

Exploring associations and subgroups 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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Abstract

This thesis aimed to explore possible associations and subgroups within the autism spectrum. First, a systematic literature review examining the relationship between anxiety and repetitive and restricted behaviours (RRBs) in children and young people with autism was conducted. Sixteen studies were selected according to inclusion/exclusion criteria. Results showed significant correlations between anxiety and the higher order RRB of insistence on sameness. Evidence suggested that this relationship was mediated by sensory differences and intolerance of uncertainty. Evidence for a relationship between anxiety and the lower order RRBs of self-injurious behaviours and repetitive sensory motor behaviours was mixed and inconclusive. Methodological weaknesses of included studies are discussed as well as clinical implications and recommendations for future research.

Second, the empirical report concerned the development and validation of a brief autism subgrouping questionnaire (BASQ) for children with autism spectrum condition (ASC). Procedures for developing and validating the BASQ are described. The BASQ was found to demonstrate acceptable internal consistency and test-retest reliability. The convergent validity of the BASQ was explored with promising results. The BASQ was then used to explore potential ASC subgroups and showed evidence for the presence of three subgroups within our sample with distinct ASC symptom profiles across the BASQ subscales. The limitations of the study, implications of findings and recommendations for future research are all discussed.

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Section 1: Literature Review

The relationship between restricted and repetitive behaviours and anxiety in autism spectrum condition (ASC).

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Abstract

Objectives. The aim of the review was to examine whether there is a relationship between restricted and repetitive behaviours (RRBs) and anxiety in children with autism, and what factors influence this relationship.

Methods. A systematic literature review was undertaken. Six electronic databases (Web of Science, Science Direct, PsycInfo, PubMed, Medline and Cochrane Library) were searched (August 2017). Search terms related to RRBs, anxiety and autism were used. The search was limited to English language papers only.

Results. Studies reviewed (n=16) found evidence for significant relationships between total RRBs and anxiety. The most robust finding was of a significant relationship between anxiety and the RRB subtype of insistence on sameness (IS). Evidence suggested that sensory processing differences and intolerance of uncertainty mediated the IS-anxiety relationship. Findings were inconclusive for a significant relationship between anxiety and the RRBs of repetitive sensory motor behaviours (RSMB) and self-injurious behaviours (SIB).

Conclusions. This review suggests a moderate relationship between IS and anxiety, which is influenced by sensory processing differences and intolerance of uncertainty. Findings related to RSMB and SIB are inconclusive and require further investigation.

Practitioner points.

- When developing interventions to reduce RRBs in children with ASC, clinicians should assess the potential role that anxiety may play in the development and maintenance of these behaviours.
- Clinicians may find it useful to address the potential influencing factors of sensory processing issues and intolerance of uncertainty in children who present with high levels of anxiety and RRBs.
- The inclusion of anxiety and RRB subtypes within analyses in research is important so that the differential relationships do not get masked within total scores.
- More longitudinal and experimental studies are needed in this area.

Autism spectrum condition¹ is a neurodevelopmental syndrome characterized by difficulties in social communication and interaction skills, alongside the presence of restricted, repetitive patterns of behaviour (RRBs) (American Psychiatric Association, 2013). In addition to these core symptom areas, children with ASC frequently have clinically elevated levels of anxiety (van Steensel, Bogels, & Perrin, 2011).

Evidence suggests that there is an association between RRBs in autism and level of anxiety (Joosten, Bundy & Einfeld, 2009) however the nature of this relationship is currently unclear. Anxiety may be a by-product of ASC symptoms or a moderator of ASC symptom severity (Factor, Condy, Farley & Scarpa, 2016). This current review aims to investigate RRBs and their relationship with anxiety levels in individuals with ASC.

Restricted and repetitive behaviours in ASC

The 5th edition of the Diagnostic and Statistical manual (DSM-V, APA, 2013) has categorised RRBs into four subcategories B1.) stereotyped or repetitive speech, motor movements or use of objects, B2.) excessive adherence to routines, ritualized patterns of behaviour and excessive resistance to change, B3.) highly restricted, fixated interests that are abnormal in intensity or focus and B4.) hyper- or hypo- reactivity to sensory input or unusual interest in sensory aspects of environment. Turner (1999) conceptualized RRBs into two clusters: ‘lower order’ actions (such as DSM-V B1 and B4 symptoms) and ‘higher order’ behaviours (such as

¹ Throughout this paper I will use the term ‘Autism Spectrum Condition’ when referring to a diagnosis of ‘Autism Spectrum Disorder,’ in line with the view that ‘condition’ is a less stigmatizing, more neutral and respectful term (Lai & Baron-Cohen, 2015).

DSM-V B2 and B3 symptoms). Evidence from factor analyses have also supported this categorization, yielding two factors: 1.) 'repetitive sensory motor behaviour' (RSMB) - which includes motor mannerisms, sensory seeking, repetitive use of objects and self-injurious behaviours and 2.) 'insistence on sameness' (IS) behaviours -which encompasses ritualistic habits, compulsions, strict routines and difficulties with change (Cuccaro et al., 2003; Bishop, Richler & Lord, 2006; Szatmari et al., 2006).

Research suggests that level of cognitive functioning and age are associated with variations in the manifestation of RRBs in children with ASC (Gabriels, Cuccaro, Hill, Ivers & Goldson, 2005; Lam & Aman, 2007). Patterns of change over time are thought to differ, with RSMBs reported to either persist at the same level of frequency/severity or improve over time (Kim & Lord, 2010), whilst IS and higher order behaviours are reported to stay the same or to show an increase in frequency/severity over time (Richler, Huerta, Bishop & Lord, 2010).

It is important to note that not all RRBs are regarded as problematic, for some people they may represent a particular strength or skill. However, for many individuals with ASC, RRBs can take up large amounts of time, interfere with the individual's ability to learn and can lead to aggression if disrupted (Grahame et al., 2015). Higher levels of RRB have also been reported as among the most stressful and stigmatizing behaviours for caregivers to manage (Bishop, Richler, Cain & Lord, 2007).

The current literature on interventions for RRB suggests that there are evidence based practices to treat "lower order" behaviours (such as stereotypies) which utilise a behavioural approach. However, there is presently a need to establish more evidence based interventions to address

higher order behaviours (such as IS) and to consider the underlying cognitive mechanisms involved (Boyd, McDonough & Bodfish, 2012).

Anxiety and RRBs

Anxiety disorders in children with ASC are estimated to be at 40% (van Steensel et al., 2011) compared to 27% prevalence in typically developing children (Costello, Egger & Angold, 2005). Severity of anxiety is higher in children with ASC than typically developing children (Kuusikko et al. 2008) and associated with significant functional impairment and reduced quality of life (Wood & Gadow, 2010).

