies within current review used
The Autism Diagnostic Interview- Revised (ADI-R, Rutter, LeCouteur & Lord, 2003)	Clinician-Interview with parents	3 items – 'unusual sensory interest, noise sensitivity and idiosyncratic response to sensory stimuli'	Authors found good inter-rater validity for all domains (>.80). Good sensitivity (.92) and specificity (.89). Esbensen et al., (2009) suggested it underreports RRBs	1
Repetitive, and restricted Behaviours				
The ADOS-RRB subscale (Lord, Rutter, DiLavore & Risi 2001)	Standardised Play-based assessment	0-8 points. RRB algorithm included repetitive play, unusual sensory interests, stereotypical body movements and restricted/stereotyped interests or behaviours	Good inter-rater reliability and test retest reliability (.59-.86). Poor diagnostic discrimination. Good predictive validity (90-97% sensitivity and 87-94% specificity.	3
Repetitive Behaviour Scale revised (RBS-R) (Bodfish & Lewis, 2002)	43-item Caregiver report	Six subscales; stereotyped, self-injurious, compulsive, sameness, restricted and routine behaviour	Lam & Aman (2007) found good internal consistency (.83) and inter-rater reliability (.60-.66).	5
Repetitive Behaviour Questionnaire (RBQ, Turner, 1995)	33-item caregiver report	33 items with 4 domains; repetitive language and movement sameness behaviour & circumscribed interests	Honey et al., 2012 found 2 reliable factors, sensory-repetitive motor behaviours (.79) and insistence on sameness/circumscribed interests (.85).	1
Repetitive Behaviour Questionnaire-2 (Leekam, et al., 2007)	20-item Caregiver report	Either 4 factor (same as RBQ) or 2 factors; motor-sensory and rigidity/routines/occupations	Arnott et al. (2010) found Fair-good internal consistency for all factors (.51-.82). Not validated on ASD sample.	1

Table 2 (continued)

Name (Author)	Type	Behaviours Assessed	Reliability and validity	*N* studies within current review used
The SSIMI (Turner, 1999)	32-item Clinician administered questionnaire	Examines stereotypical movements and object use and self-injury.	Not validated. Internal consistency .76 (Gal et al., 2009).	1
The CRI (Evans et al. 1997)	19-item Caregiver report	Examines routine adherence and need for sameness. 3 factor structures; 'just right', repetitive behaviours and sensory sensitivities (Challman et al., 2012)	Constructed with neurotypical children. Excellent internal consistency (.80) and test-retest reliability (.90). Moderate construct validity (Systma, Kelley, Wymer, 2001)	1
The SBC (Berkson, Gurermurh & Baranek,1995)	54-item Caregiver report	8 factors; rigidity/sameness, auditory/repetitive vocalisations, visual orientation, music-motor behaviours, object stereotypy and abnormal focused attention. Two unnamed factors - items included 'only eating certain foods, bothered by tooth brushing & washing'	None reported	1
ICD-10 RRB symptom count (World Health Organisation, 1992)	Clinician assessment tool	Examines preoccupation with object parts, stereotyped language and repetitive behaviour, routines and rituals	Limited evidence of reliability and validity of ICD-10. Ability to discriminate between ASD and Asperger's diagnosis (Woodbury-Smith, Klin & Voklmar, 2005)	1

Results

Sensory behaviour and RRBs

Nine studies reported significant correlations between RRBs and an overall sensory behaviour score (Table 3). Five studies (Boyd et al., 2009; Chen et al., 2009; Gabriels et al., 2008; Johnston et al., 2014; Rogers et al., 2003) found significant moderate effect sizes, ranging from .42 to .53. Two studies (Inada et al., 2015; Wiggins et al., 2009) found significant strong effect sizes (r = .62 and .63 respectively). In contrast, Green et al. (2016) and Gal et al. (2010) found significant but small correlations between RRBs and sensory behaviours total score (r = .045-.28). Differences may be due to the type of measure used. Studies which found small correlations used RRB measures which show poor or unmeasured reliability and validity (ICD-10 and SSMI). In contrast, most of the studies which found moderate-strong relationships used the RBS-R (*n* =4) or the ADOS (*n* =2). Moreover, Gal et al. (2010) only measured stereotypical movements whereas the other studies examined several RRBs.

Table 3. Relationship between overall sensory behaviour scores and RRB scores

Study	**Measures**	Analysis	Result
Boyd et al. (2009)	RBS-R and SQ	Correlation and regression	SQ total moderately correlated with RBS-total score (r = .43, p <.001), stereotypy (r = .41, p = .01) and compulsion (r = .34, p =.05).
Chen et al. (2009	CRI and SSP	Correlation	Significant correlation between increased number of repetitive behaviours and sensory abnormalities (r = -.42, p =.02).
Gabriels et al. (2008)	RBS-R & SP	Correlation	Sensory behaviours and RRBs significantly correlated after controlling for age and intelligence (r = .53, p = .001).
Gal et al. (2010)	SSIMI & SSP	1 way (group) ANCOVA controlling for DD	ASD group showed small correlation between stereotypical movements and total sensory behaviour score (r=-.28, p .05),
Green et al. (2016)	ICD-10 & SSP	Regression	Increased sensory abnormalities significantly correlated with increased RRBs (t = 2.18, p = .05) b = -5.49, eta squared .045)
Inada et al. (2015)	RBS-R & SP	Correlation	RBS-R and SP total score significantly correlated (r=.62, p<.001).
Johnston et al. (2014)	RBS-R SSP	Correlation	Repetitive and sensory abnormalities significantly correlated (r = -.53, p<.001).
Rodgers et al. (2003)	ADOS RRB score & SSP	1 way (group) ANOVA	Repetitive and sensory behaviours moderately correlated for autism group only (r=.43, p<.05).
Wiggins et al. (2009)	ADOS & SSP	Correlation	Significant association between SSP and ADOS stereotyped interests and behaviours (r = .63, p < .01).

Hyper-responsiveness and RRBs

Nine studies (Table 4) examined the relationship between hyper-responsiveness and RRBs. No significant relationships were found when studies used the TDDT as a measure of tactile hyper-responsivity (Boyd et al., 2010; Foss-Feig et al., 2012). The remaining studies all found significant relationships between hyper-responsiveness and repetitive behaviours (r = .31-.78). The most consistent finding was a significant moderate association between hyper-responsiveness and insistence on sameness (r =.36-.56), found in 4 studies (Baranek et al., 1997; Lidstone et al., 2014, Boyd et al., 2010; Wigham et al., 2015). Findings were mixed when exploring the relationship between

hyper-responsiveness and stereotyped behaviour. Baranek et al. (1997) found no relationship between the two variables. However, 3 studies (Gal et al., 2010; Boyd et al., 2010; Wigham et al., 2015) found significant correlations (r=.31-.39). One reason for this discrepancy may be due to the type of measures used. Baranek et al. (1997) used objective measures in only one sensory domain. They also used a smaller sample size (*N* = 29) and an RRB measure with unestablished psychometric properties.

Only 3 studies examined the relationship between repetitive behaviours and specific sensory modalities (Gal et al., 2010; Baranek et al. 1997 and Chen et al., 2009). Within these studies an association between repetitive behaviours was found across several sensory domains, including visual/auditory (Chen et al., 2009; Gal et al., 2010) and tactile (Chen et al., 2009; Baranek et al., 1997). However, a critique of this research is that two of the latter studies combined auditory and visual modalities into one score, which may mask subtle differences between the two modalities. Moroever, Baranek et al (1997) only examined one modality and so cannot make comparisons between the strength of the relationship between tactile sensory abnormalities and RRBs in comparison to other sensory modalities.

Table 4. *Relationship between hyper-responsiveness and RRBs*

Study	**Measures**	Analysis	Result
Baranek et al. 1997)	SBQ, TIPFACE-HAB & TDDT	Correlation	Tactile hyper-responsiveness, significantly associated with rigidity/sameness, repetitive auditory vocalisation and unusual/narrow interests on FACE-HAB (r = .50,.56 and .48) and TIP (r =.39,.45 & .55).
Boyd et al. (2010)	RBS-R & sensory score amalgamated from SEQ, SP & TDDT-R	Correlation	No relationship with object/motor stereotypies Hyper-responsiveness positively associated with RBS subscales stereotypy, compulsions and rituals/sameness ($p<0.01$).
Chen et al. (2009)	CRI and SSP	Correlation	Significant relationship between hyper-responsivity and repetitive behaviours (r = .72-.78, $p<.05$). Tactile sensitivity significantly associated with CRI frequency (r = .73, $p <.05$).
Foss Feig et al. (2012)	ADOS, SEQ, sensory profile & TDDT-R	Correlation	No relationship
Gal et al. (2010)	SSIMI & SSP	Correlation	ASD group showed significant correlation between stereotyped movements and visual-auditory hyper-responsivity (r = .31, $p < .05$)
Inada et al. (2015)	RBS-R & SP	Correlation	Significant correlations between RBS-R, sensory sensitivity (r = -.57, $p < 0.001$) and sensation avoidance (r = .62, $p < .001$).
Joosten & Bundy (2010)	SP	Independent t-tests	ASD group showed increased sensory sensitivity (t = 2.5, $p <0.01$, d = 0.70) and sensation avoidance (t = -1.7, $p<.05$, d =.47) compared to ID group.
Lidstone et al. (2014)	RBQ-2 & SP	Correlation	Repetitive motor behaviours correlated with sensory avoidance (r=.42). Insistence on sameness correlated with sensory sensitivity (r=.49) and avoidance (r=.43, all $p<.001$).
Wigham et al. (2015)	RBQ & SSP	Correlation	Sensory hyper-responsiveness significantly correlated with insistence on sameness (r = -.56, $p <.001$) and repetitive motor subscales (r= -.39, $p <.01$).

Hypo-responsiveness and RRBs

Eight studies examined the relationship between hypo-responsiveness and RRBs. Three studies (Chen et al., 2009; Boyd et al., 2010; Joosten & Bundy, 2010) found no significant relationship between the latter variables. Foss Feig et al., (2012) found tactile hyposensitivity was significantly associated with the ADOS, but not ADI-

R RRB subscales. This difference suggests the type of RRB measure used influences the results. Hus, Gotham & Lord, (2014) argue given repetitive behaviours can be time and context dependant, the ADOS (a one-off assessment) may not capture the true amount of repetitive behaviours conducted in every-day life.

Nonetheless, four studies (Gal et al., 2010; Inada et al., 2015; Wigham et al., 2015; Lidstone et al., 2015) found significant associations between hypo-responsivity and RRBs (r = .36-.70). In examining specific repetitive behaviours, Wigham et al. (2015) and Gal et al. (2010) found stereotyped motor behaviour was predicted by hypo-responsivity (r =.70 and .43), though Lidstone et al. (2015) did not. Lidstone et al. (2015) used the RBQ-2 which has not yet been validated on an ASD population and includes significantly fewer stereotyped motor behaviour items compared to the older version used by Wigham et al. (2015), which has been extensively validated within an ASD population. However, both Lidstone et al. (2015) and Wigham et al. (2015) found insistence on sameness was correlated with hypo-responsivity (r =.38 and -.36 respectively).

No studies examined hypo-responsiveness in terms of specific sensory modalities. Instead all sensory modalities are combined into one score. Implications of this approach are discussed within the discussion.

Table 5. *Relationship between hypo-responsiveness and RRBs*

Study	**Measures**	Analysis	Result
Boyd et al. (2010)	RBS-R & sensory score amalgamated from SEQ, SP & TDDT-R	Correlation	No relationship
Chen et al. (2009)	CRI and SSP	Correlation	No relationship
Foss Feig et al. (2012)	ADOS, SEQ & TDDT-R	Correlation	Tactile hypo-responsiveness significantly correlated with ADI-R (r = .38, p = .05) but not ADOS
Gal et al. (2010)	SSIMI & SSP	Correlation and multiple regression	Significant correlation between stereotyped movements and hypo-responsiveness (r=.43, p <.01) Hypo-responsiveness predicted stereotyped movements in ASD group (r^2 = .17, F= 1,54 = 12.76, p < .001)
Inada et al. (2015)	RBS-R & SP	Correlation	RBS-R and hypo-responsiveness significantly correlated (r= .39, p < .001),
Joosten & Bundy (2010)	SP	Independent t-tests	No relationship
Lidstone et al. (2014)	RBQ-2 & SP	Correlation	Significant correlation between hypo-responsiveness and insistence on sameness (r=.38, p<.001) but not repetitive motor behaviours
Wigham et al. (2015)	RBQ & SSP	Correlation	Sensory hypo-responsiveness significantly correlated with insistence on sameness (r =-.36, p<.01) and repetitive motor RRB subscales (r = -.70, p<.001)

Sensory-seeking and RRBs

Five studies examined the relationship between sensory-seeking and repetitive behaviours. Joosten & Bundy (2010) compared two groups of children displaying high RRBs, those with both ASD and ID and those with ID only. They and found no significant group for sensory-seeking Joosten & Bundy (2010) found no significant differences between high and low RRB groups for sensory-seeking. However, four studies (Boyd et al, 2010; Foss Feig, 2012; Inada, 2015; Lidstone et al., 2014) found significant associations

between the two variables (r =.39-.63). In terms of specific repetitive behaviours, both Lidstone et al. (2014) and Boyd et al. (2010) found a relationship between sensory-seeking and insistence on sameness (r = .49, p < .01 and *b* = 5.92, p < .046 respectively). However, only Lidstone et al. (2014) found sensory-seeking significantly correlated with stereotypical motor behaviours. Lidstone et al. (2014) and Boyd et al. (2010) used different RRB and Sensory measures which may account for differences. One strength of Boyd et al. (2010) is that they used several sensory measures to calculate a sensory seeking score and omitted sensory items comparable to RRBs, indicating a more accurate measure of sensory-seeking. Only one study (Foss-Feig, 2012) examined sensory-seeking within a specific sensory modality. Nonetheless, whilst they examined tactile sensory-seeking, they did not examine any other sensory modality. Implications of this are explored within the discussion.

Table 6. *Relationship between sensory seeking and RRBs*

Study	**Measures**	Analysis	Result
Boyd et al. (2010)	RBS-R & sensory score amalgamated from SEQ, SP & TDDT-R	Regression	Sensory-seeking predicted insistence on sameness (b = 6.49, p<.05) not repetitive motor behaviours.
Foss Feig (2012)	ADOS, SEQ & TDDT-R	Correlation	Tactile sensory-seeking and repetitive behaviours significantly correlated (r=.36, p = .05)
Inada et al. (2015)	RBS-R & SP	Correlation	RBS-R significantly associated with SP sensory-seeking (r = -.63, p< .001),
Joosten & Bundy (2010)	SP	Independent t-test	No significant difference between high and low RRB groups for sensory-seeking.
Lidstone et al. (2014)	RBQ-2 & SP	Correlation	Sensory-seeking significantly correlated with repetitive motor behaviour (r =42, p<.001) and insistence on sameness (r =.49, p <.001).

Factors influencing the relationship between sensory behaviours and RRBs

Anxiety. Three studies (Table 7) examined the impact of anxiety on the relationship between sensory and repetitive behaviours (Lidstone et al., 2014; Joosten and Bundy, 2010; Wigham et al., 2015). Lidstone et al. (2014) found whilst hypo-responsivity and sensory-seeking were significantly associated with anxiety and insistence on sameness, when the latter sensory behaviours were controlled for, there was no impact upon the relationship between anxiety and insistence on sameness. In contrast, the relationship between insistence on sameness and anxiety vanished when hyper-responsivity was partialled out, suggesting hyper-responsivity mediates the relationship between anxiety and instance on sameness.

Moreover, Wigham et al. (2015) found anxiety mediated relationships between hypo/hyper responsiveness and repetitive motor behaviours and insistence on sameness. Whilst Joosten and Bundy (2010) did not explicitly examine mediating effects, in a previous study Joosten, Bundy, & Einfeld, (2009) found significant levels of anxiety within the ASD group reduced when conducting stereotypical behaviours. In the current study, Joosten & Bundy (2010) found the same ASD group showed increased hyper-responsivity. This may suggest hyper-responsivity behaviours in ASD increase anxiety, resulting in individuals employing strategies such as increasing repetitive behaviours to off-set anxiety.

Impact of demographic factors. Four studies found a significant relationship remained between repetitive and sensory behaviours when age was either partialled out or entered a predictor, suggesting that age is not an influencing factor. Additionally, Joosten and Bundy (2010) controlled for intelligence between the two high RRB groups suggesting this does not impact upon group differences relating to sensory behaviours. However, Gal et al., (2010) found whilst intellectual disability does not increase

stereotypical movements per se, an interaction effect exists between intellectual disability, autism and abnormal sensory behaviours to increase the prevalence of stereotypical movements. Given that Joosten & Bundy (2010) split groups into high and low RRBs based upon referrer information, it may be that their RRB criteria was too broad to detect an effect. Alternatively, intelligence may influence only particular types of RRBs; stereotypical movements.

Impact of cognitive and clinical factors. Three studies examined the impact of specific cognitive factors. Intolerance of uncertainly (IoU) is defined as 'the tendancy to react negatively on an emotional, cognitive and behavioural level to uncertain situations and events' (Dugas & Koerner, 2005, p. 62). Wigham et al., (2015) found IoU mediated relationships between hyper-responsiveness, insistence on sameness and repetitive motor behaviours. This was also found for hypo-responsiveness. However, executive functioning (Boyd et al., 2009) and a weak central coherence cognitive style, which refers to a reduced ability to integrate a perceptual whole whilst displaying intact or superior ability to focus upon detail (Chen et al., 2009) did not explain the relationship between repetitive and sensory behaviours. Additionally, Johnston et al. (2014) found that sensory difficulties and increased repetitive behaviours both predicted feeding difficulties in children with autism (Table 7).

Group comparisons. Three studies compared the relationship between sensory and repetitive behaviours within both the ASD and comparison groups. Two studies (Gal et al, 2010; Boyd et al., 2010) found a relationship between the sensory and RRBs exists across different clinical groups, but the presentation differed depending on diagnostic group. For example, Gal et al., (2010) found stereotypical movements were significantly associated with hypo-responsivity within the ASD group, but hyper-responsivity for visually impaired and typically developing groups. In both studies, the

autism group showed significantly higher sensory and repetitive behavioural scores. However, Rodgers (2003) found no significant differences between autism and fragile X groups for sensory abnormalities, but the ASD group exhibited higher RRBs.