There is a growing body of research reporting that raised levels of anxiety in ASC are associated with a greater presence of RRBs (Gotham et al., 2013; Lidstone et al., 2014; Rodgers, Glod, Connolly & McConachie, 2012a). Current hypotheses for this association are that children with ASC may engage in repetitive behaviours as a ‘coping response’ to reduce anxiety (Joosten et al., 2009) or manage an optimal arousal level (Baker, Lane, Angley & Young, 2008), that the behaviours may potentially elicit anxiety themselves (Sofronoff, Attwood & Hinton, 2005) and/or that anxiety acts a moderator of RRBs, e.g. experiencing anxiety exacerbates RRB (Wood & Gadow, 2010).

Research suggests the RRB-anxiety relationship is complex with anxiety subtypes reported to be differentially associated with the various RRB subtypes (Black et al., 2017; Rodgers et al., 2012a). Atypical sensory features have been associated with both RRBs and anxiety and it has been proposed that RRBs may function as an attempt to cope with sensory-linked anxiety (Joyce, Honey, Leekam, Barrett & Rodgers, 2017).

Intolerance of uncertainty (IU) is another concept linked with the RRB-anxiety relationship. IU is associated with anxiety in the typically developing population, being considered a risk factor for the development and maintenance of clinically significant anxiety (Carleton, 2012). IU involves a negative perception of uncertain situations which results in high levels of stress, worry and avoidance (Dugas, Gagnon, Ladouceur & Freeston, 1998). The higher order RRBs and IU show conceptual similarities, with their shared associated features such as avoidance of unexpected events and the desire to make life predictable and routinized. IU has been implicated in pathways between ASC symptoms and anxiety and with sensory responsivity (Boulter, Freeston, South & Rodgers, 2014; Wigham, Rodgers, South, McConachie & Freeston, 2014).

Rationale for review

RRBs and anxiety are an important feature of ASC to understand because of the impact that both can have on a child's functioning and quality of life. The relationship between RRBs and anxiety in ASC also has clinical implications. If anxiety and RRBs are related, current assessments and interventions might need to be adapted to address both elements and to take into account mediating factors in this relationship. To date, there has not been a review of the literature examining this topic.

Aims

The current review aims to systematically review the available literature concerning the potential relationship between anxiety levels and RRBs in children and adolescents with ASC in order to address the following questions:

- 1.) Is there a relationship between anxiety and RRBs in children/adolescents diagnosed with ASC?
- 2.) What factors influence the relationship between anxiety and RRBs?

Method

The review was carried out in accordance with the ‘Preferred reporting items for systematic reviews and meta-analyses’ (PRISMA) checklist (Moher, Liberati, Tetzlaff, Altman & The PRISMA Group, 2009).

Search strategy

A systematic search of the literature was conducted in August 2017 to identify papers in which the primary research aim was to identify the relationship between RRBs and anxiety in individuals with autism. Six electronic databases; Web of Science, Science Direct, PsychINFO via OvidSP (1806 to August 2017), Medline via OvidSP (1946 to August, 2017), PubMed (up until August Week 2, 2017), and Cochrane Library (up until August week 2, 2017) were searched. The following search terms were used “repetitive” OR “stereo*” OR “restrict*” paired with autism related search terms “autism”, “ASD”, “asperger*”, “pervasive developmental disorder” OR “PDD” AND “anxiety”, “anxious”, “anxiety disorder”, “psychological disorder” OR “comorbidity.” In addition to the database search, reference lists of all included articles were hand searched to ensure that all relevant papers had been identified.

Inclusion Criteria

Studies examining the RRB-anxiety relationship were included if 1.) the child/young person had a diagnosis of an ASC and was aged between 2-

21 years, 2.) they included an anxiety measure, 3.) they included an RRB measure, 4.) they explicitly reported on the association between RRBs and anxiety in their ASC group, 5.) were published in a peer review journal, 6.) were written in English, 7.) were quantitative in design.

The lower age limit was set so as to exclude children younger than the typical age of an ASC diagnosis (Jo et al., 2015). The upper age limit was set at 21 years to allow for variance across countries/studies in the definition of ‘adolescent/young person’ but to exclude studies with adults as the focus.

Exclusion criteria

1.) No single case studies or case series designs.

This decision was based upon the consideration that results from these designs would not provide quantitative statistical data to allow further generalisation of the findings.

Data extraction

Data were extracted from articles that met the inclusion criteria. Table one provides information on methodological characteristics of studies. Table two provides information on the measures used to assess RRBs and anxiety, including validity and reliability information. In order to satisfy the main aims of this review, studies exploring the relationship between total RRB and anxiety are summarised in table three; studies exploring the relationship between anxiety and lower order RRBs are summarized in tables four and five; studies exploring the relationship between anxiety and higher order RRBs are summarised in table six. Lastly, studies reporting on mediating factors in the relationship between anxiety and RRBs are summarized in table seven.

Quality Assessment

An adapted version of Downs and Black (1998) appraisal tool was used to assess the methodological quality of the studies included in this review (n=16). This 27-item checklist covers quality of reporting, external and internal validity and power. It is considered to be a valid and reliable checklist that performs with good internal consistency, test-retest and inter-rater reliability (Downs & Black, 1998).

Items relating to intervention studies were omitted from the checklist because they were not relevant. For correlational studies two items were omitted due to not being applicable. Item 27 (power question) was adapted, with a single point being awarded if the paper reported a sample size power calculation and zero if not. All items carried a possible score of “yes”=1 or “no/unable to determine” =0, except for one question in the reporting section that carried a possible two-point score. The modified quality appraisal tool resulted in correlational studies being assessed on 14 items and between groups studies being assessed on 16 items, with maximum scores of 15 or 17 respectively. A higher score indicates greater study quality.

To enhance reliability four papers were randomly selected to be scored by an independent reviewer (a Clinical Psychologist). Cohen’s kappa was used to assess interrater reliability. Due to interrater reliability being strong ($\kappa = .90$; McHugh, 2012), it was not felt necessary to co-rate any further papers.

Results

The searches retrieved 1,530 articles from which sixteen articles were retained. See Figure 1 for PRISMA flow diagram, detailing search strategy and screening process. Although the age range of interest was 2-21 years of age, one article (Uljarevic, Richdale, Evans, Cai & Leekam, 2017) was included with a sampling age range that went above this bracket (14-24yrs). The judgment to include this paper was based on the fact that the overall sample age range sat largely within the desired range and the mean age was within the age bracket outlined in the inclusion criteria, therefore it was considered a relevant sample to this review.

Quality of included studies

The quality score for each article is provided in table one. The mean score for quality of papers was 71% and the scores ranged from 53-87%, highlighting considerable variation in quality. The most common limitation (14/16 studies) was within the domain of external validity, specifically the representativeness of the samples. Only one of the studies reported a power analysis and 6 of the studies had a sample size of under 50. The appendix shows the quality scores of each paper using the modified Downs and Black (1998).

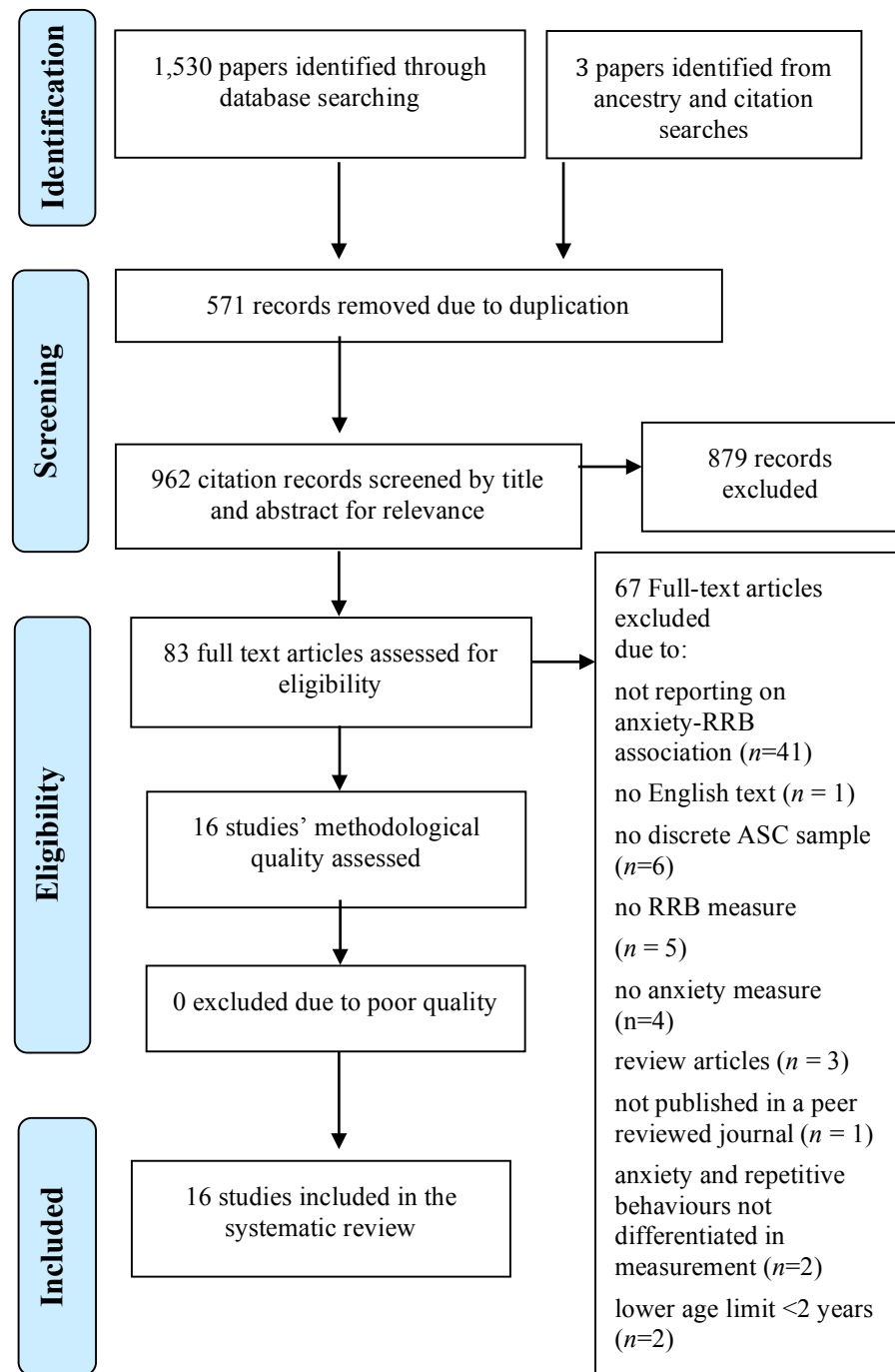


Figure 1. PRISMA flow diagram (adapted from Moher et al., 2009).

Methodological Characteristics

Table one depicts the methodological characteristics of the studies included in this review. Fifteen used a cross sectional design with one longitudinal design also. Fifteen (94%) of studies were published within the last ten years.

Participants. The overall sample included 4,867 participants, of which 4,807 had a diagnosis of ASC. The remaining 60 participants comprised of a typically developing group of children (n=40) and a group of children with William's syndrome (n=20) who were used as comparison groups in two studies. Sample sizes varied greatly from n=19 to n=2341. One study (Teh, Chan, Tan & Magiati, 2017) followed up a subset of a sample (n=54) from an earlier study (Magiati et al., 2016) included in the review.

The ages of participants across studies ranged from 2 – 24 years (overall mean age = 11.25yrs). The majority (11/16) of studies focused on middle childhood to mid/late teens and two studies focused on adolescents and young adults only.

All of the studies reported on the gender of the participants and the proportion of males to females across ASC groups was high (overall mean=83% male, range = 69-92% male). The percentage of males in the comparison groups was 40% and 57%.

Eight studies did not report the ethnicity of their sample. Five studies reported a high proportion of Caucasian participants (70-84%) and two studies reported the majority of participants to be Chinese (77% in both).

Cognitive ability. Cognitive abilities varied greatly across studies and were assessed and reported in a range of ways. Eight papers used standardised measures of cognitive assessment and provided mean IQ scores for their samples. Three studies excluded participants with an intellectual disability (Joyce et al., 2017; Spiker, Lin, Van Dyke & Wood, 2012; Wigham et al., 2015). Two studies stratified participants into high (IQ>70) and low (IQ<70) functioning subgroups for their analyses (Dempsey, Dempsey, Guffey, Minard & Goin-Kochel, 2016; Sukhodolsky et al., 2008). Two papers did not provide any indicators of cognitive functioning for their participants (Lidstone et al., 2014 and Uljarevic et al., 2017).

Anxiety Comorbidity. Eight of the studies included in the review reported on prevalence of anxiety difficulties, these varied greatly from no presence of anxiety disorder (Teh et al., 2017) to 75% of the sample being in the clinical range for anxiety (Wigham et al., 2014). Two studies stratified their sample into clinically ‘anxious’ and ‘non-anxious groups for their analyses (Lidstone et al., 2014; Rodgers et al., 2012a). Anxiety measures used across studies are listed in table two and discussed later in the review.