Table 7. *Anxiety and cognitive factors influencing relationship between sensory behaviours and RRBs*

Study	Analysis	Result
Anxiety and Intolerance of Uncertainty		
Joosten & Bundy, 2010	T-test	ASD group significantly higher sensory avoidance (r=.47, .05) and sensitivity (.70, p<.001) compared to ID group. Same group had higher levels of anxiety, reducing when engaged in stereotypical behaviour.
Lidstone et al. (2014)	Mediation model	Anxiety predicted hyper-responsiveness (F,1,47) = 28.17, R^2=.36, p<.001) and insistence on sameness (F (1,47) =12, R^2=.20, p<0.001). Non-significant when hyper-responsiveness partialled out. Association between insistence on sameness and anxiety remained (r=.37, p<.01) when sensory seeking and hypo-responsiveness partialled out.
Wigham et al. (2015)	Mediation model	Anxiety and intolerance of uncertainty mediated relationships between hypo-responsiveness and Insistence on sameness (B = .16; LL = -.34, UL = .04) and repetitive motor behaviours (B = .09: LL = .22, UL = .01) Anxiety and intolerance of uncertainty mediated relationships between hyper-responsiveness and insistence on sameness (*B* = .07; LL = -.15, UL = .01) and repetitive motor behaviours (*B* = .05: LL = .11, UL = .01).
Other cognitive/Clinical Factors		
Johnson et al. (2014)	Correlation	Eating behaviours correlated with RRBs (r =.60, p<0.001) and sensory behaviours .48, p < .001)
Chen et al. (2009)	Correlation	In-depth processing style moderately correlated with RRB total score (r=.61; p<.001) but not sensory abnormalities
Boyd et al. (2009)	Correlation	Executive functioning (BRIEF) moderately correlated with RBS-R (r=.43, p< .001) but not SQ.

Discussion

The relationship between sensory and repetitive behaviours

The first aim of the review was to explore whether there is a relationship between sensory and repetitive behaviours in individuals with ASD. Similar to Glod et al. (2015) the current review found all studies reported an association between repetitive behaviours and sensory behaviours total score. Overall, studies suggest a moderate relationship exists between sensory and repetitive behaviour total scores. Additionally, several studies found a significant association between repetitive behaviours and at least one of the sensory patterns defined by DSM-V. Given all studies found a significant result, research within this area could be subject to publication bias.

The current study found studies examining hypo-responsivity were most mixed when examining its relation to repetitive behaviours, with some finding no association (n = 4) and others (n=5) finding a moderate to large effect size. In contrast, most studies examining the relationship between hyper-responsivity and repetitive behaviours found significant correlations. Two studies which did not find an association only examined the tactile sensory modality. This may suggest that the relationship may be stronger or weaker depending upon the sensory modality explored. Most studies examining sensory-seeking and repetitive behaviours found a significant moderate correlation between the two.

The most robust finding from multiple studies was that insistence on sameness significantly moderately correlated with all sensory behaviours (r =.36-.56). Whereas, mixed findings for an association between stereotyped movements and sensory behaviours were highlighted. Thus, it could be argued that higher order RRBs are more likely to be associated with sensory abnormalities. Although of note, there were more studies which found an association between lower level stereotypies and sensory behaviours (n =6) than those who did not (n=3), with some studies reporting a

significantly strong association. One reason for the mixed findings may be due to the vast array of RRB measures used to examine the same constructs. Leekham, Prior & Uljarevic (2011) argue there is still a lack of agreement for the definition of RRBs, which makes it difficult to develop reliable and valid measures. RRBs have been weighted differently depending on the measure, and different tools measure RRBs in a variety of ways. For example, some focus upon the frequency of behaviours, whereas others examine intensity or distress caused (Honey et al., 2012).

The finding that insistence on sameness is related to sensory behavioural patterns may give some weight to Hutt et al.'s (1964) hyper-arousal theory that individuals present with an insistence on sameness to block becoming overwhelmed by any more environmental stimuli. However, it is still unclear how this explains the association with hypo-responsivity and sensory-seeking. The latter studies within the review do not provide clear evidence as to whether the relationship between sensory and repetitive behaviours are related to over-arousal (Hutt, Hutt, Lee & Ounsted, 1964) or under-arousal (Rimland, 1964; DesLauriers & Carlson, 1969) in the brain. Future research will require the inclusion of an objective measure of arousal (for example electrodermal techniques or brain imaging within the reticular formation/other brain areas associated with arousal control), examining its impact upon behavioural manifestations of arousal and its link to repetitive behaviours.

Factors influencing the relationship between sensory and RRBs

The second aim was to examine what factors influence the relationship between sensory and repetitive behaviours. Findings from the review may support the notion that repetitive behaviours are employed as compensatory behaviours to reduce high levels of arousal (Liss et al., 2014). Specifically, some studies suggest high arousal levels may present as increased anxiety, causing hyper-responsiveness but not sensory-seeking or

hypo-responsiveness. (Joosten and Bundy, 2010; Lidstone et al., 2014). A theoretical implication of the latter findings may be that the three sensory patterns have different rather than shared underlying mechanisms.

Age did not appear to be an influencing factor in the relationship between sensory and repetitive behaviours. Though, this finding needs to be interpreted cautiously as studies in the current review use narrow age ranges within each study. For example, whilst Rogers et al. (2003) and Wiggins et al. (2009) recruited children under 5, Wigham et al. (2015) and Inada et al's. (2015) sample consisted of teenagers. It may be that age changes the relationship between sensory and repetitive behaviours over time. Indeed, longitudinal studies have found that lower level RRBs are more frequent in younger children and higher order RRBs (e.g rituals) increase with age (Kim & Lord, 2010). The studies included within the review are all cross-sectional and so cannot examine this. Future studies should develop longitudinal designs to examine whether the relationship between repetitive and sensory behaviours changes over time and include a range of ages.

Moreover, results suggest that cognitive processing style and executive functioning are not the shared mechanisms underlying sensory and repetitive behaviours. Nonetheless, Chen et al. (2009) only examined whether a detailed cognitive processing style predicted sensory abnormalities total score. If one were to take the view that hyper-responsivity, hypo-responsivity and sensory-seeking have distinct rather than shared underlying mechanisms, then combining all scores together may be masking any relationships between specific sensory patterns and specific cognitive variables. Nonetheless, intolerance of uncertainty was shown to mediate a relationship between sensory and repetitive behaviours, which may have important intervention implications.

Methodological critique

One of the main critiques of the studies included within the review is that most have relied upon parent/caregiver reported questionnaires, which lends itself to bias. Whilst parent rated questionnaires can provide detailed information across time and contexts from someone who knows the child extremely well (Rogers et al, 2003), Hoyle et al (2001) suggest their retrospective nature increases the risk of erroneous reporting and recollection bias. Moreover, parental stress (Ooi et al., 2016) and parent's knowledge about symptoms of ASD (Dahlgren and Gillberg, 1989) influences reporting. Schauder & Bennetto (2016) argue that more objective measurements should be used to assess sensory behaviours that can be easily missed using questionnaires (for example an absence of behaviours in hypo-responsivity or an abnormally high sensory threshold). Additionally, many studies use the Sensory Profile or Short Sensory Profile to assess sensory behaviours. However, the face validity of the questionnaire in examining sensory symptoms has been questioned. Green et al. (2016) question how 'weak grasp' relates to sensory behaviours and highlight that frequent hypo-sensitivities such as a high threshold to pain and temperature are absent in the Short Sensory Profile. Additionally, sensory items may overlap with repetitive items and vice versa, potentially artificially inflating the relationship. For example, the Sensory Profile includes the item 'is distressed by change in routines'. Moreover, sensory items may not fit into one discrete sensory pattern but several. Only three studies within the review (Boyd et al., 2010; Gabriels et al., 2008; Lidstone et al., 2014) removed overlapping items. Nonetheless, both found a significant relationship after excluding items.

A further difficulty is that sensory processing literature uses a range of interchanging terminology, which can be confusing. For example, hyper-responsiveness is used interchangeably with sensory responsivity, hyper-sensitivity, low threshold, tactile/oral/visual defensiveness, enhanced perception and over-sensitivity (Schaaf &

Lane, 2015). Such a wide variation of terms makes it difficult to comprehend the sensory literature**.**

Additionally, only a limited number of studies within the review included ethnicity of participants. Of those, three overrepresented Caucasian participants, which may confound results. Moreover, males were over-represented in all studies except one. Although a larger percent of males was expected, given they are five times more likely to be diagnosed (US centre for disease prevention, 2012), 11 were above the 80% expected if one were to account for the ratio difference in research. Additionally, the ratio of males was lower in comparison groups. Moreover, none of the studies included adults, despite inclusion within the reviews criteria. This highlights the paucity of research examining links between repetitive and sensory behaviours within the adult ASD population. These potential confounding variables of gender, age and ethnicity therefore limit the generalisability of findings. Furthermore, none of the studies included a power analysis and many used a small sample size. Thus, studies may not have been sufficiently powered to find significant effects. This may explain for example why Chen et al. (2009), Joosten and Bundy (2010) and Boyd et al. (2009) did not find any associations between hypo-responsivity and repetitive behaviours, despite Ben-Sasson et al. (2009) suggesting that this is more prevalent in individuals with ASD.

Additionally, most of the studies included within the review adopted a profile approach that combined sensory modalities into an overall hyper-responsivity, hypo-responsivity or sensory-seeking score. Moreover, 7/15 of the studies included only examined the relationship between RRBs and sensory behaviours using an overall total sensory score. Some studies whichich did examine specific modalities still comined some of the sensory modalities together (for example Chen et al. 2009 and Gal et al. 2010 both combined visual and auditory hyper-responsivity difficulties into one score) Implications of not examining specific modalities is that interventions could be

implemented on the basis of a generic hypo/hyper score, without acknowledging the fact that an individual may be hypo-responsive to sound but hyper-responsive to touch and vision for example. Thus, adopting a generic intervention to reduce hyperresponsivity without examining specific modalities is may be extremely detrimental. Future research must therefore examine the relationship within specific modalities so that the intervention can be tailored to one's individual profile, rather than looking at sensory abnormalities generically.

A strength of some studies was the use of multiple measures and methods, increasing validity and reliability. Future studies should aim to include objective data, through observational methods, brain imaging techniques or paradigms designed to assess sensory behaviours. An additional strength of the review is that it excluded studies considered to have poor methodological quality. However, the review methodology can be criticised in terms of the search terms used. Specifically, Autism was used instead of autis* which would have allowed for the word 'autistic' to be searched. This may have provided additional relevant papers. Moreover, experts within the field were not contacted to ensure the author had not missed any relevant papers or papers due to be published. Additionally, the author did not search databases for unpublished work, which would have reduced the likelihood of a publication bias. Thus, in future the author will search unpublished databases such as ProQest and EthOS when conducting a systematic review.

Clinical implications

Given the reviews findings of influencing factors, it may be useful for psychological interventions to address anxiety, insistence on sameness and intolerance of uncertainty when treating ASD individuals who exhibit high levels of sensory abnormalities and RRBs. Boyd et al. (2012) concluded no effective interventions

currently exist for treating higher-order RRBs. Similar conclusions have been made for sensory interventions and their impact upon RRBs (Sniezyk & Zane, 2014). It could be argued they are ineffective because they do not target the underlying mechanisms associated with these behaviours.

Given the reviews findings that individuals with ASD show increased levels of sensory behaviours compared to controls (with exception of Fragile X) it may be useful to for sensory questionnaires to develop clinical cut off scores for ASD. As individuals with ASD may show all sensory patterns, this cut off score may only be reliable for a total sensory score. Sensory questionnaires should be developed which include sensory symptoms commonly and specifically reported by individuals with ASD.

Additionally, findings suggest at best a moderate relationship between sensory and repetitive behaviours, which is likely to be conflated by many measures assessing both sensory behaviours and RRBs. It could therefore be argued that they should not be included within the RRB domain. Research has also found a correlation between sensory and social and communication difficulties (Glod et al., 2015). Thus, it remains unclear why they are classified under RRBs. The current review supports an argument proposed by Lord & Jones (2012), who suggest rather than concentrating on ASD as either categorical or dimensional, it would be more useful to reflect upon how dimensions can predict particular behavioural manifestations, in order to develop suitable interventions to modify these. It may therefore be useful to explore whether particular ASD subgroups exist which fall along different dimensions (e.g. higher order RRBs with increased hyper-responsivity) as it is unlikely that there will be a 'one size fits all' intervention to meet the needs of such a heterogeneous diagnosis.

More evidence is required to examine whether there would be merit in sensory behaviours having their own subcategory within future classification systems. In doing so, it is clear from the review that researchers need to develop measures assessing each

construct individually. This may be more difficult for sensory-seeking since its definition (e.g. preoccupation with smelling and movement) appears intrinsically intertwined with lower order RRBs.

Finally, the findings suggest there may be a higher risk of eating difficulties in children presenting with high levels of sensory and repetitive behaviours. It may be useful for clinicians to assess eating behaviours in those displaying high levels of repetitive and sensory behaviours and evaluate the value of providing early intervention to offset future eating difficulties. Nonetheless, most studies were correlational and so cannot make assumptions about causality. Results should therefore be interpreted cautiously. More experimental studies and mediation models are required within this field.

Conclusions

The review suggests a moderate relationship between repetitive and sensory behaviours, though no integrated model exists to explain this association. Underlying mechanisms that may drive and mediate the relationship require further exploration, as does the relationship between RRBs and specific sensory modalities. As this moderate association is likely to be artificially inflated, it will be beneficial for future studies to focus upon using a small number of valid and reliable measures which do not include overlapping sensory and RRB items, specifically developed for individuals with ASD. Other direct methods (e.g. brain imaging) are required to substantiate theories linking RRB and sensory behaviours. Until this is empirically supported, there seems no valid evidence to indicate that sensory behaviours should be merged with RRBs in the DSM-V classification system. There is a paucity of longitudinal and experimental studies within this area, which is compounded by the lack of research conducted within the

adult ASD population as well as the wide range of measures and language used to describe repetitive and sensory behaviours.

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Appendix A. *Down and Blacks Quality Appraisa*

Author (Date)	1.Hypothesis/aim clearly described?	2. Outcomes clearly described	3.Participant characteristics	5. Description of confounding variables?	6. Findings clearly described?	7. Estimates of random variability provided?	10. Probability values reported?	11. Representative of source population?	12. Representative of recruited participants?	16. Any data dredging?	18. Statistical tests appropriate?	20. Outcome measures accurate?	21. Groups recruited From same population	22. Groups recruited At same time?	25.Adjusted for confounding	27. Conducted power analysis	Total score (%)
Baranek et al., (1997)	1	1	0	1	0	1	0	0	0	1	1	1	N/A	N/A	1	0	53%
Boyd et al., (2009)	1	1	1	1	1	1	1	0	0	1	1	1	1	0	0	0	65%
Boyd et al., (2010)	1	1	1	0	1	1	1	1	0	1	1	1	1	0	1	0	71%
Chen et al., (2009)	1	1	1	1	1	1	1	0	0	1	1	1	N/A	N/A	1	0	73%
Foss Feig at al., (2015)	1	1	1	1	1	1	1	0	0	1	1	1	N/A	N/A	0	0	67%
Gabriels et al., (2008)	1	1	1	1	1	0	1	0	0	1	1	1	N/A	N/A	1	0	67%
Gal et al., (2010)	1	1	1	1	1	1	0	0	0	1	1	1	0	0	0	0	53%
Green et al., (2010)	1	1	1	0	1	1	1	1	0	1	1	1	1	0	0	0	65%

Appendix A. (continued)

Author (Date)	1.Hypothesis/aim clearly described?	2. Outcomes clearly described	3.Participant characteristics	5. Description of confounding variables?	6. Findings clearly described?	7. Estimates of random variability provided?	10. Probability values reported?	11. Representative of source population?	12. Representative of recruited participants?	16. Any data dredging?	18. Statistical tests appropriate?	20. Outcome measures accurate?	21. Groups recruited From same population	22. Groups recruited At same time?	25.Adjusted for confounding variables?	27. Conducted power analysis	Total score (%)
Inada et al., (2015)	1	1	1	0	1	1	1	1	0	1	1	1	1	0	0	0	65%
Johnson et al., (2014)	1	1	1	1	1	1	0	1	0	1	1	1	N/A	N/A	1	0	73%
Joosten & Bundy (2010)	1	1	1	1	1	1	1	0	0	1	1	1	1	0	0	0	65%
Lidstone et al., (2014)	1	1	1	1	1	1	1	0	0	1	1	1	N/A	N/A	1	0	73%
Mccormick et al. (2014)	1	1	1	1	0	0	0	0	1	1	1	1	0	0	0	0	47%
Rogers et al., (2003)	1	1	1	1	1	1	1	0	0	1	1	1	0	0	0	0	59%
Schauder et al. (2015)	1	0	1	2	0	0	0	0	0	1	1	1	0	0	0	0	41%
Wiggins et al., (2009)	1	1	1	1	1	1	1	0	0	1	1	1	1	1	1	1	82%
Wigham et al., (2015)	1	1	1	1	1	1	1	0	0	1	1	1	N/A	N/A	1	0	73%

Appendix B. Additional methodological critiques of studies included within the review

Study and Country	Critique
Baranek, Foster & Berkson (1997) USA	• Small sample size recruited from 1 school. • Males overrepresented. • Used a range of sensory tactile measures and controlled for age.
Boyd, McBee, Holtzclaw, Baranek & Bodfish (2009) USA	• Small sample size recruited through 1 university registry. • All measures parent reported. • Controlled for age and intelligence. • Did not account for medication use as a confounding variable.
Boyd et al. (2010) USA	• Sample recruited via variety of methods, though within one state. • Used objective and carer reported sensory measures. • Gender a potential confounding variable
Chen, Rodgers & McConachie (2009) U.K	• Small sample size with overrepresentation of males. • Poor response rate, increasing likelihood of recruitment bias. • Reliable measures used, though all parent reported.
Foss-Feig, Heacock & Cascio (2012) USA	• Small sample size and limited representativeness. • All measures demonstrate good psychometric properties and used a range of sensory measures.
Gabriels et al. (2008) USA	• Sample recruited from a range of settings, but exact sources not stipulated. • No random variability estimates for main outcomes. • Controlled for intelligence, age, medication.
Gal, Dyck & Passmore (2010) Israel	• Large sample size. Controlled for intellectual disability. • Only used 1 teacher rated sensory measure.