Table 1

Characteristics of studies

Study and Country	Sample size	Mean age in years (S.D) range	Study design	Main Outcome measures	Quality rating (%)
Black et al. (2017) USA	ASC: 39 TD: 40	ASC: 12.1(2.6), 7-17 TD: 11.0 (3.0), 7-18	Cross-sectional study. Between and within group differences examined. 2 groups – ASC and TD	RRB: RBQ-2 Anxiety: SCAS-P Sensory: SSP	71
Dempsey et al. (2016) USA	ASC: 2341	9 (3.6), 4-17	Cross sectional design, within group (ASC) correlational study.	Anxiety: CBCL, Sensory: ADI-R RRB: ADI-R & RBS-R	87
Factor et al. (2016) USA	ASC: 44	6.91(3.64) 2-17	Cross sectional design, within group (ASC) correlational study.	RRB: RBS-R Anxiety: CBCL Social motivation: SRS-2	80
Gotham et al. (2013) USA	ASC: 1429	10.17 (3.08) 5.8-18	Cross sectional design, within group (ASC) correlational study.	RRB: ADI-R Anxiety: CBCL	80
Joyce et al. (2017) USA	ASC: 19	16.81 (2.39), 13-20	Cross sectional design, within group (ASC) correlational study.	RRB: RBQ-2, RBQ-2A Anxiety: SCAS-P/C Intolerance of uncertainty: IUS-C/P.	53

Table 1 (continued)

Study and Country	Sample size	Mean age in years (S.D) range	Study design	Main Outcome measures	Quality rating (%)
Lidstone et al. (2014) (Study 2) UK	ASC: 49	10.7 (3.1), 3-17.9	Cross sectional design, within group (ASC) correlational study.	RRB: RBQ-2 Sensory: SP Anxiety: SCAP-P & PAS	73
Magiati et al. (2016) Singapore	ASC: 241	10.25 (3.0), 5-17	Cross sectional design, within group (ASC) correlational study.	RRB: DBC-P Anxiety: SCAS-P	80
Rodgers et al. (2012a) UK	Anxious Group: 33 Non anxious Group: 34	Mean 11.2, 8-16 Anxious Group: 11.6 (1.8) Non anxious group: 12.2 (1.7)	Cross sectional, between and within group differences examined. 2 groups – anxious ASC and non-anxious ASC	RRB: RBQ Anxiety: SCAS-P	76
Rodgers, Riby, Janes, Connolly & McConachie (2012b) UK	ASC: 34 Williams syndrome: 20	ASC: 12.17 (2.12), 8-16 WS: 9.40 (3.45), 6-15	Cross sectional, between and within group differences examined. 2 groups – ASC and WS	RRB: RBQ Anxiety: SCAS-P	76

Table 1 (continued)

Study and Country	Sample size	Mean age in years (S.D) range	Study design	Main Outcome measures	Quality rating (%)
Spiker, Lin, Van Dyke & Wood (2012) USA	ASC: 68	9.36 (1.58), 7-13	Cross sectional design, within group (ASC) correlational study.	RRB: YSIS Anxiety: CY-BOCS, ADIS & MASC-P	53
Sukhodolsky et al. (2007) USA	ASC: 171	8.2 (2.6), 5-17	Cross sectional design, within group (ASC) correlational study.	RRB: ADI-R Anxiety: CASI Adaptive functioning: VABS	67
Stratis & Lecavalier (2013) USA	ASC: 72	11.0 (3.3), 5-17	Cross sectional design, within group (ASC) correlational study.	RRB: RBS-R Anxiety: CSI-4 Level of functioning: ABA-II	73
Teh, et al. (2017) Singapore	ASC: 54	T2 -11(2.69), 5-17	Longitudinal	RRB: DBC-P Anxiety: SCAS-P	67

Table 1 (continued)

Study and Country	Sample size	Mean age in years (S.D) range	Study design	Main Outcome measures	Quality rating (%)
Ujijarevic et al. (2017) Australia	ASC: 71	18.71 (2.51) 14-24- 24.81	Cross sectional design, within group (ASC) correlational study.	RRB: RBQ-2, RBQ-2A Anxiety: DSM-5 DAS Self-regulation: A/ATQ	60
Wigham et al. (2015) UK & USA	ASC: 53	12.5 (2.3) 8-16	Cross sectional design, within group (ASC) correlational study.	RRB: RBQ Anxiety: SCAS-P Sensory: SSP Intolerance of uncertainty: IUS-P	73
Williams, Leader, Ammnion & Chen (2015) Ireland	ASC: 109	9.74 (3.17), 6-17	Cross sectional design, within group (ASC) correlational study.	RRB: BPI-S Anxiety: CBCL	67

Note: *ADI-R* Autism Diagnostic Interview – Revised (Rutter, LeCouteur & Lord, 2003), *ADIS* Anxiety Disorders Interview Schedule (Silverman & Albano, 1996), *A/ATQ* Adolescent/Adult Temperament Questionnaire (Evans & Rothbert, 2007), *BPI-S* Behaviour Problems Inventory- Short Form (Rojahn et al., 2011), *CASI* Child and Adolescent Symptom Inventory, *CBCL* Child Behaviour Checklist (Achenbach & Rescorla, 2001), *CSI-4* Child Symptom Inventory – 4th Edition (Gadow & Sprafkin, 2002), *CY-BOCS* Children’s Yale-Brown Obsessive Compulsive Scale (Goodman et al., 1989), *DBC-P* Development Behaviour Checklist – Parent Version (Einfield & Tonge, 2002), *DSM-5 DAS* DSM-5 Dimensional Anxiety Scales (Knapppe et al., 2013), *IUS-C/P* Intolerance of Uncertainty Scale – Child and Parent (Walker, 2009), *MASC-P* Multidimensional Anxiety Scale for Children – Parent Version (March, 1997), *PAS* Preschool Anxiety Scale (Spence, Rapee, McDonald & Ingram, 2001), *RBS-R* Repetitive Behaviour Scale – revised (Bodfish, Symons & Lewis, 1999), *RBQ* Repetitive Behaviour Questionnaire (Turner, 1995), *RBQ-2* Repetitive Behaviour Questionnaire – Second Edition (Leekam et al., 2007), *RBQ-2A* Adult repetitive behavior questionnaire (Barrett et al., 2015), *SCAS-P* Spence Children’s Anxiety Scale – Parent (Spence, 1998), *SRS-2* Social Responsiveness Scale 2 (Constantino & Gruber, 2012), *SSP* Short sensory profile (McIntosh, Miller, Shyu & Dunn, 1999), *YSIS* Yale Special Interest Survey (Klin, Danovitch, Merz, & Volkmar, 2007).

Measures

RRB. Table two shows the range of RRB measures used across studies included in this review, alongside information on their reliability and validity. The majority of RRB measures were parent report with the exception of one self-report measure. The RRB measures used varied with regards to the construct under examination and in some cases were adapted for use. For example, of the three studies using the RBQ-2, two of these (Black et al., 2017; Lidstone et al., 2017) excluded sensory items from their analyses so as to avoid artificially inflating the relation between the RBQ-2 and sensory features. Magiati et al. (2016) modified the DBC-P, removing non-autism specific items and adding autism specific items. The modified DBC-P was also used by Teh et al. (2017). No objective measures were used.