Appendix B. (continued)

Study and Country	Sample size	Critique
Green et al., (2016) U.K	ASD; 116 SEN; 72	• Large sample size recruited from a large cohort sample. • Group age differences uncontrolled.
Inada et al. (2015) Japan	ASD; 274 Atypical; 36	• Large ASD sample recruited from 28 Japanese clinics. • Confounders (e.g. medication, age) uncontrolled.
Johnson et al. (2014) USA	256	• Large sample recruited from 5 sites across USA. • Controlled for medication effects. • However, sample was largely Caucasian.
Joosten & Bundy (2010) Australia	ASD & ID; 29 ID; 23	• Small sample size, recruited from 1 school. • High RRBs measured based upon referral for high RRB – subjective. • Gender may be a confounding variable.
Lidstone et al. (2014) study 2 UK	49	• Participants recruited from one source in Wales. • Measures used were parent reported, though, though illustrate good psychometric properties and controlled for age • Excluded sensory items on RBQ-2 to reduce multi-collinearity
Rodgers, Hepburn &Wehner (2003) USA	*ASD*; 26 Fragile X; 20 *DD*; 32 Control; 24	• Population source not stipulated. • Participant gender ratio unknown. • Ethnicity, socioeconomic status and verbal age controlled for
Wiggins, Robins, Bakeman & Adamson (2009) USA	34 (34 months; unknown)	• Small sample sizes recruited from 1 source, though did report effect sizes. • Sample ethnically diverse. • Controlled for age
Wigham, Rodgers, South, McConachie & Freeston (2015) UK and USA	53	• Small sample sizes recruited from 1 source, though did report effect sizes. • Sample ethnically diverse. • Controlled for age

Section 2: Research Report

Child Visual Perception in the Autism Spectrum: A Quantitative Study

Abstract

Objectives. The inhibition theory attributes superior visual abilities in autism (ASD) to increased neuronal inhibition. However, this contradicts the excitation theory, suggesting ASD is caused by increased neuronal excitation. The aim of the current study was to reconcile these competing theories by examining the impact of epilepsy (caused by increased neuronal excitation) on visual discrimination abilities. In line with the inhibition theory, it was hypothesised that the ASD group would show superior sensory discrimination abilities compared to neuro-typical and epilepsy control groups. Epilepsy participants would show the poorest performance. Associations between discrimination thresholds and repetitive and sensory behaviours were also explored.

Design. A non-randomised experimental design

Method. Three groups of children (autism, epilepsy and neuro-typical controls) completed a child adapted visual orientation discrimination task. To control for confounding variables participant's visual accuracy and non-verbal abilities were assessed. Caregivers also completed autism, depression and anxiety questionnaires.

Results. Orientation discrimination abilities were significantly correlated with age. Therefore, an ANCOVA, with age inputted as a co-variate was used to explore group differences. Results found no evidence of superior discrimination abilities within the ASD group. However, the epilepsy group showed significantly poorer discrimination abilities compared to the neuro-typical group. No significant correlations between threshold and restricted or sensory behaviours was found. Although, there was a trend towards significance.

Conclusions. The current study found no evidence of superior discrimination abilities in children diagnosed with ASD. Results suggest visual discrimination may not be a reliable index of increased inhibition within the child population.

Practitioner Points

- The finding that inhibition is associated with age and that other research has found superior discrimination abilities in adults diagnosed with ASD suggests more research is needed examining the developmental trajectory of visual discrimination abilities in children with ASD.
- As no significant differences between ASD and epilepsy groups was found, the research cannot assume that participants with epilepsy and ASD have different neural aetiologies. Orientation discrimination tasks do not seem a beneficial to use as a marker for identifying who may be more likely to benefit from different pharmacological interventions.
- The inclusion of a fourth group (children with co-morbid epilepsy and ASD) is required to more precisely reconcile the two conflicting hypotheses.
- Future studies require a much larger sample size, with the inclusion of participant electroencephalograms to examine inhibition and excitation more precisely.

Autism and sensory sensitivity

Autism spectrum disorder (ASD) is characterised by The Diagnostic and Statistical Manual of Mental Disorders, fifth edition, (DSM-V, 2013) as encompassing reciprocal social interaction and communication deficits, as well as restricted and stereotypical behaviours. The recently published DSM-V contains additional criteria within the restricted and stereotypical behaviours index, namely hypo/hyper sensitivities and sensory-seeking behaviour. This reflects a large body of literature suggesting children with ASD frequently display abnormal sensory behaviours (Ben-Sasson et al., 2009). For example, individuals with autism may be overly sensitive to and avoidant of certain noises, textures or sounds. Hypo-sensitivities may include having a high pain threshold or not acknowledging loud noises such as a fire alarm.

A range of sensory processing abnormalities have been examined under experimental conditions (see Haesen at al., 2011 and Simmons et al., 2009 for reviews). Across the modalities, some evidence suggests that individuals with ASD (or at least an ASD subgroup) show superior auditory (Bonnel et al., 2010; Jones et al., 2009; Mottron, Peretz & Menard, 2000; O'Riordan & Passetti, 2006), tactile (Blakemore et al., 2006) and visual discrimination abilities relative to neuro-typical controls. In relation to the latter, individuals with ASD have performed significantly better than neuro-typical controls in various cognitive tasks, including embedded figures (Brosnan, Gwilliam & Walker, 2012; Taylor et al., 2014), block design (Shah & Frith 1993) and visual search tasks (Gonzalez et al., 2013; Plaisted, O'Riordan & Baron-Cohen, 1998). These findings have also been replicated in individuals who exhibit ASD traits without a formal diagnosis (Milne et al., 2013; Almeida et al., 2012). The explanation given for superior performance on these types of tasks is enhanced discrimination abilities in ASD (O'Riordan & Plaisted, 2001; Mottron et al., 2007). Mottron et al. (2007) proposed the enhanced perceptual functioning theory. They argued that ASD individuals show superior performance because brain

regions associated with low level perceptual processing are over-functioning, leading to superior performance on tasks which process local details. This enables individuals with ASD to better recognise and distinguish subtle details and patterns, evidenced within the latter studies described.

More recently, visual orientation discrimination abilities have been assessed within the ASD population with mixed results reported. Bertone (2005) asked ASD and neuro-typical groups to identify the orientation (either vertical or horizontal) of luminance defined static gratings. They found individuals with ASD had superior orientation discrimination thresholds compared to neurotypical controls. Brock et al. (2011) employed a similar paradigm but instead asked participants to identify cardinally (i.e. vertically) oriented target gratings from oblique (i.e. slanted) distractor gratings. They found orientation discrimination thresholds and autistic traits were not significantly correlated. However, the latter study's methodology was criticised by Dickinson, Jones and Milne (2014), who argued that the results may be due to the oblique effect, a phenomenon whereby individuals perform better when angles are cardinally rather than obliquely aligned (Apelle 1972), thus giving rise to ceiling effects. Dickinson, Jones and Milne (2014) instead used oblique orientation gratings for both target and distractor stimuli. They found a significant negative correlation between autistic traits and orientation discrimination within a group of neurotypical participants, suggesting superior orientation discrimination thresholds for those with higher autistic traits. Dickinson, Bruyns-Haylett, Smith, Jones & Milne (2016) replicated the previous study using a clinical sample of adults diagnosed with ASD. They found that individuals with ASD had significantly lower (i.e. superior) orientation discrimination thresholds relative to matched neuro-typical controls. Gomez, Bennett, Dickinson & Milne (2015) extended Dickinson et al's. (2014) research to children. Using the same task they found no significant difference in discrimination thresholds for children with or without ASD.

However, it could be argued that the paradigm Gomez et al., (2015) used was not "child friendly" enough. The task used was the same as that given to the adult population. Consequently, the task may have been too long and /or boring to obtain reliable thresholds from children. Additionally, Gomez et al. (2015) used a standard psychophysical paradigm whereby task difficulty changes in response to the participant's performance (Levitt 1971). However, this method cannot differentiate participants who genuinely had a high threshold from those who had a high threshold score because they were not attending to the task. Therefore, the current study aimed to explore whether orientation discrimination thresholds are lower in children with ASD when a more suitable task is used.

Biological mechanisms of visual discrimination

Superior visual discrimination appears to be a result of individual differences within the neuronal excitatory and inhibitory (E:I) balance (Dickinson et al., 2016). Excitatory neurons are cells that release particular neurotransmitters (e.g. Glutamate) which positively charge the cell. The positively charged neuron will then transmit information to another neuron, which in turn will do the same. To impose order and ensure that the brain is not always excited and active, the brain also produces inhibitory neurons. Inhibitory neurons release Gamma-Aminobutyric acid (GABA), resulting in a negatively charged cell, stopping neuronal excitation in its tracks (Purves et al., 2001). The E:I balance therefore refers to the ratio of excitatory to inhibitory neurons in the brain.

Research suggests that increased inhibition leads to better discrimination. Using the Autism Spectrum Quotient (AQ, Baron-Cohen, 2001) Dickinson et al. (2016) examined the relationship between autistic traits and gamma activity using electroencephalography. They examined peak gamma frequency, as its oscillatory activity provides an index of the E:I balance (Buzsaki & Wang, 2012), with higher peak gamma frequency associated with higher inhibition levels (Brunel & Wang, 2003). The

authors found a significant positive relationship between AQ scores and peak gamma frequency. The same sample showed lower discrimination thresholds were associated with increased AQ scores (Dickinson et al., 2014). Moreover, Edden, Muthukumaraswamy, Freeman & Singh (2009) found superior discrimination thresholds were associated with increased peak gamma frequencies. In combination, results from the latter three studies imply that the relationship between superior perceptual discrimination and autistic traits is a result of increased neural inhibition. Bertrone (2005) provided further support, arguing that increased perceptual sensitivity arises from abnormal neural connectivity, specifically increased inhibition.

Nonetheless, the latter literature suggesting an association between neural inhibition, perceptual discrimination and ASD is at odds with the dominant theory proposed by Rubenstein and Merzenich (2003), which suggests an increased ratio of excitatory to inhibitory neuronal circuits cause some types of autism. Support for this hypothesis comes from co-morbidity research suggesting that 30% of individuals with autism also have epilepsy, a disorder caused by increased neuronal excitation (Gillberg & Billstedt, 2000). The increased excitation to inhibition hypothesis has led to drug trials addressing the imbalance by increasing inhibitory neurotransmission (Erickson et al., 2014). However, they have shown varying success, with only a subgroup of individuals with ASD improving. An alternative explanation which would help reconcile the two competing theories is that perhaps co-morbid epilepsy is influencing the relationship between autism and increased excitation. If non-comorbid epilepsy and autism individuals instead have different types of abnormal connectivity (with epilepsy showing an increased ratio of excitatory to inhibitory neuronal circuits and autism showing the opposite), then it would be expected that, relative to the epilepsy group, the autism group would show superior discrimination abilities, given this is thought to be a marker for increased inhibition.

Research examining discrimination abilities in individuals diagnosed with epilepsy is mixed. Grant et al. (2007) found no significant differences between participants with temporal lobe epilepsy and neuro-typical controls when undertaking two visual discrimination tasks. As epilepsy participants performed normally whilst also taking multiple anti-epileptic drugs, Grant et al. (2007) argued antiepileptic drugs did not impact upon visual discrimination abilities. Similarly, Arnedo et al. (2009) found whilst adults with temporal lobe epilepsy were poorer on auditory discrimination tasks relative to matched controls, this did not reach significance. Nonetheless, Grant et al. (2005) conducted a tactile grating orientation task with epilepsy and neuro-typical controls. They found the epilepsy group's mean discrimination threshold was twice as high as neurotypical controls, suggesting that individuals with epilepsy show impaired tactile discrimination. Moreover, performance remained impaired without the presence of anti-epileptic drugs, suggesting this cognitive impairment is not due to medication.

Associations between orientation discrimination and specific symptoms of autism and other clinical variables

To date, there is a lack of research examining the association between enhanced perceptual discrimination abilities and particular clusters of ASD symptoms. Jones et al. (2009) suggested one under-researched hypothesis is that abnormal processing of auditory information results in abnormal patterns of sensory behaviours. The same could be said for abnormal processing of visual information (i.e. perceptual correlates map onto their behavioural counterparts). Indeed, Baron Cohen (2009) found enhanced perception is associated with particular sensory abnormalities, namely hyper-responsiveness.

Superior discrimination abilities have also been associated with an increased number of repetitive and restricted behaviours (Chen et al., 2009; Evans et al., 2001; Kargas et al., 2015). Thus, it could be questioned whether enhanced perceptual discrimination

abilities are associated with the second Autism DSM-V category; repetitive and restricted behaviours (which also includes abnormal sensory behaviours). This may help to explain why some studies have found only some individuals with ASD have enhanced perception (Bonnel et al., 2010; Jones et al., 2009).

Morevoer, Reports suggest children with ASD (Gurney et al., 2006; Stewart et al., 2006) and epilepsy (Barry et al., 2008; Kanner, 2003a) are also more likely to have depression and anxiety compared to neuro-typical controls. Depression and anxiety have also been shown to influence visual perception (Yilmaz, Akca, Acikel, Bilgic & Kilic, 2015; Fam, Rush, Haaland, Barbier & Luu, 2013). No previous visual orientation studies have examined the impact of depression and anxiety upon visual discrimination thresholds. The present study aimed to exploritarily investigate whether these variables do impact. Therefore, to control for anxiety and depression influencing threshold scores and confounding results the Revised Depression and Anxiety Scale-parent version (RCADS; Chorpita, Yim, Moffitt, Umemoto & Francis 2000) was administered.

Aims

In order to reconcile the two conflicting theories (increased inhibition versus increased excitation) of ASD, the aim of the current study will be;

1) To establish whether orientation discrimination thresholds are lower (i.e. superior) in children with ASD when the orientation discrimination task is adapted to make it more child friendly. To explore this, discrimination thresholds from three groups of children will be compared; children with epilepsy, children with ASD and neuro-typical children.
2) To examine whether certain clusters of ASD symptoms are more related to visual orientation threshold scores, to increase understanding surrounding how (if at all) visual perceptual correlates map onto their behavioural counterparts.

Clinical and theoretical implications

Whilst there is some evidence to suggest superior discrimination in ASD, there is also counter evidence. The study will therefore provide further insight into the nature and prevalence of atypical sensory processing in ASD. If the study is able to infer increased inhibition in individuals with ASD through the identification of discrimination thresholds, this will imply that different groups of ASD may have different neural aetiologies, and would therefore require different interventions (e.g. individuals with ASD may require a different intervention to those with both ASD and epilepsy). Whilst one group may require drugs to increase inhibition, this may be detrimental to those who exhibit increased inhibition. Results from this study may indicate that only some individuals with ASD are likely to benefit from these types of drugs, and may provide way of identifying which individuals may benefit through the use of an orientation discrimination task. This will also provide an objective measure of perceptual sensory behaviour to compliment self-reported parent measures. Additionally, the study will examine the impact of epilepsy on perceptual sensitivity, in an attempt to reconcile the two competing E:I balance theories.

Moreover, if orientation discrimination thresholds are related to sensory or repetitive and restricted behaviours, this may suggest that visual discrimination abilities serve as a risk factor in the development increased repetitive and sensory behaviours. It will also add to the paucity of research which has explored an association between perceptual processes at the cognitive level and their behavioural manifestations.

Hypotheses

H[1] In line with Dickinson et al's (2014) increased inhibition theory it is hypothesised that children with autism will show superior sensory discrimination abilities compared to neuro-typical controls and the epilepsy group, after taking into account depression and anxiety scores

H[2] Given that increased neural excitation is identified in epilepsy, it is hypothesised that the epilepsy group will show significantly poorer sensory discrimination than the neuro-typical and autism groups, after taking into account depression and anxiety scores

H[3] Better discrimination scores will be found in those individuals with higher repetitive and sensory behaviour scores

Methods

Design

A between subjects' cross-sectional non-randomised experimental design was used to examine perceptual discrimination thresholds. The independent variable was defined as neurodevelopmental diagnosis. Participants were categorised into one of three groups; those with a diagnosis of autism (ASD group), those with a diagnosis of epilepsy (epilepsy group), and those with neither diagnosis (neuro-typical control group).

Ethical considerations

Ethical approval was granted by the NHS Research Ethics Committee (REC) and the Research and Development Department at Sheffield Children's Hospital NHS Trust (Appendices A). All parents and participants provided written informed consent (Appendices E & F). Participants were told prior to beginning the study that they had

the right to withdraw from the study at any time. This was highlighted on the consent and debrief forms (Appendices E, F, G, H, I).

Recruitment

The clinical (epilepsy and ASD) groups were recruited from the Ryegate Children's Centre, Sheffield Children's NHS Foundation Trust between August 2016 – March 2017. Participants in the neuro-typical control group were recruited via opportunistic sampling using social media (an open Facebook page connected to the main researchers Facebook) and the staff Sheffield University email system. The following inclusion and exclusion criteria were applied;

Inclusion criteria. To be included within the study, participants were required to fall within the normal intellectual functioning range (I.e. an IQ above 70). The participants were over 6 years of age and under 14 years, eleven months. The clinical groups had to have received either a diagnosis of autism or epilepsy. This was screened by the Ryegate staff.

Exclusion criteria during participant screening stage. To control for confounding variables such as the presence of other co-morbidities, Ryegate staff excluded those with a diagnosis of Attention Deficit Hyperactivity Disorder (ADHD) or a learning disability. Moreover, given that the task was conducted on a computer with quickly moving stimuli and flashing animation, potential participants who had photosensitive epilepsy were also excluded. Ryegate professionals ensured that the ASD group participants did not have a co-morbid diagnosis of epilepsy and the epilepsy group participants did not have a co-morbid diagnosis of ASD by examining their health records held at the Ryegate Centre.

Exclusion criteria following research participation. To ensure group homogeneity, participants in the neuro-typical and epilepsy groups were excluded if

they scored higher than the diagnostic cut off value (65) on a parent-reported ASD questionnaire, namely the Social Responsiveness Scale-2 (SRS-2; Costantino & Gruber, 2012; Appendix J). Participants in the neurotypical groups were excluded if they had a 1st degree relative (parents or siblings) with autism and/or epilepsy or reported a history of epileptic seizures. Finally, basic vision was screened using the Log Mar visual acuity test (Keeler ophthalmic instruments). Those who did not have corrected to normal vision (a Log MAR score of 0.2 or above) were also excluded from the study. None of the participants included within the study met the latter exclusion criteria. Additionally, participants were excluded if they performed at chance level on the orientation discrimination task.

Participants

Forty-six participants (autism group *n* =14; epilepsy group *n* = 10; neurotypical group *n* = 22) took part in the study. Though 1 participant was excluded following participation, as their visual orientation results indicated only chance performance, suggesting they may not have not have understood the task. Figure 1 depicts the recruitment process. Overall, 290 clinical participants were initially identified (123 epilepsy participants and 167 ASD participants). Nine percent of the screened ASD sample and eight percent of the epilepsy sample agreed to take part. However, 1 participant diagnosed with ASD did not attend due to increased levels of anxiety and was subsequently excluded. For the control group, thirty-seven responded to email and social media adverts asking for more details. Fifty nine percent agreed to take part after being emailed study information sheets. One neurotypical control was excluded after performing at chance level on the orientation discrimination task. Forty-five participants were therefore included within the analysis (20 males and 25 females, age range 72-178 months, Mean = 123 months, SD = 34.24).

The epilepsy group was classified according to the International League Against Epilepsy (ILAE) classification system (Scheffer et al., 2017). Four participants were classified as having a focal epilepsy and six having generalised epilepsy. Seven epilepsy participants (70%) were using anti-epileptic medication. Anti-epileptic medication included sodium valproate (*n*= 2), Levetiracetam (*n* =2), lamotrigine (*n*= 1), ethosuximide (*n* =1) and carbamazepine (*n* = 1). In terms of co-morbidities, 2 of the ASD group had albinism and 1 participant had a diagnosis of developmental co-ordination disorder. In the epilepsy group 1 participant had tuberous sclerosis and 1 had idiopathic chronic constipation.

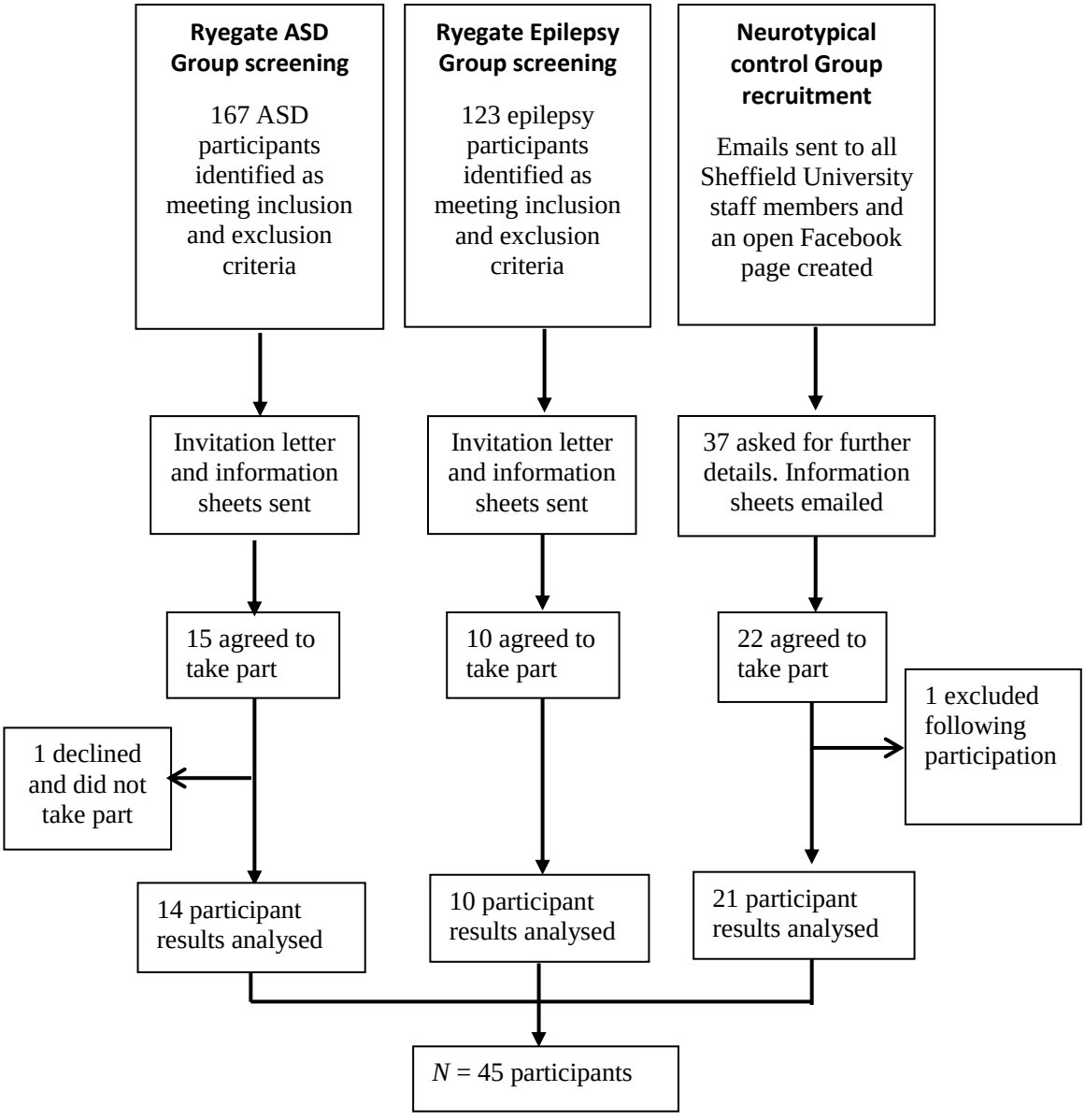

Figure 1. *Participant recruitment flowchart*

Table 1. *Participant characteristics*

	Neurotypical	Autism	Epilepsy
Age Mean (SD)	119 (35.58)	130 (32.76)	118 (36.04)
Gender *N* male (%)	9 (43%)	7 (50%)	4 (40%)
Location tested *N* (%)			
Home	16 (46%)	8 (57%)	5 (50%)
Ryegate	0	4 (29%)	4 (40%)
Sheffield University	5 (24%)	2 (14%)	1 (10%)
Seizure Type *N* (%)			
Focal	-	-	4 (40%)
Generalised	-	-	6 (60%)
Combined	-	-	0
Medication (%)			
Anti-epileptic drugs	-	-	7 (70%)

Power analysis

Due to time constraints, the sample size was below that expected from an initial a priori power analysis, calculated using G*power (Faul, Erdfelder, Lang, & Buchner, 2007). Given that similar studies conducted in the past have only compared two groups (Dickinson et al., 2015; Grant et al., 2005; Gomez et al., 2015), Cohen's conventions were used to estimate the effect size for the current study. Whilst Dickinson et al. (2014) only compared two groups, an effect size of 0.46 was evident when comparing the ASD group (Mean; 5.81; SD; 2.26) to the neuro-typical control group (Mean; 6.88; SD; 2.37). Therefore, assuming a Cohens D effect size (f = 040), with alpha set at .05 and power set at .80, the initial a priori power analysis suggested 66 participants would

need to be recruited to compare the three groups against each other using a one way between subject's ANOVA analysis.

Apparatus and Stimulus Material

Gomez et al. (2015) used a one up three down adaptive staircase procedure (Levitt 1971), whereby task difficulty is based upon the individual's ongoing performance. However, this method cannot differentiate participants who genuinely had a high threshold from those who had a high threshold score because they were not attending to the task. Therefore, the current study used the method of constant stimuli, whereby the difference between target and distractor orientation stimuli are randomised throughout the task.

A preselected set of stimuli was presented several times in a fixed randomised order. The perceptual discrimination task was presented using EPRIME on a 15.6 inch HP Pavillion 15-p248sa laptop screen, with a screen resolution of 1366 x 768 pixels. Participants were seated 57cm away from the monitor, in a dark room.

The stimuli used in the perceptual discrimination task consisted of 6 stimuli, each depicting a horizontal line rotated at differing degrees. A single horizontal line rather than oblique gratings was used to make the task easier for children. The stimuli were developed using Matlab (The Mathworks, 2000) using the PsychToolbox set of functions (Brainard, 1997). Stimuli have a diameter of 4^{o} and a mean luminance of 83 cd/m2. The reference stimulus was always orientated at 45 degrees. The 5 remaining target stimuli were oriented away from the reference stimulus by a range of degrees, namely 1^{o}, 3^{o}, 5^{o}, 7^{o} and 9^{o}. Each target stimulus was presented at 24 times, 12 times clockwise and 12 times anticlockwise from the reference stimulus. The number of target stimulus values chosen and the frequency of trials for each value was based upon recommendations made in psychophysics literature (Ehrenstein & Ehrenstein, 1999).

The specific degrees of difference between the reference and target gratings were based upon previous findings by Dickinson et al. (2014) and Gomez et al. (2015). The current study piloted these values on a 7 year old child who was able to understand and complete the paradigm and reported enjoying it.

The discrimination task consisted of one practice block, consisting of 10 trials. The trial presented all the combinations of degrees (1^o, 3^o, 5^o, 7^o and 9^o clockwise and anticlockwise once each) in a fixed order. If participants understood the practice task they then conducted the full experiment. The main experiment consisted of 120 trials split into 4 blocks. Within the blocks each of the 5 values were presented 3 times clockwise and 3 times anticlockwise from the reference grating.

In line with Dickinson et al. (2014) and Gomez et al. (2015), in each trial (illustrated in Figure 2 below) a central fixation cross was presented (250ms) followed by the reference grating (350ms), another fixation (100ms), the target stimulus (350ms) and finally the response screen. To make the task more child friendly the orientation discrimination task was presented in the context of a game. The task followed the story of a surfer bear called Bob who asked participants for help in deciding which direction the waves (i.e. the detection of a change in line orientation) were going and help him move his surf board in the right direction. On each trial participants were asked to identify whether the target rotated left (anticlockwise) or right (clockwise) compared to the reference stimulus. Given that the participants were children, they were asked to verbally report whether it was left or right or/and indicate the direction by pointing left or right. Giving the option of pointing helped younger children who had difficulty with concepts such as ‘left’ and ‘right’. The experimenter recorded each response using the keyboard. At the end of each block of 30 trials the participants saw a picture of Bob surfing closer to the shore and received verbal encouragement from the experimenter. At the end of the experiment participants saw a "Well done!" message which showed a

picture of Bob at the shore. After each block the participant was given the opportunity for a short break of approximately 2-5 minutes before beginning the next block. The entire procedure lasted between 15 to 30 minutes depending on how much time it took to explain the task to each child, how quick they were to respond to each trial and also how long they chose to break for between the blocks.

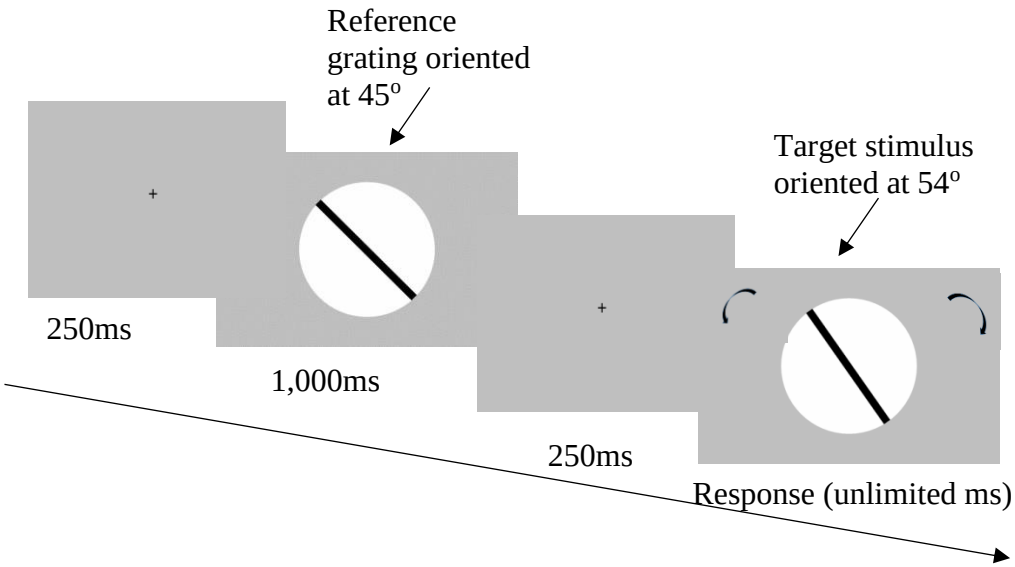

Figure 2. *Example of a trial in which the target grating is 9° (clockwise) from the reference grating*

Visual acuity task

The Log MAR uncrowded test (Keeler ophthalmic instruments) was administered to ensure that uncorrected visual difficulties did not confound results. In the Log MAR test individuals are presented with sets of two black letters printed on a white background. With each new card presented, the two letters decreased in size. The task ends when the participant cannot identify any of the two letters presented on one card. A score is calculated in log units, based upon the number of letters correctly identified. 20/20 (i.e. average) vision will produce a log MAR score of 0.0. To minimise harm it, if children were identified as having a visual impairment, defined as a Log MAR score of 0.2 or above (equivalent to 6/9 or 20/32 vision), they would be excluded from the orientation discrimination task. The task stopped once participants had

achieved 0.2. If a participant obtained a score above the threshold there was a protocol in place for further action to be taken. This was in the form of writing a letter to the GP **(**Appendix K), informing parents of findings and encouraging them to book an appointment with an optician. However, all participants passed the test and so this protocol was not used.

Demographic and Clinical Measures

Demographic Questionnaire. Information collected from participants included age, gender, current medication and type of epilepsy/autism diagnosis. The number of epileptic seizures per year, medical or mental health diagnoses and any family history of developmental disorders was also collected (Appendix L).

Depression and Anxiety. Reports suggest children with ASD (Gurney et al., 2006; Stewart et al., 2006) and epilepsy (Barry et al., 2008; Kanner, 2003a) are also more likely to have depression and anxiety compared to neuro-typical controls. Depression and anxiety have also been shown to influence visual perception (Yilmaz, Akca, Acikel, Bilgic & Kilic, 2015; Fam, Rush, Haaland, Barbier & Luu, 2013). Therefore, to control for anxiety and depression influencing threshold scores and confounding results, the Revised Depression and Anxiety Scale-parent version (RCADS; Chorpita, Yim, Moffitt, Umemoto & Francis 2000) was administered. The RCADS (Appendix M) was chosen as it is a valid and reliable measure of depression and anxiety for children both within the general population (Chorpita et al., 2000) and within ASD populations (Hallett et al., 2013; Kaat & Lecavalier, 2015; Sterling et al., 2015). Although the reliability and validity of RCADs within an epilepsy sample has not been conducted, the questionnaire has been used with an epilepsy sample in the past (Rizou et al., 2015). The RCADs consists of 47 items, with each item rated on a 4 point Likert scale. Items are divided into six subscales; separation anxiety disorder, social

phobia, panic disorder, generalised anxiety disorder, obsessive compulsive disorder and major depressive disorder. It also provides total anxiety and depression scores.

Social and Communication difficulties. To measure social and communication impairments in ASD, the Social Responsiveness Scale Second Edition for 4-18 year olds (SRS-2, Costantino & Gruber, 2012) was completed by parents. The SRS-2 (Appendix J) is 65 item questionnaire which rates the severity of behaviours using a 4 point Likert scale. The SRS-2 provides 5 subscales; social awareness, social cognition, social communication, social motivation and repetitive behaviours. SRS-2 scores range from 0-195, with increased scores representing higher social impairments. SRS-2 scores are converted into t scores. The SRS-2 suggests a t score between 60-65 indicates mild difficulties, whereas t scores between 66-75 and 76 or higher suggest moderate and severe difficulties respectively. Therefore, the cut off score for an ASD diagnosis is a t score of 65. Given that participants within the ASD group already have a diagnosis of autism this cut-off score did not change their group allocation. However, for those in the neuro-typical and epilepsy groups, any individual scoring above the latter cut-off scores would have been excluded from the analysis. The SRS-2 also provides conversion t scores for DSM-5 compatible scales, namely social communication and interaction (SCI) and repetitive and restricted behaviour (RRB) scales. The SRS-2 has good internal consistency (0.95), interrater agreement (0.61) and convergent validity (Costantino & Gruber, 2012).

Repetitive and Restricted behaviours. To measure repetitive and restrictive behaviours, parents completed the Repetitive Behavioural Questionnaire-2 (RBQ-2, Leekham et al., 2007). The RBQ-2 (Appendix N) is a 20-item questionnaire which includes items from the original 33 item RBQ (Turner, 1995) and The Diagnostic Interview for Social and Communication Disorders (DISCO, Wing, Leekham, Libby, Gould & Larcombe, 2002). It examines a wide range of repetitive and restrictive

behaviours including stereotyped motor behaviours, rituals, routines and sensory behaviours. The RBQ-2 items relate to current behaviours observed within the last month. It rates frequency of behaviours using a three point Likert scale; never/rarely (1 point), mild/occasional (2 points) and marked/notable (3 points). The total score is calculated by summing all item scores and dividing by the number of items completed. The RBQ-2 has good psychometric properties and high internal consistency (Leekham et al., 2007)

Patterns of sensory behaviour. The Short Sensory Profile 2 (SSP-2, Dunn, 2014; Appendix O) is a caregiver questionnaire for children up to aged 14 years 11 months. The SSP-2 is a 38-item questionnaire examining four sensory patterns of behaviour; sensory seeking, sensory avoiding, sensory sensitivity and sensory registration. Caregivers respond using a five-point Likert scale ranging from 1 (always) to 5 (never). The Short Sensory Profile 2 has good internal consistency (Cronbachs.79-.93) and test-retest reliability (.93-.97). Raw scores are provided as well as a percentile range which offers qualitative information regarding the frequency of sensory patterns in relation to other children (much less than others, less than others, average, more than others or much more than others). However, the test has only been validated on American individuals.

Non-Verbal Intelligence. The Matrix Reasoning subtest from the Wechsler Abbreviated Scale of Intelligence (WASI; Wechsler, 1999) was completed by all participants to ensure no differences in non-verbal intelligence existed between groups. The test involved individuals identifying the missing element that makes a pattern.