Anxiety. Eight different measures of anxiety were used across the reviewed papers (table two). Two of the tools were used in several of the publications; the Child Behaviour Checklist (used in 4/16 studies) and the Spence Children's Anxiety Scale (used in 8/16 studies). The ADIS, CASI, DSM 5-DAS, MASC-P and SCAS-P all assess a child's anxiety across the main DSM categories of anxiety disorders and are tools that show good reliability and validity for use with a typically developing child population. Whilst the CBCL, CSI-4 and SCAS-P have all been used within ASC populations, they have not been developed specifically for this purpose. Studies varied between using total anxiety scores or specific anxiety subtype scores in their analyses. Spiker et al. (2012) were the only authors to use anxiety measures administered by trained clinicians.

Table 2

Measures used to assess RRBs and anxiety

Measure (author)	Age range	Type of RRB/anxiety examined	No. of items	Type of measure	Reliability and validity	N papers that used measure
RRB measures						
ADI-R (Rutter et al., 2003).	From 2 years	Motor, sensory, rituals/routines, circumscribed interests and self injurious behaviours.	14	Parent report interview	Good to excellent validity and reliability (Rutter et al., 2003).	2
BPI-S (Rojahn et al., 2011)	Not reported.	Self injurious behaviour, stereotyped behaviour and aggressive/destructive behaviour.	30	Informant based behaviour rating	Psychometrically sound properties but test-retest reliability is yet to be examined (Rojahn et al., 2012).	1
DBC-P (Einfeld & Tonge, 2002)	4-18	Disruptive, antisocial, self absorbed, communication disturbance, anxiety and social relating.	96	Caregiver report checklist	Good internal consistency of $\alpha=0.94$, and strong psychometric properties.	2
RBS-R (Bodfish et al., 1999)	Not reported	Stereotyped, self injurious, compulsive, ritualistic, sameness and restricted behaviour.	43	Parent report questionnaire	Factor et al. (2016) report reliability (Cronbach's alpha) as $r=0.77$ for Stereotyped and $r=0.87$ for Sameness behavior.	3

Table 2 (continued)

Measure (author)	Age range	Type of RRB/anxiety examined	No. of items	Type of measure	Reliability and validity	N papers that used measure
RBQ (Turner, 1995)	Not reported	Repetitive movements, sameness behaviour, repetitive use of language and circumscribed interests.	33	Caregiver report	Honey, McConachie, Turner & Rodgers (2012) found 2 reliable and valid factors, sensory-repetitive motor behaviours (.79) and insistence on sameness/circumscribed interests (.85).	3
RBQ-2 (Leekam et al., 2007)	Not reported	Repetitive movements, sameness behaviour, repetitive use of language and circumscribed interests.	20	Caregiver report	Reported to demonstrate good internal consistency for IS and repetitive motor movements (.76/.83) in a sample of children aged 2-17yrs.	3
RBQ-2A (Barrett et al., 2015)	Adults	Repetitive movements, sameness behaviour, repetitive use of language and circumscribed interests.	20	Self report version of RBQ-2.	Good psychometric properties when tested with adults with ASC (Barrett et al. 2015).	2
YSIS (Klin et al., 2007)	Preschool-adolescence	Circumscribed interests	Not reported	Parent report interview	Inter rater reliability reported at $k=.85$, 94% agreement between raters (Spiker et al., 2012).	1

Table 2 (continued).

Measure (author)	Age range	Type of RRB/anxiety examined	No. of items	Type of measure	Reliability and validity	N papers that used measure
Anxiety measures.						
ADIS (Silverman & Albano, 1996).	Not reported	Provides diagnoses of anxiety disorders and a severity score for the primary anxiety diagnosis.	Not reported	Semi-structured interview administered by trained clinician.	Not reported	1
CASI: CSI-4 (Gadow & Sprafkin, 2002) and the ADI-4 (Gadow & Sprafkin, 1998) combined.	Not reported	Generalized anxiety disorder, separation anxiety disorder, PTSD, somatization, social phobia and obsessive-compulsive disorder, specific phobia and panic disorder.	26	Parent report	Satisfactory internal consistency, reliability and validity in community-based, clinic-referred and PDD samples (Sprafkin, Gadow, Salisbury, Schneider & Loney, 2002).	1
CBCL (Achenbach & Rescorla, 2001).	Preschool (1.5-5 years) or School age (6-18yrs)	Social competence and emotional and behavior problems	118	Parent report questionnaire	Strong psychometric properties and has been used among samples of children with ASC (Pandolfi, Magyar & Dill, 2012). Reliability was $r=0.8$ for the anxiety problems subscale (Achenbach and Rescorla 2001).	4

Table 2 (continued).

Measure (author)	Age range	Type of RRB/anxiety examined	No. of items	Type of measure	Reliability and validity	N papers that used measure
CY-BOCS (Goodman et al., 1989)	Not reported	OCD related obsessions and compulsions	Not reported.	Symptom checklist administered by trained clinician	Good internal consistency ($\alpha=.90$). Has maintained consistency with children as young as five (Storch et al., 2004).	1
DSM-5 DAS (Knapppe et al., 2013)	Not reported	Dimensional assessment of anxiety symptoms	Not reported	Screening questionnaire	Not reported	1
MASC-P (March, 1997).	Not reported.	Number and severity of anxiety symptoms.	39	Parent report	Good internal consistency and convergent validity (Wood, Piacentini, Bergman, McCracken & Barrios, 2002).	1
SCAS-P (Spence, 1998)	Preschool (PAS) and parent (SCAS-P) versions	Panic/agoraphobia, separation anxiety, fears of physical injury, obsessive compulsive disorder and generalized anxiety	38	Caregiver report	Acceptable reliability and validity for use with children with ASC (Wigham et al. 2015). Subscales demonstrate good internal consistency (.47-.87) and test retest reliability (.60-.82) with TD children (Spence, 1998).	8

The relationship between RRBs and anxiety

Four studies examined the relationship between total RRB and total anxiety scores and all reported significant correlations (table three), ranging from moderate to strong (according to Evans (1996) classification for interpreting correlational strength). Two studies found significant moderate effects (Lidstone et al., 2014; Stratis & Lecavalier, 2013) of $r=0.41$ and $r=0.56$ respectively. Two studies reported significant strong effects of $r=0.68$ (Joyce et al., 2017) and $r=0.69$ (Rodgers et al., 2012b). Joyce et al. (2017) included self-report versions of measures and found positive correlations between self-reported total RRB and anxiety, $r=0.60$.

Table 3

Relationship between total RRB and total anxiety scores

Study	Analysis	Result
Joyce et al., (2017)	Correlation	Parent report: RBQ total + SCAS total ($r=0.68$, $p=0.001$); Self report: RBQ total + SCAS total, ($r=0.60$, $p=0.032$)
Lidstone et al., (2014)	Correlation	Anxiety positively correlated with RBQ-2 Total score ($r=0.41$, $p=0.004$)
Rodgers et al., (2012b)	Correlation	Significant positive relationship between RBQ total score and SCAS-Parent ($r=0.69$, $p=0.000$)
Stratis & Lecavalier (2013)	Correlation	RBS-R total score and CSI-4 anxiety ($r=.56$, $p<0.05$)

Anxiety and lower order RRBs

Repetitive sensory motor behavior (RSMB). Eight studies examined the relationship between anxiety and the ‘lower order’ RRB domain of RSMB (table four). Results were mixed with four reporting no significant relationship between RSMB and anxiety (Factor et al., 2016; Lidstone et al., 2014; Rodgers et al., 2012a, 2012b), three reporting significant but weak associations (Joyce et al., 2017; Magiati et al., 2016;

Sukhodolsky et al., 2008) of $r=.24$, $r=.38$ and $r=.22$ respectively and one reporting a significant moderate association of $r=.40$ (Wigham et al., 2015). Of note, Magiati et al. (2016) found the strength of the RSMB-anxiety correlation to vary from $r=.20$ - $r=.48$ dependent on the anxiety subtype under investigation.

These different findings are unlikely to be due to variance in the outcome measures used because the RBQ and SCAS-P were used in the majority (6/8) of these studies. Interestingly of the four studies that did report an association between RMSB and anxiety, two had excluded participants with an intellectual disability (Joyce et al., 2017; Wigham et al., 2015) and one reported that higher IQ was associated with anxiety and stereotyped behaviours (Sukhodolsky et al., 2008). These findings suggest that anxiety subtype and IQ level are factors that might mediate the potential relationship between anxiety and RSMB.

Table 4

Relationship between anxiety and RSMB

Study	Analysis	Result
Factor et al. (2016)	Correlation	Anxiety was not found to be associated with RSMB (stereotyped behavior scale) ($\beta=0.10$, $p=0.499$).
Joyce et al. (2017)	Correlation	Parent reported anxiety was significantly and positively associated with RSMB ($r=.79$, $p<0.001$) Self-reported anxiety was positively associated with RSMB but not significantly ($r=0.51$, $p=0.075$).
Lidstone et al. (2014)	Correlation	Anxiety was not correlated with RSMB $r(49) = .24$, $p=.10$.
Magiati et al. (2016)	Correlation	Repetitive speech/behavior score was significantly positively correlated with total anxiety ($r=.38$, $p<.001$) and the subscales of: separation anxiety ($r=.20$, $p<.001$), generalized anxiety ($r=.33$, $p<.001$), panic/agoraphobia ($r=.48$, $p<.001$) and OCD ($r=.42$, $p<.001$).
Rodgers et al. (2012a)	Correlation	No significant relationship between RSMB and anxiety found in the anxious group ($r=.06$, $p=.36$) nor the non-anxious group ($r=.21$, $p=.26$).
Sukhodolsky et al. (2008)	Correlation	Higher levels of anxiety associated with stereotyped behaviours ($\beta=0.23$, $p<.05$).
Wigham et al. (2015)	Correlation	Anxiety significantly positively correlated with RSMB ($r=.40$, $p=.003$).
Williams, et al. (2015)	Correlation	No significant correlations found between anxiety and stereotyped behavior.

Self injurious behavior (SIB). Three studies (table five) focused on the specific ‘lower order’ RRB of SIB and its association with anxiety with mixed results: one reported no significant relationship between these variables (Williams et al., 2015) and two reported a significant association (Dempsey et al., 2016; Stratis & Lecavalier, 2013). However, Dempsey et al. (2016) went on to report that their model only explained 12% of the variance in SIB. Stratis et al. (2013) found that the relationship between SIB and anxiety was moderated by level of functioning, with SIB only predicting anxiety in those with conceptual composite scores 1sd

above/below the mean. The potential moderating effect of level of functioning might be a reason why results are mixed in this category.

Each of these studies employed a different measure of SIB (RBS-R, ADI-R and BPI-R) which also makes comparing findings difficult.

Table 5

Relationship between anxiety and self-injurious behaviour (SIB).

Study	Analysis	Result
Dempsey et al. (2016)	Regression and mediation	Anxiety significantly associated with SIB, for a one unit increase in anxiety, SIB scores increase by 0.03 ($p < 0.001$). However, this model was found to only account for 12% variance in continuous SIB measure.
Stratis & Lecavalier (2013)	Multiple regression	Significant relationship between SIB and anxiety which was moderated by level of functioning.
Williams, et al. (2015).	Correlation	No significant correlations found between anxiety and self-injurious behavior.

Anxiety and higher order RRBs

All of the studies ($n=10$), examining the relationship between the higher order RRB domain and anxiety (table six), reported a significant positive correlation between anxiety and IS ranging from weak to strong ($r=.28-.61$). A significant moderate association ($r=.45-.54$) was the most consistent finding reported (Black et al., 2017; Joyce et al., 2017; Lidstone et al., 2014; Uljarevic et al., 2017).

Eight of the studies used total anxiety scores within their analyses, however two studies (Black et al., 2017; Rodgers et al., 2012a) used anxiety subtype scores and both reported differential associations for IS with the various anxiety subtypes. Consistent across both studies was a positive correlation between IS and separation anxiety ($r=0.54$ and $r=.40$

respectively). Rodgers et al. (2012a) reported no significant correlations between IS and panic/agoraphobia, social phobia, OCD or GAD subscales.

Stratis & Lecavalier (2013) considered a wider range of higher order RRBs within their study and reported IS to be a positive predictor of anxiety but compulsive behavior or restricted interests did not predict anxiety.

Table 6

Relationship between anxiety and higher order RRBs.

Study	Analysis	Result
Black et al. (2017)	Correlation	IS positively correlated with specific phobia ($r=0.50$, $p=0.001$), separation anxiety ($r=0.54$, $p<0.0001$) and social anxiety ($r=0.33$, $p=0.04$).
Factor et al. (2016)	Mediation model	Anxiety was positively associated with sameness behavior ($\beta=0.49$, $p<0.01$).
Gotham et al. (2013)	Correlation	Anxiety and IS were minimally though significantly positively associated with each other ($r=.28$, $p<.001$).
Joyce et al. (2017)	Correlation	Parent report anxiety and IS significantly positively associated ($r=0.60$, $p=0.007$). Self-report anxiety and IS showed a positive but not significant association ($r=0.49$, $p=0.087$).
Lidstone et al. (2014)	Correlation	IS factor (without sensory items) was significantly associated with anxiety ($r=.46$, $p<.001$).
Rodgers et al. (2012a)	Correlation	Anxious sample: IS/circumscribed interests significantly positively associated with total anxiety score ($r=.36$, $p=.03$). Non-anxious sample: IS/circumscribed interests positively associated with total anxiety score but not significantly ($r=.32$, $p=.07$).
Spiker et al. (2012)	T-tests	Children who symbolically enacted restricted interests (RI) showed greater degree of anxiety as measured by the ADIS ($p=.05$) and the MASC-P ($p<.01$) than children who did not.
Stratis & Lecavalier (2013)	Correlation and regression	Ritualistic/sameness behaviour was a significant positive predictor of anxiety ($\beta=.662$, $p<.01$). Compulsive behavior ($\beta=-.010$, $p>.05$) and restricted interests ($\beta=-.175$, $p>.05$) were <u>not</u> found to predict anxiety symptom severity.
Uljarevic et al. (2017)	Mediation model	IS was positively associated with anxiety ($r=.45$, $p<.001$).
Wigham et al. (2015)	Correlation	Anxiety significantly positively correlated with RBQ sameness ($r=.61$, $p=.000$).