Procedure

An invitation letter and information sheets (Appendices A, B & C) were sent to 290 participants diagnosed with autism or epilepsy whom met inclusion and exclusion

criteria. For the neuro-typical control group, parents/caregivers responded to a university staff email or Facebook page. They then emailed or rang the main researcher to confirm whether they wished to take part in the study. Participants chose whether to conduct the research at Sheffield University (*n* =8), the Ryegate centre (*n* = 8) or at their home (*n* = 29 plus 1 excluded) to provide participants with flexibility. Upon arrival, the information sheets were reviewed and written consent was obtained from parents (Appendices E) and children (Appendix F). The duration of the study was approximately 45-60 minutes. Parents were asked to complete the demographic and clinical information sheet (Appendix L), the Sensory Profile-2 (Appendix O), RCADS (Appendix M), SRS-2 (Appendix J) and RBQ-2 (Appendix N). Before beginning the visual discrimination task the participants had their vision assessed using the Log MAR test (approximately 10 minutes).

Participants then conducted the computerised visual discrimination task (approximately 15-30 minutes). Following the visual discrimination task participants completed the WASI (approximately 10-15 minutes). Finally, participants and their caregivers were then given debrief forms (Appendices G, H & I) and asked if they had any questions.

Data Analyses

Data was analysed using version 23 of the Statistical Package for Social Sciences (SPSS, IBM, 2015) data processing software. All data was screened to ensure it met assumptions of normality, linearity and homogeneity. Independent measures ANOVAs were conducted to examine differences between the three groups (ASD, epilepsy and neuro-typical groups) in terms of age, non-verbal intelligence (matrix reasoning) scores, depression, anxiety (measured using RCADs), social and communication difficulties (measured using SRS-2) and repetitive behaviours

(measured using the RBQ-2 and SRS-RRB subscale). This aimed to ensure groups were homogeneous in terms of age and non-verbal IQ. It also aimed to ensure that groups were different in expected domains (e.g. ASD group scoring significantly higher on ASD questionnaires). Correlations were used to examine associations between age, depression, anxiety, matrix reasoning and the dependant variable visual orientation threshold, to detect any potential confounding variables. Significant correlations were inputted as a co-variate using an ANCOVA.

The effect of neurodevelopmental group on visual orientation discrimination was analysed using a one factor (3 levels; autism, epilepsy and neuro-typical groups) between measures ANCOVA design, with age (months) inputted as a co-variate. Main effects were examined using Bonferroni pairwise comparison tests.

Additionally, Spearman correlational analyses were conducted to examine associations between orientation discrimination thresholds, repetitive and restricted interests (measured using the RBQ-2 and SRS-RRB subscale), sensory abnormalities (measured using the SSP-2).

Visual orientation discrimination thresholds (dependant variable) were calculated using the psychometric function, which is an inferential model often applied in discrimination tasks and psychophysical research (Klien, 2001; Wichmann & Hill, 2001). Firstly, the proportion of correctly identified and incorrectly identified responses is calculated for each of the five stimulus values (1^o, 3^o, 5^o, 7^o, 9^o). The percentage of correct responses is plotted onto the Y axis and the five stimulus values plotted on the X axis See Figures 3 and 4 below). Using matlab version 9.1 a psychometric curve was fitted to calculate the threshold where participants got 85% of responses correct. A value of 85% was used as psychometric research suggests this constitutes a reliable threshold (Smith, Gamble & Heil, 2010).

Results

Data screening

Orientation threshold scores, age (months), matrix reasoning scores, the SRS-2 total and domain scores, sensory profile-2 domains, the RBQ-2 and the RCADs total, depression and anxiety scores were screened to measure assumptions of normality, homogeneity and linearity. The data was also screened for outliers. Matrix reasoning scores and age were the only variables which did not violate any assumptions. Threshold scores violated the assumption of normality for the epilepsy group and a significant Levene statistic suggested homogeneity was not assumed. Therefore, the data was transformed using a square root transformation. Assumptions were met once the data was transformed and no outliers were identified. Thus, parametric models were used to examine hypotheses 1 and 2. The SRS-2, SRS-RRB, SRS-SCI, RBQ-2 and RCADs total, depression and anxiety scores violated at least one assumption. Transformation procedures did not eliminate violations. Thus, a non-parametric correlational equivalent (Spearman) was used when examining the third hypothesis exploring the relationship between threshold scores and ASD characteristics. Group differences were also explored using non-parametric models with exception to age and matrix reasoning.

Hypothesis 1 and 2 - Group differences in visual orientation threshold

Calculating orientation discrimination thresholds. Using the method of constant stimuli individuals are expected to get most of the easy discriminations correct, with a reduction of correct responses as the discriminations become more difficult (see Figure 3 for an example of one participant's normal threshold curve). If participants were providing incorrect answers for the easiest trials and were showing similar responses for easy and hard trials (i.e. chance rate responses), it was assumed that they

either did not understand the task or were not paying attention. This was used to discriminate them from participants who genuinely have high discrimination thresholds. One participant was excluded as their threshold curve highlighted chance rate responses (see Figure 4 for excluded participant's threshold curve). All other participants produced data curves which indicated that they were performing as expected, thus their threshold estimate can be considered reliable.

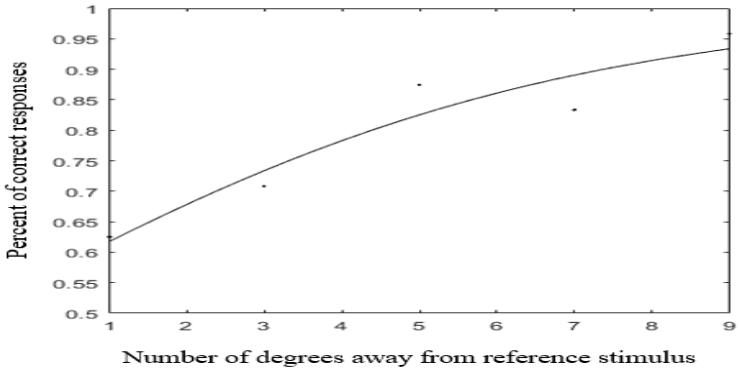

Figure 3. *An example of a typical orientation threshold curve, whereby the number of correct responses is higher when the judgement to be made is easier than when it is harder*

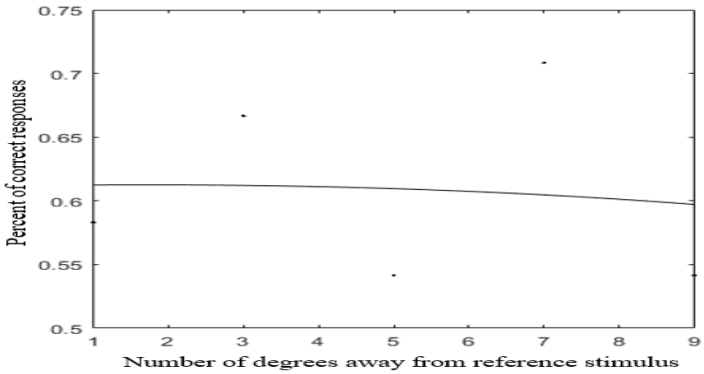

Figure 4. *Excluded participant's threshold curve indicating they are performing only slightly higher than chance levels and performance is unaffected by task difficulty*

Orientation discrimination group differences. It was hypothesised that the ASD group would show superior visual discrimination abilities compared to the epilepsy and neuro-typical control group, after taking into account depression and anxiety scores. Additionally, it was hypothesised that the epilepsy group would show significantly poorer visual discrimination compared to the Autism and neurotypical control groups.

To investigate any potential confounding variables influencing group differences in perceptual orientation thresholds, correlations (Pearson's for parametric data and Spearman's for non-parametric data) were conducted to examine the relationship between perceptual orientation threshold scores and the variables depicted in Table 2 below. A Pearson correlation found age (months) was the only variable significantly associated with orientation threshold ($r(43) = -.46$, $p = .001$). As participants got older, their orientation threshold became lower, suggesting better performance with increasing age. Moreover, the relationship between orientation discrimination and anxiety and depression was examined at the sungroup level. Due to multiple comparsions a more conservative 0.02 probability value was selected calculated using a Bonferroni comparison ($p = 0.05 / 3$ comparisons; total anxiety and depression score, depression score and anxiety score) to reduce type 1 errors. Analysis as the subgroup level showed no significant relationships between any group and depression (neurotypical group $r = .19$; ASD group $r = .04$; epilepsy group .47. all $p > 0.02$), anxiety (neurotypical group $r = .28$; ASD group $r = .32$; epilepsy group .64. all $p > 0.02$) and total anxiety and depression scores (neurotypical group $r = .22$; ASD group $r = .24$; epilepsy group .64, all $p > 0.02$.

Therefore, to examine differences in orientation discrimination thresholds a 3 level between subjects (Group; neurotypical, ASD and epilepsy) ANCOVA was

conducted, with age (in months) entered into the analysis as a co-variate, to ensure any significant results were not influenced by this variable.

The ANCOVA revealed that the covariate, age, was significantly related to orientation threshold, F (1, 41) = 13.44, p = .001, ηp^2 = .25. There was also a significant main effect of group on orientation threshold after controlling for age, F (2, 41) = 4.33, p = .02, ηp^2 = .18. Post hoc Bonferroni comparisons revealed that the epilepsy group had significantly higher threshold scores (Mean = 2.26, SD = .61) than neurotypical controls (Mean = 1.74, SD = .42, r = .008 p =.17), suggesting that the epilepsy group performed worse, though the effect size was extremely small. However, no significant differences were found between the ASD group threshold scores compared to epilepsy (p = .46) or neurotypical control (p = .48) groups (Figure 5). Moreover, the epilepsy group was further divided into non medicated (n =3_ and medicated (n=7) groups, to examine whether the possibility of medication effects serving as a confounding variable. Due to small sample sizes, only descriptive statistics were computed. The medicated epilepsy subgroup had a higher mean threshold compared to the non-medicated group, suggesting anti-epileptic medication may negatively impact upon orientation discrimination abilities.

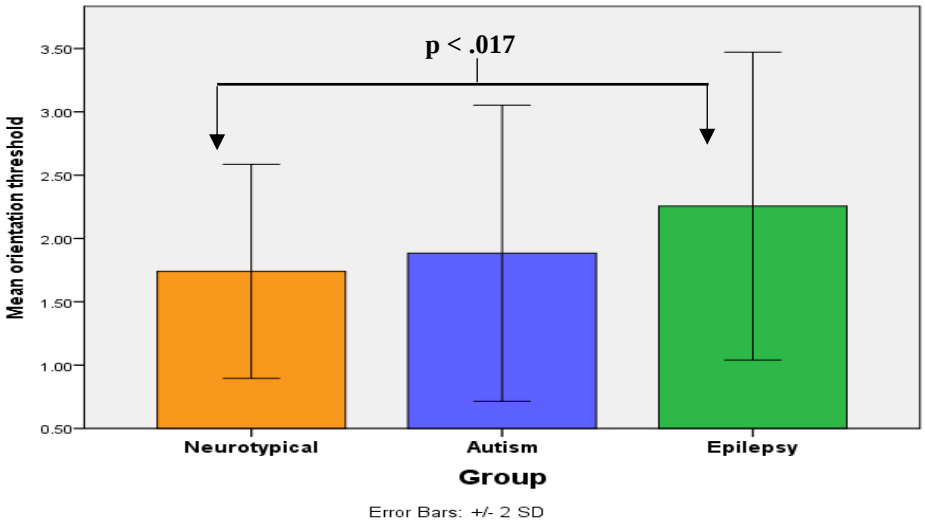

Figure 5. *Bar chart depicting group orientation discrimination means and standard deviations*

Group comparisons for demographic and clinical variables. Group comparisons were conducted to ensure the groups were well matched or showed expected group differences (for example in terms of ASD symptoms). An independent measures ANOVA showed no significant differences between ASD, epilepsy and neurotypical groups in terms of matrix reasoning scores and age, suggesting the groups were well matched on the latter variables (Table 2).

Characteristics of Autism. As expected, an independent samples Kruskal-Wallis found groups significantly differed with regards to the SRS-2 total score, H (2) = 30.01, p = .001. Pairwise comparisons with adjusted p values found the ASD group showed significantly higher SRS-2 scores (Median = 64, Range = 39) compared to epilepsy (Median = 48, Range = 17, r = -.41, p = .019) and neurotypical control groups (Median = 43, Range = 19, r = -.82, p = .001). Similarly, when examining the SRS-2 social communication and interaction subscale (SCI), a main group effect was found (H (2) =27.28, p = .001). Adjusted pairwise comparisons found the ASD group showed significantly higher social communication and interaction difficulties (Median = 62, Range = 36) compared to epilepsy (Median = 46, Range = 15, r = -.48 p = .004) and neurotypical controls (Median = 43, Range = 54, r = -.77 p = .001).

Additionally, a main group effect on the SRS-RRB subscale scores were also evident (H (2) = 26.42, p = .001). However, interestingly, compared to neurotypical controls (Median = 44, Range = 16) RRB scores were significantly higher for both epilepsy (Median = 59, Range = 27, r = .44, p < .001) and ASD groups (Median = 71, Range = 47, r = .74, p <.001). Moreover, there was a significant main effect of RBQ-2 scores (H (2) = 16.36, p = .001). Pairwise comparisons showed RBQ-2 scores were only significantly different between neurotypical (Median = 22, Range = 15) and ASD groups (Median = 32, Range = 15), with the ASD group displaying higher RBS scores

(r = .60, p = .001). No significant differences between the ASD and epilepsy groups were found. Descriptive statistics for the SRS total, SRS—SCI, SRS-RRB and the RBQ and are depicted visually in Figures 6, 7, 8 and 9 respectively.

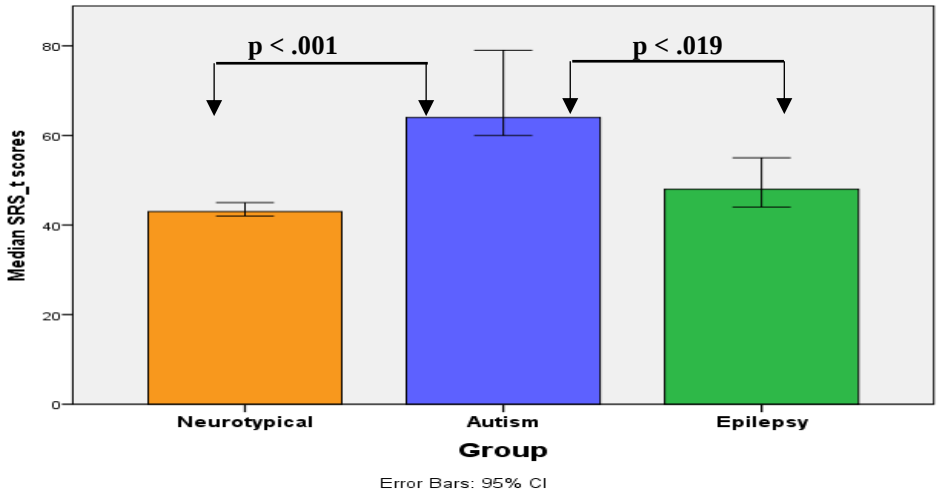

Figure 6. *Median SRS-2 total scores and error bars depicting confidence intervals*

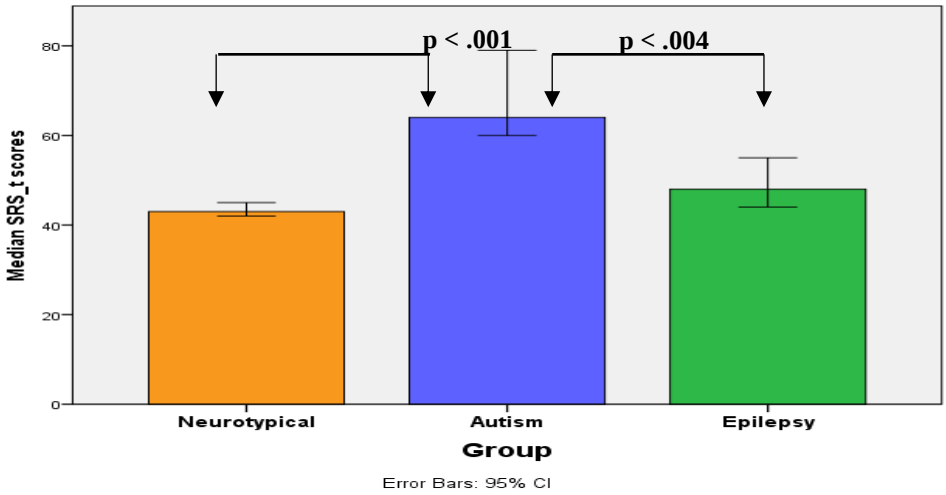

Figure 7. *Median SRS social and communication domain scores and error bars depicting confidence intervals*

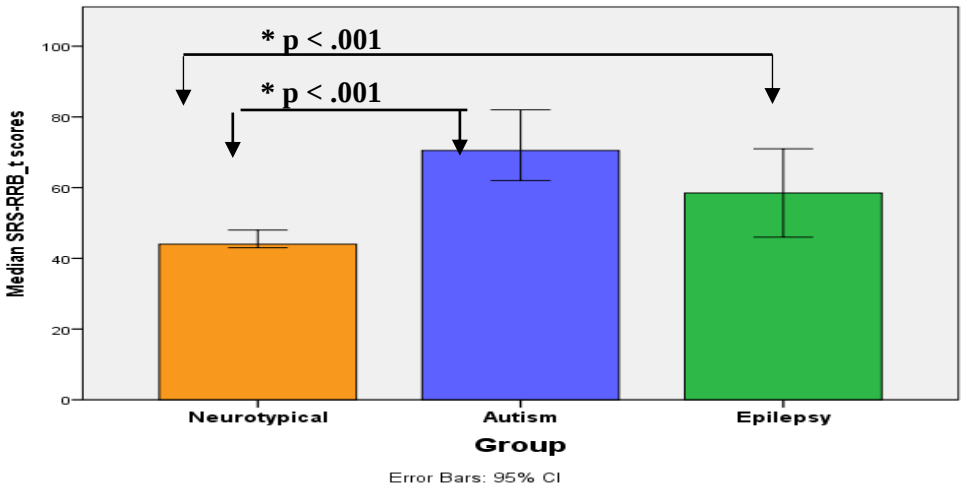

Figure 8. *Median SRS restricted and repetitive behaviour scores and error bars depicting confidence intervals*

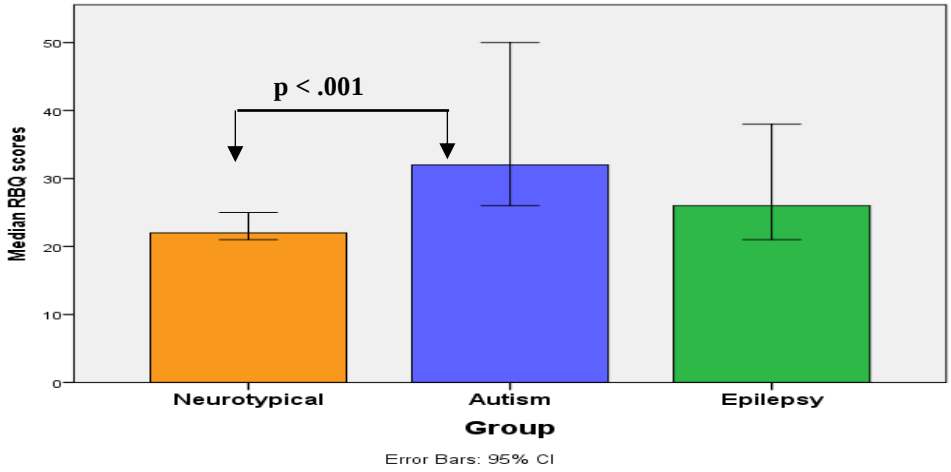

Figure 9**.** *Median RBQ-2 scores and error bars depicting confidence intervals*

Depression and anxiety. An independent samples Kruskal-Wallis found a significant group difference for RCADs total t score, which combines both anxiety and depression scores (H (2) = 20.40, p = .001). Pairwise comparisons found the autism group exhibited significantly higher RCADS scores (Median = 57, Range = 46) compared to the neurotypical group (Median = 36, Range = 21, r = .67; p = .001). When depression and anxiety scores were analysed separately, a main effect was found for both (H (2) = 20.59, p = .001 and H (2) = 16.60, p = .001 respectively). Pairwise comparisons showed compared to neurotypical controls (Median = 34, Range = 17). depression scores were significantly higher for both epilepsy (Median = 49, Range =32, r = -0.45, p = .001) and ASD (Median = 56, Range = 50, r = -.63, p =.001). Additionally, the autism group showed significantly higher levels of anxiety (Median = 58, Range = 43) compared to neuro-typical controls (Median = 37, Range = 24, r = -.61 p = .001). No significant differences for depression and anxiety levels were found between the clinical groups (Figures 10 and 11 respectively). Descriptive statistics for the latter variables are depicted in Table 2 below.