Mediating factors in the anxiety-RRB relationship

Sensory processing features. Three studies (table seven) explored the mediating role of sensory features in the anxiety-IS relationship. Black et al. (2017) reported that hypersensitivity mediated 67% of the relationship between symptoms of specific phobia and IS and 57% of the relationship between separation anxiety and IS. Lidstone et al. (2014) reported that the relationship between anxiety and IS was mediated by sensory avoiding and to a lesser degree by sensory sensitivity. Wigham et al. (2015) found significant direct effects from sensory under responsiveness to RSMB ($B=-.39$, $p=.001$) and IS ($B=-.29$, $p=.009$) and from sensory over responsiveness to IS ($B=-.13$, $p=.001$) but with no significant direct effect to RSMB.

Both the Black et al. (2017) and Lidstone et al. (2014) studies removed sensory items from their RRB measure and so we can be confident that their correlation results weren't artificially inflated by overlapping items with the sensory measure however caution must be applied when interpreting Wigham et al's (2015) results due to potential for overlap.

Cognitive mechanisms. Two studies looked at the potential role of intolerance of uncertainty (IU) as a mediating variable (table seven). Wigham et al. (2015) found evidence for pathways involving IU and anxiety from both over and under sensory responsivity to both types of RRB (higher and lower order). Joyce et al. (2017) found both parent and self-reported IU to be significantly positively correlated with anxiety ($r=0.82$ for both). A significant positive relationship was also found between parent reported IU and both RSMB ($r=0.66$) and IS ($r=0.55$). Finally, Uljarevic et al. (2017)

found effortful control and Factor et al. (2016) found low social motivation to mediate the IS-anxiety relationship (table 7).

Table 7

Mediating factors in the relationship between anxiety and RRBs.

Study	Analysis	Result
Sensory processing		
Black et al. (2017)	Mediation	Significant indirect pathway from specific phobia to IS through hypersensitivity (ab=0.33, SE=0.11, 95% CI [0.10, 0.55], p<0.0001), with hypersensitivity mediating 67% of total effect. Significant indirect pathway from separation anxiety to IS through hypersensitivity (ab=0.33, SE=0.12, 95% CI [0.12, 0.59], p<0.0001), with hypersensitivity mediating 57% of the total effect. No relationship observed between sensory hypersensitivity and social anxiety.
Lidstone et al. (2014)	Correlation	IS significantly correlated with low registration (r=-.38, p<.01), sensation seeking (r=-.49, p<.01), sensory sensitivity (r=-.31, p<.01) and sensation avoiding (r=-.49, p<.01). Anxiety was significantly associated with low registration (r=-.40, p<.01), sensory sensitivity (r=-.61, p<.01) and sensation avoiding (r= -.71, p<.01). RSMB significantly correlated with sensation seeking (r=-.42, p<.01) and sensation avoiding.
	Mediation	Both IS (F (1,47) = 12.70, R ² =.196, p=.001) and sensation avoiding (F, (1,47) = 46.81, R ² =.488, p<.001) predicted by anxiety. IS predicted by sensation avoiding (F (1, 47) =15.23, R ² =.229, p<.001)
Wigham et al. (2015)	Correlation	Sensory over responsiveness significantly associated with RSMB (r=-.39, p=.004) IS (r=-.56, p=.000) and anxiety (r=-.35, p=.01). Sensory under responsiveness significantly associated with RSMB (r=-.70, p=.00) and IS (r=-.36, p=.01) but not anxiety.
Cognitive factors		
Factor et al. (2016)	Mediation	Low social motivation mediated relationship between anxiety and IS (M=0.13, 95% CI [0.02, 0.271]).
Uljarevic et al. (2017)	Mediation	Effortful control mediated the relationship between insistence on sameness and anxiety (B=1.62; BCa 95%CI [.59,3.24])
Wigham et al. (2015)	Mediation	Intolerance of uncertainty and anxiety mediated relationships between sensory under-responsiveness and IS (B=.16; LL=-.34, UL=.04) and repetitive motor behaviours (B=.05; LL=.11, UL=.01)
Joyce et al. (2017)	Correlation	Intolerance of uncertainty found to be significantly positively correlated with anxiety (parent report: r=0.819, p=0.001 and self-report: r=0.817, p=0.001) and RSMB (r=0.656, p=0.002) and restricted interests (r=0.553, p=0.014)(parent report only).

Impact of demographic factors

Two studies reported significant correlations between anxiety and age; Wigham et al. (2015) reported a negative correlation and Williams et al. (2015) reported a positive one. Similarly, Gotham et al. (2013) reported a positive association between anxiety and IQ, whilst Williams et al. (2015) reported a negative one. Given these results are conflicting and limited to a small number of studies caution must be applied in their interpretation. The studies use different measures and differ in terms of their sample demographics. However, previous studies have reported anxiety to be associated with higher IQ (Witwer & LeCavalier, 2010) and age (Kuusikko et al., 2008).

Wigham et al. (2015) reported RSMBs to significantly negatively correlate with age but reported no significant relationship between age and higher level RRBs, these findings are in keeping with those from longitudinal research (Richler et al., 2010). Finally, there was some evidence for SIB being predicted by IQ (Dempsey et al., 2013) and adaptive functioning (Stratis & Lecavalier, 2013).

Longitudinal study

Time one repetitive behavior symptoms were found to predict time two (10-19 months later) total anxiety scores in Teh et al.'s (2017) longitudinal study. However, this predictive relationship was fully mitigated by time one anxiety scores when these were included in the regression. These findings suggest that earlier severity of RSMB predicts later anxiety, when earlier anxiety is not accounted for. These results must be interpreted

with the limitations of the study in mind, a relatively small sample size (n=54), short follow up period and a low follow up response rate (33%).

Between groups

Two studies compared the RRB-anxiety relationship in individuals with ASC and comparison groups: of typically developing (TD) children (Black et al., 2017) and children with William's syndrome (WS) (Rodgers et al., 2012b). Both studies reported that the significant positive correlations found between RRBs and anxiety in their ASC samples were not replicated in either of the comparison groups and thus concluded that RRBs role in the development and maintenance of anxiety may be significant in ASC specifically.

Discussion

Relationship between anxiety and RRBs

The primary purpose of this systematic literature review was to explore whether there was a relationship between anxiety and RRB in individuals with ASC.

Studies examining total scores of RRB and anxiety (n=4) found a moderate-strong relationship exists. However, studies examining the associations of specific RRB categories with specific anxiety disorders (rather than total score) found these relationships to be differential.