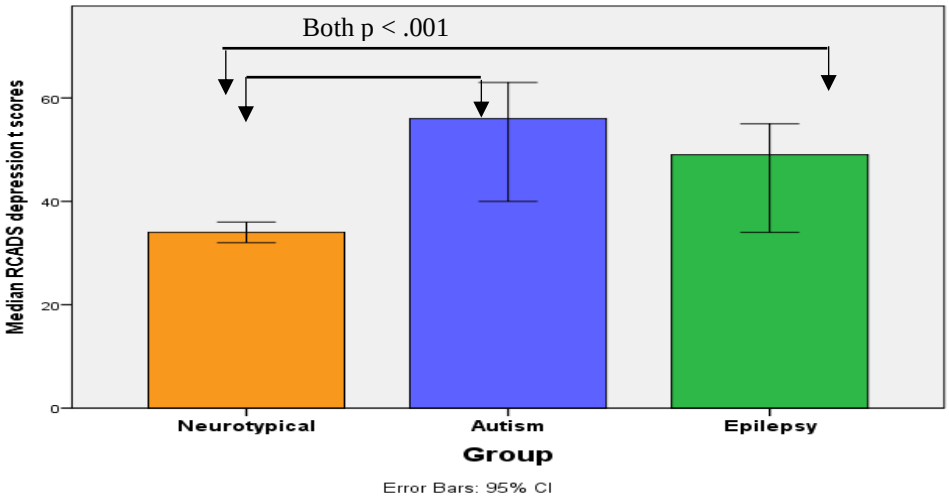

Figure 10**.** *Median RCADS depression scores and error bars depicting confidence intervals*

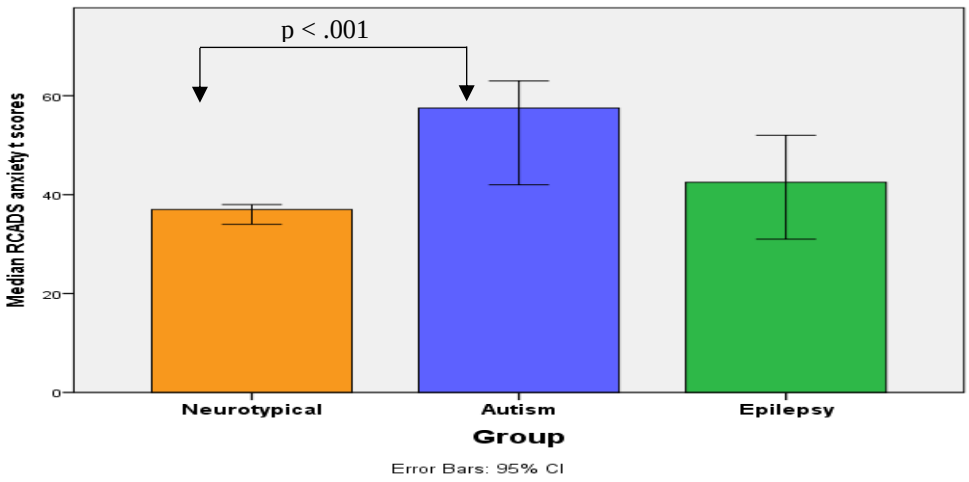

Figure 11. *Median RCADS anxiety scores and error bars depicting confidence intervals*

Table 2. Means and Standard deviations for Group comparison variables

Variable Score	Neurotypical (n=21)		ASD group (n=14)		Epilepsy group (n = 10)		F or H* Statistic	*p*
	Median	Range	Median	Range	Median	Range		
SRS-2 t score	43	19	64	39	48	17	30.01	.001
SRS-RRB t score	44	16	71	47	59	27	26.42	.001
SRS-SCI t score	43	54	62	36	46	15	27.28	.001
RBQ-2	22	15	32	37	26	19	16.36	.001
RCADs total t	36	21	57	46	43	28	20.40	.001
RCADs depression	34	17	56	50	49	32	20.59	.001
RCADs anxiety t	37	24	58	43	43	26	16.60	.001
Age (months)	119[1]	35.58[1]	130[1]	32.76[1]	118[1]	36.04[1]	.520[2]	NS
Matrix reasoning	54[1]	7.52[1]	54[1]	7.21[1]	49[1]	8.68[1]	2.36[2]	NS
Threshold	1.74[1]	.42[1]	1.88[1]	.58[1]	2.26[1]	.61[1]	4.33[2]	.02

Note. [1] As data was parametric the Mean and Standard Deviation was used rather than the Median and Range; [2] refers to when the F ANOVA statistic was reported

Hypothesis 3. Associations between orientation threshold and symptoms of Autism

It was hypothesised that better discrimination scores would be found in those individuals with higher repetitive and sensory behaviour scores. To test this hypothesis, Spearman correlations (Table 3) were conducted between orientation threshold and the SRS-2-RRB subscale, as well as the RBQ-2 and sensory profile-2 subscales; sensory seeking, sensory avoidance, sensory sensitivity and sensory registration. Due to each variable being correlated with 13 additional variables, a bonferroni correction (0.05/13) was applied to control for type 1 errors. Thus, significance level was set at 0.004. The findings showed that none of the variables were significantly associated with orientation threshold.

Table 3. *Correlations between variables*

	Threshold	SRS-2 Total	SRS-SCI	SRS-RRB	RBQ-2	SP-seeking	SSP-2 Avoidance	SSP-2 sensitivity	SSP-2 registration	Age (Months)	Matrix reasoning	RCADS Total	RCADs Depression
SRS-2 Total	.19 [.12-.46]	-											
SRS-SCI	.13 [-.16-.42]	**.97**** [.92-.98]	-										
SRS-RRB	.28 [-.05-.54	**.82**** [.63-.92]	**.71**** [.50-.87]	-									
RBQ-2	.28 [-.07-.61]	**.67**** [.45-.82]	**.63**** [.41-.78]	**.74**** [.56-.86]	-								
SP - seeking	.27 [-.03-.50]	**.51**** [.26-.71]	.45** [.19-.68]	**.74**** [.55-.86]	**.64**** [.39-.80]	-							
SP Avoidance	.14 [-.18-.42]	**.83**** [.70-.90]	**.78**** [.63-.86]	**.80**** [.62-.91]	**.77**** [.61-.87]	**.68**** [.44-.86]	-						
SP – Sensitivity	.19 [.14-.46]	**.82**** [.70-.90]	**.74**** [.57-.86]	**.87**** [.75-.94]	**.69**** [.49-.85]	**.64**** [.39-.83]	**.86**** [.72-.93]	-					
SP Registration	.25 [-.07-.52]	**.63**** [.40-.79]	**.55**** [.30-.75]	**.79**** [.62-.89]	**.71**** [.52-.85]	**.76**** [.56-.86]	**.74**** [.57-.87]	**.75**** [.55-.87]	-				
Age (months)	**-.46**** [-.66-.20]	.05 [-.26-.36]	.03 [-.30-.34]	-.08 [-.37-.25]	**.35*** [-.60-.06]	-.26 [-.53-.04]	-.07 [-.35-.20]	.01 [-.31-29]	-.20 [-.48-.08]	-			
Matrix Reasoning	-.09 [-.41-.24]	.07 [-.36-.23]	.01 [-.36-.23]	.01 [-.30-.28]	.12 [-.18-.39]	.15 [-.15-.44]	.01 [-.28-.29]	.01 [-.28-.30]	.13 [-19-.42]	-.24 [-.53--.07]	-		
RCADs total	.06 [-.24-.39]	**.78**** [.67-.85]	.73** [.60-.82]	**.76**** [.58-.86]	**.72**** [.52-.87]	**.58**** [.32-.77]	**.77**** [.57-.89]	**.73**** [.51-.87]	**.59*** [.32-.79]	.08 [-.22-.40]	-.16 [-.42-.11]	-	
RCADs Depression	.18 [-.14-.49]	**.61**** [.38-.78]	**.51**** [.26-.71]	**.66**** [.44-.79]	**.40*** [.12-.63]	**.56**** [.29-.73]	**.57**** [.32-.74]	**.64**** [.43-.79]	**.52*** [.24-.73]	.16 [.19-.48]	.20 [-.47-.12]	**.76*** [.55-.90]	-
RCADs Anxiety	-.01 [-.30-.33]	**.72**** [.55-.83]	**.62**** [.52-.81]	**.69**** [.47-.84]	**.70**** [.49-.85]	**.54**** [.26-.74]	**.75**** [.52-.88]	**.68**** [.42-.85]	**.52*** [.23-.74]	.04 [-.25-.35]	-.11 [-.39-.16]	**.98*** [.93-.99]	**.70**** [.45-.86]

*Note. *p<0.004. BCa bootstrap 95% Confidence Intervals reported in brackets*

Discussion

The present study was designed to examine visual orientation discrimination thresholds in groups of children with epilepsy, ASD and neuro-typical controls. The first aim was to help reconcile two contradictory theories of ASD, relating to whether individuals with ASD have increased or decreased neuronal inhibition. The second aim was to examine whether certain clusters of ASD symptoms are related to visual orientation threshold scores, in order to increase understanding surrounding of how (if at all) visual perceptual correlates map onto their behavioural counterparts.

Main findings

Orientation discrimination threshold group differences. Based upon Dickinson et al.'s (2014) increased inhibition theory in ASD, it was firstly hypothesised that children with autism would show superior sensory discrimination abilities compared to neuro-typical controls and the epilepsy group. However, the results found no evidence of orientation threshold differences in the ASD group relative to epilepsy and neurotypical groups. This is in contrast to findings by Dickinson et al. (2014), who found AQ scores were significantly correlated with orientation discrimination. Additionally, results do not support Dickinson et al. (2016), who found adults with ASD have lower (i.e. superior) orientation discrimination thresholds. Thus, on the face of it, these findings do not support the inhibition theory, which implies that individuals with ASD will show superior orientation discrimination abilities, indirectly implicating increased neuronal inhibition in ASD individuals. However, one difference between the current study and Dickinson et al. (2014) is that they used an adult ASD population, whereas the current study examined children with ASD. The present study's findings are

consistent with Gomez et al. (2015), who found no significant differences between children with a diagnosis of ASD and matched neurotypical controls. Thus, discrepancies may be due to developmental differences in visual threshold abilities. Indeed, the current study found orientation discrimination was highly correlated with age. It may be that orientation discrimination abilities become more superior in individuals with ASD as they age. Longitudinal studies are required to examine this hypothesis further. Using longitudinal rather than cross sectional designs to track changes in discrimination abilities is supported by Karmiloff-Smith (1998) who argued that, in opposition to innate modularity theories, abnormal and normal development changes over time, based upon complex interactions between genetic, environmental and structural/anatomic brain interactions, as well as brain plasticity. Thus, if the brain organises itself developmentally across time, then research examining developmental disorders should highlight children at risk and follow them longitudinally. Their developmental changes can then be tracked, rather than relying upon an exploration of group averages. This is because group averages fail to take into account developmental differences in visual orientation at aged 7 compared to aged 14 for example.

Another reason to account for insignificant findings may be because the task tapped into motion detection abilities. The distractor and target were presented in quick succession, which made it look like the horizontal lines were moving, especially in trials whereby target and distractor had a high degree contrast (e.g. when the target moved 9^{o} from the reference stimuli). Research has shown that ASD individuals display impaired motor perception (Koh, Milne & Dobkins, 2010) which may have masked superior discrimination abilities in this study. Instead, much research suggests that ASD individuals are superior at detecting static stimuli

(Mottron et al., 2006). It may therefore be useful for future research to examine whether a longer fixation, would reduce the stimuli being perceived as dynamic and moving, instead being viewed as separate static stimuli.

Given that increased neural excitation is identified in epilepsy, it was also hypothesised that the epilepsy group would show significantly poorer sensory discrimination than the typically developing and autism groups. This hypothesis was partially supported. Results showed the epilepsy group had significantly worse orientation discrimination abilities compared to the neurotypical group. However, no significant differences were found between epilepsy and ASD groups. According to Dickinson et al. (2016), reduced orientation abilities are implicated as an index of increased excitation. Thus, children with epilepsy showing poor discrimination abilities are consistent with what would be expected according to Rubenstein and Merzenich's (2003) excitation theory, but also what would be expected according to the inhibition theory also.

The current findings support Grant et al. (2005) who found similar results but within the tactile domain. Thus, there is now a small literature base for poor orientation discrimination abilities in several sensory modalities for individuals with epilepsy. To our knowledge, this is the first study which explored orientation discrimination abilities within the child epilepsy population. Nonetheless, findings are in contrast to Grant et al. (2007), who found no significant differences between control and epilepsy groups on two visual discrimination tasks. Similarly, although Armedo et al. (2009) found adults with epilepsy performed worse than controls on frequency discrimination tasks, this did not reach significance. However, differences between participants included within the latter studies and the current study must be noted. As well as Grant et al. (2005) and Armedo et al. (2009) using an adult

population, they limited their samples to those diagnosed with temporal lobe (a focal) epilepsy. The epilepsy group in the current study was fairly heterogeneous, including both focal and generalised epilepsy types. It could be suggested that specific epilepsy types are more susceptible to orientation discrimination deficits. Though, the inhibition hypothesis would argue that if superior orientation is an index of increased neuronal inhibition, then all epilepsy participants, regardless of type, should perform worse than controls. This is because their underlying neurophysiological causes are all the same; due to over-excitable neuronal networks. In the current study, the epilepsy group was too small to examine subgroups. As discussed earlier, it may also be that age impacts upon discrimination abilities.

Associations between orientation discrimination and specific ASD symptoms. Based on previous research (Baron Cohen, 2009; Chen et al., 2009; Kargos et al., 2015) and in line with the inhibition theory of ASD (Dickinson et al., (2014) it was hypothesised that better discrimination scores will be found in those individuals with higher repetitive and sensory behaviour scores. Findings did not support this hypothesis, with no significant associations found. Although of note, there was a trend towards significant positive correlations between orientation threshold and RRB measures, as well as sensory-seeking. These trends suggest, contrary to our hypothesis, that orientation discrimination abilities are poorer in those with increased repetitive and restricted behaviours and abnormal sensory-seeking patterns of behaviour. Though, caution must be taken when interpreting these findings**.** The results are partially consistent with Chen et al. (2009) who found superior discrimination on the embedded figures task was associated with repetitive behaviours but not sensory behaviours. The findings also similar to Jones et al.

(2009) who did not find a significant difference in auditory discrimination at the group level. Though, Jones et al, (2009) found a subgroup (20% of their sample) who exhibited late onset of verbal communication abilities and high IQ levels showed superior auditory discrimination abilities. This may suggest that if one were to examine the current sample further a subgroup exhibiting superior visual discrimination abilities may have been identified. However, the study did not record data pertaining to the onset of verbal abilities and the limited sample size limits the ability to subdivide the sample further. The study is in contrast to Kargos et al. (2015) who found enhanced auditory discrimination abilities were significantly associated with increased repetitive and restricted behaviours.

Strengths and limitations

A noteworthy strength of the current study is that it aimed to modify a visual orientation paradigm so that it was more suitable for a child population. To our knowledge, it is the first study which has examined visual orientation discrimination across clinical groups of children. However, the paradigm was only piloted on one child. Although the child was at the lower age limit, future studies using the paradigm should pilot it on children of a variety of ages, particularly 6 year olds given this was the lower age limit. Nonetheless, anecdotally, all the younger children included within the study appeared to understand the task except for one (who was subsequently excluded). Moreover, the study aimed to control for a wide range of potentially confounding variables including uncorrected visual impairments, anxiety, mood, age and co-morbid diagnoses such as ADHD and learning disabilities. This ensured that the study measured orientation thresholds more precisely. Additionally, research suggests individuals with ASD exhibit discrepancies between verbal and

non-verbal (performance) IQ, performing better on the latter (Mouga et al., 2016). Given the orientation threshold task was largely based upon non-verbal abilities, the study ensured groups did not significantly differ within this domain. Moreover, the study used reliable and valid measures of ASD symptomology, depression and anxiety.

However, one of the main limitations of the study is that it lacked power, increasing the likelihood of type II errors. Although the ANOVA found a significant result, suggesting it was able to detect a significant difference within the model, it may be why the associations between threshold and ASD symptoms were non-significant. There was a trend towards significance between threshold and repetitive behaviours (both RBQ and SRS-RRB) as well as sensory seeking. An increased number of participants were perhaps required to detect this as a significant difference.

Moreover, much of the data violated assumptions of normality and homogeneity, with a large spread across the data. This resulted in non-parametric tests being conducted when examining the third hypothesis. Non-parametric tests lack statistical power relative to their parametric counterparts, and thus it is possible that significance was missed.

Another limitation of the study is that although the RCADs is recommended for children above 6 years old, norms for 6-7 year olds have not yet been developed. Therefore, the study used the lowest norms available (i.e. 8 years) when extrapolating t scores for 6-7 year olds. To ensure that this did not influence results, raw scores for RCADs total, RCADs depression and RCADs anxiety were correlated with threshold and also inputted into a Kruskal-Wallis. The results were similar for raw and t scores ($r(43) = -.04$, $p = .77$ for anxiety raw and $r(43) = -.23$, $p = .13$ for

depression raw scores), confirming that depression and anxiety levels were unrelated to threshold scores.

A further critique relates to group homogeneity. Both epilepsy and ASD groups showed significantly higher repetitive behaviour scores compared to neuro-typical controls. Although much effort was taken to ensure groups were distinct, such as screening for co-morbid epilepsy and ensuring they did not meet SRS-2 total cut-off scores, this significant result may indicate that the epilepsy group also presented with ASD characteristics. It may have been more useful to exclude those with high repetitive scores. However, due to the small epilepsy sample size and lack of clinical cut-off scores on the RBQ-2, this was not feasible. Cuccaro et al. (2012), using latent analysis found individuals with comorbid epilepsy and ASD showed a higher number of repetitive and sensory behaviours. Given both groups showed this pattern, it could be speculated that some of the epilepsy participants, although not formally diagnosed, fell within this cluster, or at least further along one of the symptom dimensions related to ASD. This hypothesis is also supported by research which has found increased rates of ASD symptomology in epilepsy patients despite having no diagnosis (Clarke et al, 2005; Ryland, Hysing, Posserud, Gillberg & Lundervold, 2012).

It could also be argued that some of the ASD group may have undetected abnormal epileptiform activity. Research suggests for children with ASD, there are two onset peaks for epilepsy; early childhood and adolescence into early adulthood (Bolton et al., 2011; Viscidi et al., 2013). In addition, research has found some individuals with ASD show abnormal EEG epileptiform activity (Chez et al., 2006; Kim, Donnelly, Tournay, Book & Filipek, 2006). Although they were not classified as epilepsy seizures, Chez et al. (2006) argued it is likely a large proportion may go

on to develop clinical seizures during adolescence and adulthood. Thus, some of the ASD group may have been presenting with abnormal EEG activity which could have impacted upon neuronal excitation. Without EEG data it is difficult to determine whether some of the ASD group fell into this category.

Furthermore, the study classified epilepsy based upon parent reports and previous clinician's diagnoses, but did not examine electroencephalogram reports to confirm epileptiform brain activity prior to the study. It could be possible that some epilepsy participants were in remission or over-excitation was controlled with anti-epilepsy medication. Although, previous research has suggested that anti-epilepsy medication does not impact upon discrimination abilities (Grant et al., 2005), caution should be taken given that anti-epileptic drugs affect excitation, the theoretical basis of the main hypotheses examined. Moreover, current study findings indicate that when the epilepsy group was sub-divided into medicated and nonmedicated groups, the medicated group performed worse. This may suggest that medication status may be a significant confounding variable driving orientation threshold difference within the current study. Though, caution should be taken when interpreting these results due to the extremely small sample sizes.

Moreover, the ASD groups median (66) SRS-2 scores fell just within the moderate range (above 66). Only 3 participants fell within the severe range and over half (n = 8) fell within the mild range or below. It may be that the severity of symptoms moderate the impact on superior threshold. Though, a visual inspection of the three participants with ASD scoring within the severe range suggest they perform poorer, with some threshold scores (between 3.13-4.32) double the groups mean threshold score (1.88). Given the sample was opportunistic, those with milder

symptoms may have been more likely to have wanted to take part, compared to those with a more severe presentation of ASD.

Furthermore, whilst the two clinical groups were selected from the same source, neuro-typical controls were recruited via social media and staff emails. Nine (43%) of the neuro-typical control sample were recruited from staff emails in Sheffield, and the remainder recruited from Birmingham. None of the clinical groups were selected from Birmingham, and instead were from Yorkshire. Participant selection may have therefore introduced selection bias, jeopardising the studies internal validity (Downs & Black, 1998). Whilst both clinical groups showed significantly higher levels of depression and the ASD group showed significantly higher levels of anxiety, this difference appears to be representative of the wider ASD and epilepsy populations (Barry et al., 2008; Kanner, 2003a; Gurney et al., 2006; Stewart et al., 2006). Therefore, it could be argued that higher anxiety and depression scores within the clinical groups does not affect the representativeness of the sample. Nonetheless, the study recruited the clinical samples from one neuro-disability and neuro-developmental service within the Yorkshire region and only 9% of respondents invited agreed to take part in the study, thereby increasing non-response bias and limiting generalisability of findings (Fincham, 2008). Additionally, the sample may not be representative of the entire ASD and epilepsy population, as it excluded those with learning disabilities and ADHD. Though, this was felt necessary due to the difficulty of the task and to ensure that orientation discrimination was not influenced by other diagnoses which could increase excitation, potentially interfering with threshold scores.

Moreover, the current study sought to include as many eligible participants as possible. To increase the likelihood of participation, the study provided flexibility in

terms of where they could be tested; either at home, the Ryegate Centre or Sheffield University. It was felt to be important to offer the choice of being tested at home given the previous literature suggesting high levels of anxiety in the clinical groups. However, increased flexibility reduced optimal experimental conditions and increased the likelihood of experimental error. For example, the lighting in each setting was different, which could have impacted upon the experiment.

It was anticipated that the study would have a fourth group, namely participants with co-morbid epilepsy and ASD, to explore whether co-morbid epilepsy influenced the relationship between autism and increased excitation. Having this fourth group would have been a valuable addition in attempting to more precisely reconcile the two conflicting hypotheses. However, during the screening process, it was extremely difficult to identify individuals with co-morbid epilepsy and ASD who did not also meet the threshold for a learning disability. Low intellectual ability has consistently been found to be associated with co-morbid epilepsy and ASD (Bolton et al., 2011; Viscidi et al., 2013). This is supported by the current study which found only 4 potential participants with co-morbid epilepsy and ASD who met inclusion criteria from a large neuro-disability service. Due to such a small sample pool, it was decided not to invite these 4 participants to the study. Thus, it may be beneficial for future studies to widen recruitment to additional services to ensure enough participants for this group can be identified and further resolve theoretical discrepancies.

Theoretical implications

The main aim of the study was to reconcile two theories stipulating increased excitation versus inhibition within ASD, through an exploration of orientation

discrimination thresholds. The findings partially support the inhibition theory, in terms of its prediction that children with epilepsy would perform worse than children with neuro-typical controls, due to increased neuronal excitation. The study has contributed to a small literature base suggesting epilepsy participants show poor discrimination abilities across several sensory modalities. However, given ASD participants did not show superior discrimination abilities, findings would suggest that visual discrimination may not be a reliable index of increased inhibition, at least not within a child population. The finding that inhibition is associated with age, and that other research has found superior discrimination thresholds in adults with ASD implies that more research is required to examine the developmental trajectory of visual discrimination abilities for children with ASD. Similar to findings in the auditory modality (Bertone et al., 2005), insignificant group differences may suggest that visual discrimination abilities differ depending upon task complexity and stimulus type. Therefore, an exploration of differences between cardinal versus oblique and static versus dynamic stimuli should be further researched, as it may be that superior discrimination thresholds are only exhibited for abilities drawing upon low level processing skills. Additionally, the study did not shed any light on further understanding how visual processing is associated with ASD symptomology. Given the heterogeneity of ASD, it may be that examining visual discrimination abilities at the group level is less useful. Indeed, research within the auditory discrimination domain (Jones et al., 2009) found no significant differences at the group level, however found superior discrimination within a subgroup of ASD individuals who had similar characteristics, such as later onset of verbal communication abilities and higher IQs. Thus, theoretical implications may be that research should increasingly focus upon examining subgroups, as well as tracking

developmental trajectories to examine change within visual discrimination abilities across time. A final theoretical implication may be that visual discrimination abilities are moderated by symptom severity. Given the participants in the current study showed mainly mild-moderate symptoms, this may be useful to explore in further research.

Clinical Implications

As the current study did not find significant group differences in visual orientation abilities it cannot claim that participants with autism and epilepsy have different neurological aetiologies. Orientation discrimination tasks do not seem beneficial to use as a marker for identifying individuals who may be more likely to benefit from certain pharmacological interventions. Nonetheless, similar to results found previously, significantly higher rates of depression were found for both clinical groups in the current study. Additionally, the ASD group showed significantly higher anxiety levels compared to the neuro-typical group. One implication may be that individuals with ASD and epilepsy should be routinely assessed for mood and anxiety disorders and evidence based interventions for these populations provided. Moreover, the findings of similar levels of sensory and repetitive behaviours within both ASD and epilepsy groups suggests that clinicians should be increasingly screening for ASD in individuals diagnosed with epilepsy, to ensure autism is not being under-reported within the epilepsy population.

Directions for Future Research

The findings from the study do not support the inhibition theory of Autism, which implies that individuals with ASD will show superior discrimination abilities

compared to neuro-typical controls and epilepsy individuals. However, given the methodological weaknesses of the study, further research is required to attempt to resolve the contradictory literature regarding increased neuronal excitation versus inhibition within ASD. It would be beneficial for future research to conduct a similar study using EEG. This would enable one to better examine epileptiform activity within each participant and examine inhibition more precisely, to be able to directly associate inhibition with sensory discrimination. Moreover, extending recruitment pathways to identify an increased number of individuals with co-morbid epilepsy and ASD who also meet inclusion and exclusion criteria would be valuable in further examining the impact of co-morbid epilepsy on orientation discrimination abilities. This may shed more light on the contradictory findings presented in the current literature base. Future studies should also screen for ASD symptoms within the epilepsy population, having a cut off for all dimensions (social and communication, repetitive and restrictive and sensory behaviours). It may be that superior orientation abilities are present in only a subsample of ASD individuals with certain characteristics (for example particular ages). Using a Latent class model may be one statistical method which could be used to examine ASD subtypes. Future studies would need a much wider sample size with a large representation of each age group, so that developmental influences can be determined.

Conclusion

This is the first study which has attempted to address two contradictory theories of autism using different clinical samples within a child population. The current study did not find evidence to support the notion that superior visual discrimination abilities exist in children diagnosed with Autism. The study did

however provide evidence to support the hypothesis that children with epilepsy show poorer visual discrimination abilities. Additionally, no evidence supporting the association between visual discrimination abilities and particular ASD symptomology was found. Though, the study found that visual orientation thresholds were highly correlated with age. It is important that ongoing research examining superior visual discrimination abilities within individuals diagnosed with autism takes into account the influence of developmental trajectories. Moreover, it may be useful to investigate whether there is a subgroup of individuals with autism presenting with superior discrimination abilities, similar to that found in the auditory domain.

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Appendix A. Health Research Authority Confirmation

NHS
Health Research Authority

Miss Jade Smith
Trainee Clinical Psychologist
Sheffield Health and Social Care
Clinical Psychology Unit
Department of Psychology
Sheffield University
S10 2TN

Email: hra.approval@nhs.net

07 July 2016

Dear Miss Smith,

Letter of HRA Approval for a study processed through pre-HRA Approval systems

Study title:	**Child Visual Perception Sensitivity in the Autism Spectrum - A Quantitative Study**
IRAS project ID:	**196946**
Sponsor:	**Sheffield University**

Thank you for your request for HRA Approval to be issued for the above referenced study.

I am pleased to confirm that the study has been given **HRA Approval.** This has been issued on the basis that the study is compliant with the UK wide standards for research in the NHS.

The extension of HRA Approval to this study on this basis allows the sponsor and participating NHS organisations in England to set-up the study in accordance with HRA Approval processes, with decisions on study set-up being taken on the basis of capacity and capability alone.

If you have submitted an amendment to the HRA between 23 March 2016 and the date of this letter, this letter incorporates the HRA Approval for that amendment, which may be implemented in accordance with the amendment categorisation email (e.g. not prior to REC Favourable Opinion, MHRA Clinical Trial Authorisation etc., as applicable). If the submitted amendment included the addition of a new NHS organisation in England, the addition of the new NHS organisation is also approved and should be set up in accordance with HRA Approval processes (e.g. the organisation should be invited to assess and arrange its capacity and capability to deliver the study and confirm once it is ready to do so).

Appendix B. Parent Information Sheet

Department Of Psychology.
Clinical Psychology Unit.

Doctor of Clinical Psychology (DClin Psy) Programme
Clinical supervision training and NHS research training
&consultancy.

Clinical Psychology Unit
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Parent Information Sheet

Title of the project: A study examining Visual Sensory Processing in Children

We would like to invite your child to take part in a research study. Before you and your child make a decision, it is important for you to understand what the research will involve. Please take time to read the following information carefully. Take time with your child to decide whether or not they wish to take part. You can keep this information sheet.

What is the aim of the study?
This study is part of a clinical psychology doctoral thesis for a student at the University of Sheffield. The study aims to see if there are differences in the way children process visual information and if the differences are related to neuro-developmental conditions such as autistic spectrum disorder and epilepsy.

Why has my child been chosen to take part?
Young people between 6 years and 14 years 11 months are being invited to take part. Children who have a diagnosis of epilepsy and/or autistic spectrum disorder and also children with no known neuro-developmental conditions are being invited to take part.

Are there any reasons for not taking part?
The study poses very low risk of harm or discomfort. However, due to the fact that one of the tasks involves asking children to look at a computer screen for up to 20 minutes, any child with photosensitive epilepsy or suffering from frequent headaches should not take part. Unfortunately, children with uncorrected vision, a diagnosis of Attention Deficit Hyperactivity Disorder or a learning disability will also be unable to take part.

Where will the study take place?

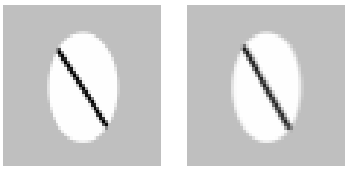
It is up to you where you would prefer the study to take place. We could either come to your home or you and your child could come to the Ryegate centre or to The Psychology Department at Sheffield University. (*Ryegate to be removed for control participants*). If you choose to travel to us we are able to provide a nominal £5:00 per family towards your travel expenses.

What will happen if my child agrees to take part?
Your child will be asked to complete three activities (described below) and you will be asked to complete a set of questionnaires. The study will take about 1 hour. This includes time at the end of the study to ask any questions. You will be able to remain with your chid during the testing if you wish.

What will my child have to do?

1. Your child will have their eyesight tested to make sure that they have no uncorrected visual difficulties which would make the other tasks difficult. The eye test is quick and straightforward and will involve your child naming letters printed in a booklet. If your child usually wears glasses or contact lenses please make sure they are wearing them during the testing session. The eye-test will take about 10 minutes. If their performance on the eye-test suggests they may have an uncorrected visual impairment then they won't take part in the rest of the study. This is because they may not be able to see the pictures in the computerised task properly and it would be unfair to ask them to do so. If it looks as though your child may have an uncorrected visual impairment, then we will let you know and suggest you book an appointment for your child at the opticians. We will also write to your GP to inform them of our findings.

2. Your child will then take part in a computerised visual task where they will see 120 sets of two pictures showing a circle with a diagonal line inside (see below). Your child will then be asked whether the lines in the second picture moved left or right compared to the first picture. This computer task will last no approximately 10-20 minutes and your child will get a break during the task. We will present this task in the form of a computer "game" where the purpose is to help Bob the surfing bear navigate his way back to shore.



An example of one of the sets of pictures your child will see

3. Your child will then complete a non-verbal intelligence task. This will involve your child looking at a set of coloured pictures or patterns with one part missing. Here they will be asked to select the missing piece from four possible solutions. This part of the study will take approximately 10-15 minutes. We will compare your child's score on this test with a normative score obtained from a large sample of the general population. If your child's score is significantly below what would be expected for their age then we will let you know. If we find that the score is significantly below what we would expect, we will also write to your GP.

What will I have to do?

You will be asked to complete three questionnaires which ask questions about your child's mood, anxiety levels and ability to interact socially. These will be completed whilst your child is conducting the computer task. Parents will also be asked to fill in a separate information form which asks questions such as your child's age, gender, ethnicity and current medication.

Will the study help my child?

We cannot promise that this study will help your child, however, the information we collect will help improve the understanding of visual perception in different groups of young people. Ultimately, this piece of research will contribute to a growing body of knowledge regarding the brain basis of autism spectrum disorder and epilepsy.

What will happen with the results of the study?

In most cases you will not receive feedback about your child's performance. However, if the eye test suggests that your child has an uncorrected visual impairment then we will inform you of this and suggest that you make an appointment for your child to see an optician. We will also inform you if your child has performed lower than expected on the intelligence task. If your child performs lower than expected on either the eye task or intelligence task we will also write a letter to your GP informing them of our findings.

If you are interested in the results of the study you may request a summary of the overall findings of the study from the researcher. The scientific results will be published in a peer-reviewed journal and used for educational purposes. All published information will be anonymous.

Will the information be kept private?
Yes. Personal information and results from the study will be kept private. Your child's information will be given a special number that only the research team will have access to, meaning that the information cannot be traced back to your child. All data and information will be kept in a locked cabinet, and destroyed once the study has been completed. Your medical doctors will not be given access to the research information. The only exception would be if your child performed lower than expected on either the eye test or intelligence testing. In these circumstances we would write to your GP and tell them the results of either the eye-test or non-verbal intelligence test.

Can I withdraw from the study?
It is important to stress that you and your child do not have to take part in the study if either you, or they, do not want to. It will have no impact on the care that your child receives. Even if you choose to participate you can withdraw from the study at any time, without having to give a reason. This will not affect your right to any future health care. If you would like to withdraw your data after you have completed the study you can do so up to 72 hours after the study has been completed. However, due to the anonymity of data, we cannot destroy data if you choose to withdraw at a later date, because we will not know which data is yours.

What if I have more questions?
You and your child will be given the opportunity to ask questions throughout the process. If you have any more questions you can contact the research team by email (jsmith31@sheffield.ac.uk) or ring the research support officer on 0112 2226650 who can take a message and I will call you back.

What if I have any concerns about the way the study was conducted?
In the first instance you should speak to the researcher who will do their best to answer your questions. If you remain unhappy and wish to complain formally, you can contact the Research Support Officer Amrit Sinha, (0114 2226650).

All research in the NHS is examined by an independent group of people, called a Research Ethics Committee to protect your rights, wellbeing, safety and dignity. This study has been reviewed and approved by *(to be confirmed)*

Our Research Team:
Jade Smith - Trainee Clinical Psychologist
Dr Elizabeth Milne – Reader in Cognitive Neuroscience at The University of Sheffield
Dr Jack Garlovsky - Clinical Psychologist
Reem Abdal-Sahib - PhD student

Department Of Psychology.
Clinical Psychology Unit.

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Child Information Sheet (under 12 years)

Title of the project: A study examining Visual Sensory Processing in Children

Hello, we would like to ask you to take some tests as part of a project. Please take your time to read the following information carefully with a parent so you know what we would like you to do. Then decide with your parents if you would like to take part. You can keep this information sheet.

What is the aim of the project?

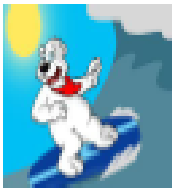
The project aims to see if there are differences in the way children process information they see, and if there are differences for different children.

How long will it take and what will it involve?

The study will take about 1 hour and involves 3 tasks. You will have time at the end to ask any questions.

1. You will have you eyesight tested. This will involve naming different letters. This eye test will take about 10 minutes. If you normally wear glasses or contact lenses you should make sure you are wearing them while we do these tests.

2. If your eyesight is fine, you will then take part in a computer task. In this task you will meet a surfer called Bob who needs your help! He is a bear who loves to surf, but sometimes the waves get too big and he needs help to know which way to turn his surf board. This task will last no longer than 10-20 minutes. We will practice this before you start. To make sure you don't become too tired we will have a break half way through.



Appendix D. Adolescent Information Sheet

3. The final part involves you looking at a set of coloured pictures with

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Adolescent Information Sheet (12 years and over)

Title of the project: A study examining Visual Sensory Processing in Children

Hello, we would like to ask you to take some tests as part of a project. Please take your time to read the following information carefully with a parent/carer so you know what we would like you to do. Then decide with your parents if you would like to take part. You can keep this information sheet.

What is the aim of the project?

The project aims to see if there are differences in the way children and teenagers process information they see, and if there are differences between different people.

How long will it take and what will it involve?
The study will take about 1 hour and involves 3 tasks.

1. You will have you eyesight tested. This will involve naming different letters. This eye test will take about 10 minutes. If you normally wear glasses or contact lenses you should make sure you are wearing them while we do these tests.

2. If your eyesight is fine, you will then take part in a computerised visual task. In the task you will see 70 sets of pictures (see picture below). The pictures will look like circles with a line through. The two pictures will be slightly different. Your task will be to identify whether the line in the second picture moved left or right compared to the first picture. This task will last no longer than 10-20 minutes. We will practice this before you start. To make sure you don't become too tired we will have a break half way through.

An example of one of the sets of pictures you will see

3. The final part involves you looking at a set of coloured pictures with one part missing. Here you will be asked to select the missing piece. This part of the study will take about 10-15 minutes.

4. Whilst you take part in the activities described above, your mom or dad will be asked to complete some questionnaires about you. This will include things like your age, gender and if you ever feel sad or afraid of things.

What if I find the eye test difficult?

If you get below a certain score on the eye test, you will not have to take part in the rest of the study and we will not include any of your details in the study. This is because it might be harder to see the pictures on the computer and it would be unfair to make you do this. We will also let your GP know and recommend that your parents/carer book an appointment with the opticians.

Will my information be private?

Yes. You will be given a number and we will use this number (not your name) when looking at your data. All of the information you give will kept safe in a locked cabinet and destroyed once the project is completed. The only reason I will discuss your results with anyone else is if I think someone may be able to help you do even better on the eye-test or the pattern-matching test. The only people I will discuss this with are your parents/carers and your doctor.

Where will the study take place?

It is up to you where you would prefer the study to take place. We could either come to your home or you and you could come to the Ryegate centre or to The Psychology Department at Sheffield University.

Can I be taken out of the project if I don't want to be in it anymore?

Yes. You do not have to take part if you do not want to. Even if you choose to take part you can withdraw from the study, without giving a reason. If you would like to withdraw your data after you have finished the study, you can do so up to 3 days after the study has been completed.

What if I have more questions?

If you have any questions about the project you can ask your parents/carer to email Jade at jsmith31@sheffield.ac.uk. You can also ask more questions on the day if you decide to take part.

Appendix E – Parent consent form

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The purpose of this form is to make sure that you are happy for your child to take part in the research and that you know exactly what this involves. Thank you for agreeing to take part in this study.

Title of Project: A study examining visual sensory processing in children

Name of Researcher: Jade Smith

	Please initial box
1. I confirm that I have read and understand the information sheet for the above project and have had the opportunity to ask questions.	☐
2. I agree for the data my child and I provide to be recorded on a computer and made anonymous, so that no one can tell it was my or my child's information	☐
3. I understand information will be kept securely so that only the research team can see it	☐
4. I agree for researchers to use data held in my Ryegate medical file (if applicable) as long as the data is annonymised so no one can tell it was my child's information	☐
5. I understand that if I withdraw consent 72 hours after I have taken part in the study, my and my child's data cannot be destroyed as it will be anonymous.	☐
6. I agree for my and my child's data to be used in the final report, as long as it Is annonymised.	☐
7. I understand that participation is voluntary. I am free to say no to taking part in the study at any time without having to give a reason and without affecting future medical care or legal rights	☐
8. I agree to take part in the above research project.	☐

Name of Participant (*or legal representative*)	Date	Signature
Lead Researcher	Date	Signature

To be signed and dated in presence of the participant. One copy to be retained by parent/caregiver and one copy retained by the researcher

Appendix F. Child consent form

The purpose of this form is to make sure that you are happy to take part in the research and that you know exactly what this involves. Thank you for agreeing to take part in this study.

Title of Project: A study examining visual sensory processing in children

Name of Researcher: Jade Smith

Please put your initials in the box

1. I have read and understand the information sheet and have had the chance to ask questions.  ☐
2. I agree for the information I give to be put on a computer and made private, so that no one can tell it is me.  ☐
3. I understand information will be kept secure  ☐
4. I agree for my data to be written in a report, as long as it is private - That means no one will know it is me.  ☐
5. I understand that I can say no to taking part in the study at any time without having to give a reason  ☐
6. I agree to take part in the above research project.  ☐

Name of Participant	Date	Signature	Parent signature
Lead Researcher	Date	Signature	Parent signature

To be signed and dated in presence of the participant. One copy to be retained by parent/caregiver and one copy retained by the researcher

Appendix G. Parent debriefing form

Department Of Psychology.
Clinical Psychology Unit.

Doctor of Clinical Psychology (DClin Psy) Programme
Clinical supervision training and NHS research training
& consultancy.

Clinical Psychology Unit
Department of Psychology
University of Sheffield
Western Bank
Sheffield S10 2TN UK

Participant Code:

Debriefing form - Parents

The aim of the study was to investigate whether certain neurodevelopmental disorders such as autism and epilepsy affect an individual's perceptual discrimination threshold, i.e. their ability to recognise differences between two very similar images. Previous research has shown that adults with autism are particularly good at this type of visual discrimination. We wanted to see if these results were also found in children with autism and what the differences were with other groups of children; those who have epilepsy, those who have both autism and epilepsy and those children without epilepsy or autism. The differences between the different groups of children may help us to further understand the biological mechanisms of autism.

The task where your child had to find the missing picture or pattern was used to make sure that everyone in the different groups had similar levels of cognitive ability, so we can make sure that this does not affect the results. If your child scored lower than expected for their chronological age on this task then I will let you know, and I will also write to your child's GP in case they want to follow this up. The initial eye-test that was completed allowed us to establish that all the children who take part in the study either have normal vision or corrected-to-normal vision (i.e. wearing suitable glasses or contact lenses). If your child scored above the threshold for being included in the study, i.e. if their performance suggests that they may have uncorrected visual impairment then I will let you know and also write to the GP. If this is the case I will suggest that you book an appointment for your child at the opticians so that their vision can be checked by a clinical professional.

Please be aware that the questionnaires you completed are not diagnostic tools and so scores do not represent any kind of disorder or condition. If you would like more information on the questionnaires please feel free to ask. These questionnaires were completed so we can make sure the groups as a whole showed similar anxiety and depression levels, so they do not affect results. However, if the study has raised any concerns about your child's anxiety or mood levels you may want to consider talking to someone at young minds, which provides information on child and adolescent mental health. You can contact them on 0808 802 5544 (Mon-Fri 9:30am-4pm) or visit their website www.youngminds.org.uk.

I will use the information gathered from this study in a research project I am carrying out at University. The findings may also be written up as a research paper and published in a scientific magazine, so that other doctors and researchers can read about what we have found out. However, anything we write about will just talk about how different groups of people do on these tests. Your child's name will never be used, and it will not be possible for anyone to work out that they have taken part in this study based on what I write. As stated previously, the only reason I will discuss your child's information with anyone else is if your child's performance on the non-verbal intelligence test or the eye-test is lower than expected. The only

person I would discuss this information with would be you and your doctor. Your data will be anonymous and is kept separate from the consent forms, so that your name is not associated with any of the information you or your child provide.

Finally, if you feel you would like to withdraw your data from the study you can do so up to 72 hours after the study. You can do this by contacting the principal researcher on jsmith31@sheffield.ac.uk or phoning the research support officer on (0114 2226650). You do not have to provide a reason for withdrawing data. However, due to information becoming anonymous we will not be able to destroy it at a later date if you withdraw consent following the 72 hour period.

The study is being supervised by Dr Elizabeth Milne at the University of Sheffield. If you are interested in hearing more about the work carried out by her research group, including a series of public lectures on autism spectrum conditions, you can visit the web-page http://autismresearchlab.group.shef.ac.uk/index.html

Thank you for you and your child's participation in the study. If you have any further questions please feel free to contact me on the above email address.

Jade Smith, Trainee Clinical Psychologist and Principal researcher

Appendix H – Adolescent debrief form

Department Of Psychology.
Clinical Psychology Unit.

Doctor of Clinical Psychology (DClin Psy) Programme
Clinical supervision training and NHS research training
& consultancy.

Clinical Psychology Unit
Department of Psychology
University of Sheffield
Western Bank
Sheffield S10 2TN UK

Participant code:

Debriefing form –Adolescents

Thank you for taking part in the study!

The aim of the study was to look at whether children and adolescents with a diagnosis of either autism or/and epilepsy look at visual information differently to children without autism or epilepsy. This may help us to understand the biology of autism a little bit better.

The task where you had to find the missing picture or pattern was used to make sure that everyone in the different groups had similar abilities so we can make sure that this does not affect the results.

I will use the information gathered from this study in a research project I am carrying out at University. The findings may also be written up as a research paper and published in a scientific magazine, so that other doctors and researchers can read about what we have found out. However, anything we write about will just talk about how different groups of people do on these tests. Your name will never be used, and it will not be possible for anyone to work out that you have taken part in this study based on what I write. The only reason I will discuss your information with anyone else is if your performance on the eye-test suggests that you may have difficulties with your eyes that nobody else has noticed yet, or if your performance on the pattern matching test suggests that you may have difficulties in learning new information. If this is the case I will mention this to your parent/carer and I will send a letter to your doctor, just to let them know how you have done. If the doctor thinks that there is something that can be done to help, then they may want to talk to you about it in a future appointment.

Finally, if you decide that you don't want to be part of the study anymore, then you can change your mind and I will delete your data. Please tell your parents and they can contact me to take your results out. You do not have to provide a reason for withdrawing your data.

Once again thank you for taking the time to be part in this study!

Jade Smith, Trainee Clinical Psychologist and Principle Researcher

Appendix I. Child (under 12) debrief Form

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Clinical Psychology Unit.

Doctor of Clinical Psychology (DClin Psy) Programme
Clinical supervision training and NHS research training
& consultancy.

Clinical Psychology Unit
Department of Psychology
University of Sheffield
Western Bank
Sheffield S10 2TN UK

Participant code:

Debriefing form –Children (under 12 years)

Thank you for taking part in the study!

The aim of the study was to see whether some groups of children who have epilepsy and autism look at things differently than children who do not. This will help us to understand if there are any differences in how our brains work.

The task where you had to find the missing picture helped to make sure that all the different groups of children had similar abilities. This makes sure that comparing between all the groups is fair.

Remember that your scores on the tasks are private. That means no one will be able to tell which results are yours. Also no one will see your results except for the researchers (researchers are people who create studies and test out different ideas to see if they are true or not). Your results will be kept locked away. The only reason I will discuss your results with anyone is if I think someone may be able to help you do even better on the tests. The only people I will discuss this with are your parents/carers and your doctor.

If you have any worries about anything that you did today tell your parents/carers and they will be able to speak to me about any of your worries or questions. If you decide that you don't want me to use your results you can talk to your parents and I can take them out. You do not have to give me a reason for taking them out, just tell your parents that you no longer want them included.

Thank You!

Jade Smith, Trainee Clinical Psychologist

Appendix J. SRS-2 (removed due to copyright restrictions)

Appendix K. G.P letter for uncorrected visual impairment

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Clinical Psychology Unit.

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Department of Psychology
University of Sheffield
Western Bank
Sheffield S10 2TN UK

Dear Dr X,

Child Y has recently participated in a research study examining differences in visual perceptual abilities between different groups of children. As part of the study child completed the LogMAR visual acuity test to ensure they had no uncorrected visual difficulties. Unfortunately, Y scored above the threshold required to participate in our study, that is, above +0.2 LogMAR (which is equivalent to 6/9 or 20/32 vision). This may indicate the presence of an uncorrected visual impairment. We have informed Mrs/Mr Z/parents of our findings and have encouraged them to book an appointment with an optician. We are also writing to you to inform you of our findings, so you can follow-up if necessary. If you would like to discuss this with me, you can contact me via the university research support officer on 0114 2226650.

Yours sincerely,

Jade Smith
Trainee Clinical Psychologist and Principal researcher

Appendix L. Demographic Questionnaire

Please fill in the following details about your child

Age: ..... **Gender:** FEMALE / MALE (please circle correct response)

Age they started school: .....

GP name and address ……

Do they have a visual impairment: YES / NO (please circle correct response)
If yes, has this been corrected for (e.g. glasses): YES / NO (please circle correct response)

Does your child have a diagnosis of epilepsy? YES/NO
If yes, on average how many seizure days in the past year has your child had? .....

What type of seizures does your child have (e.g. focal/generalised)? ……

When was their last seizure? ……..

Does your child take medication for their seizures? YES/NO
If **YES** please specify ……

If your child has emergency medication for their seizures such as buccal diazepam, when did they last require treatment?
…….

Do they suffer from photosensitive epilepsy YES/NO **(Please circle)**

Do they suffer from any other medical conditions YES/NO **(Please Circle)**
If **yes** please specify ……
……

Current medication you chid takes: ……..

When was the last dose of medication given?.....

Do any family members have a diagnosis of an Autistic Spectrum Disorder?
YES/NO **(please circle correct response)**

If yes please specify what relation they are to your child……

Has your child ever been formally assessed for Attention Deficit Hyperactivity Disorder (ADHD)?
If Yes what was the outcome? ……..
Does your child take medication for ADHD?
What medication?

Does your child suffer from frequent headaches or migraines? YES/NO (please circle correct response)
If YES please give details……

Appendix M. RCADS Example Questions

1. My child worries about things	Never	Sometimes	Often	Always
2. My child feels sad or empty	Never	Sometimes	Often	Always
3. When my child has a problem, he/she gets a funny feeling in his/her stomach	Never	Sometimes	Often	Always
4. My child worries when he/she thinks he/she has done poorly at something	Never	Sometimes	Often	Always
5. My child feels afraid of being alone at home	Never	Sometimes	Often	Always
6. Nothing is much fun for my child anymore	Never	Sometimes	Often	Always
7. My child feels scared when taking a test	Never	Sometimes	Often	Always
8. My child worries when he/she thinks someone is angry with him/her.	Never	Sometimes	Often	Always
9. My child worries about being away from me	Never	Sometimes	Often	Always
10. My child is bothered by bad or silly thoughts or pictures in his/her mind	Never	Sometimes	Often	Always
11. My child has trouble sleeping	Never	Sometimes	Often	Always
12. My child worries about doing badly at school work	Never	Sometimes	Often	Always

Appendix N. Repetitive Behaviour Questionnaire-2 Example Questions

	Does your child:	***Never** or **rarely***	***One** or more times daily*	***15** or more times daily (or at least once an hour)*	***30** or more times daily (or twice an hour)*
1.	Arrange toys or other items in rows or patterns?	**1**	**2**	**3**	**4**
2.	Repetitively fiddle with toys or other items? (e.g. spin, twiddle, bang, tap, twist, or flick anything repeatedly?)	**1**	**2**	**3**	**4**
3.	Spin him/herself around and around?	**1**	**2**	**3**	**4**
4.	Rock backwards and forwards, or side to side, either when sitting or when standing?	**1**	**2**	**3**	**4**
5.	Pace or move around repetitively? (e.g. walk to and fro across a room, or around the same path in the garden?)	**1**	**2**	**3**	**4**
6.	Make repetitive hand and/or finger movements? (e.g. flap, wave, or flick, his/her hands or fingers repetitively?)	**1**	**2**	**3**	**4**

Appendix O. Short Sensory Profile-2 example questions – removed due to copyright restrictions