The most robust finding across studies examining the higher order RRBs was that IS behaviour was positively and significantly associated with

anxiety. 100% (n=10) of studies examining IS reported this association, with the majority reporting a moderate association ($r=.45-.54$).

The lower order RRBs were considered under two further subtypes: repetitive sensory motor behaviour (RSMB) and self-injurious behaviour (SIB) and the findings for both were limited and inconclusive. Half of the studies (4/8) examining a possible RSMB-anxiety relationship reported no significant relationship and half reported a significant weak-moderate association. Magiati et al. (2016) found the RSMB-anxiety association to vary by anxiety disorder.

Evidence regarding a possible SIB-anxiety relationship was limited to three studies. Two studies initially reported a significant relationship, however one went on to report a poor model fit (Dempsey et al., 2016). At this stage there is not sufficient evidence to make a conclusion on the relationship between SIB and anxiety and further research in this area is needed.

The mixed and limited findings within the lower level domain of RRB may be a result of variance in how lower order RRBs are defined and measured, the tendency for studies to use total anxiety scores rather than subtypes and the wide range in individual cognitive functioning levels across studies. Dempsey et al. (2016) suggested that the etiology of SIBs may differ according to functional level and that the strength of predictors would be weakened in analyses that aggregate, rather than stratify individuals with low and high functioning. Additionally, evidence suggests that RSMBs are the result of an interaction between motivational states and environmental events (Holden & Gitlesen, 2008) and thus looking at

behaviours in one setting reported by one individual might not provide a sufficient evidence to understand the complex inter-relationships at play.

If further studies were to confirm that anxiety is not linked to RSMBs or SIBs but instead is only significantly associated with the IS domain then this would suggest that the different RRB domains are associated with different motivators and underlying mechanisms and would have implications for assessment, formulation and approaches to intervention when working with RRBs in children with ASC.

The importance of separating measures of total anxiety into their component anxiety subscales was highlighted by findings that the existence or strength of the RRB-anxiety relationship varied by anxiety subtype and RRB subtype.

Mediating factors in the anxiety-RRB relationship

The secondary aim of this review was to examine what factors influence the relationship between anxiety and RRBs. Findings from the review suggest that sensory processing differences may mediate the relationship between anxiety and RRBs. Evidence was limited by number of studies (n=3) but consistent for a relationship between sensory processing differences (particularly sensory hypersensitivity), anxiety and the higher order RRB of IS. This finding adds some support to arousal theories (Baker et al., 2008) and fits with proposals that hypersensitivity to stimuli may drive a preference for sameness (Baron-Cohen, Ashwin, Ashwin, Tavassoli & Chakrabarti, 2009).

There was less evidence for a relationship between the lower level RRB of RSMB with anxiety but significant associations were reported between RSMB with sensory under/over responsiveness and sensation seeking/avoiding by two studies (Lidstone et al., 2014; Wigham et al., 2015).

This review also found initial evidence for intolerance of uncertainty mediating the inter relationship between the higher order RRB of IS, anxiety and sensory processing differences. If further studies were to corroborate the mediating role of intolerance of uncertainty this might have important implications for formulation and intervention strategies and may open up a new way of understanding RRBs.

Additional findings were that low social motivation and self-regulation, in the form of effortful control, may also mediate the IS-anxiety relationship. However, evidence around these cognitive mechanisms is sparse with only one study on each and so further research is needed before conclusions can be drawn.

Demographic factors such as age, IQ and adaptive functioning were shown in a minority of studies to have significant associations with anxiety and differential relationships to the different RRB subtypes but with contrasting findings in terms of whether these were positive or negative associations. Given the cross-sectional nature of the studies it is difficult to make conclusions on these relationships, beyond stating that they are all factors to be considered in future longitudinal research.

A further significant finding from between group studies is that the link between RRB and anxiety and the mediating influence of sensory sensitivity appears to be specific to ASC and was not replicated in comparison groups.

Methodological Considerations

A major limitation of studies included within this review is their reliance on caregiver reported outcome measures from one informant. Parent rated questionnaires can be subject to report bias in the form of selective recall, social desirability, and influence of informant characteristics (such as parental understanding about anxiety or RRBs) which may all compromise their reliability. Caution must be applied in the interpretation of findings because of the lack of the use of multiple informants or observational methods to corroborate parent report.

Another limitation was the use of measures which have not been standardised for use with children with ASC. Research is beginning to show that some young people with ASC may have an atypical anxiety presentation (Kerns et al.2014), so measures developed for typically developing children may not accurately capture anxiety within the ASC population. We cannot be confident that the measurement tools selected in all of the studies are appropriate and so some caution in interpreting results is warranted.

Consideration must be given to the potential for symptom overlap between obsessive compulsive disorder (OCD) and features of ASC such as compulsive and ritualistic behaviours, which may have led to inflated correlations between measures. Stratis and Lecavalier (2013) removed the

OCD items from their analysis to address this issue and other studies reported on anxiety subtypes other than OCD. The fact that the anxiety subtype of separation anxiety was most consistently associated with IS suggests that the anxiety-IS link is more than a case of ‘symptom overlap’ with OCD. This issue adds further weight to the argument for using anxiety subscale scores rather than total scores in analyses.

The sample size of studies varied greatly and only one study (Factor et al., 2016) reported a power calculation. A number of studies cited small sample size as a limitation and these smaller studies may not have been sufficiently powered to find significant effects. Correlational analyses prevent any conclusions being made about the directions of causality in these relationships.

Demographic factors varied considerably across studies in terms of age, ethnicity, gender, IQ and adaptive functioning and there was a lack of studies conducted using adult samples. Studies were predominantly cross-sectional and often a wide age range of participants were pooled together, making it difficult to consider the possible effect of age and developmental differences on the presentation of RRBs or anxiety.

Eight of the studies included in this review did not report the ethnicity of their sample. Of the seven studies that did report ethnicity, four reported an over representation of Caucasian participants and three reported an over representation of Chinese participants, which may confound results and limit generalisations.

This review is the first review to investigate the potential relationship between RRBs and anxiety in children with ASC. The systematic approach to the search process, critique and synthesis of the literature reduced systemic bias. The use of an appraisal tool and an independent reviewer, ensured that the quality of included studies was examined in a valid and reliable format. However, the review also has some limitations. Only research papers published in English were included and there was no search of grey literature and so the review may be subject to publication bias. A further limitation is the exclusion of qualitative studies. A mixed methods review including qualitative papers may further our understanding of this area and the complex factors contributing to the RRB-anxiety relationship in ASC.

Clinical Implications

Anxiety was found to be a common feature in autism, associated with higher levels of total RRBs, in particular the higher order subtype (IS). Anxiety should therefore be considered in clinical assessment, evaluation and treatment planning, especially when individuals present with difficulties with IS behaviours.

Given the review's findings on mediating factors, it may be useful for psychological interventions to consider and address sensory processing issues and intolerance of uncertainty in children with ASC who exhibit high levels of anxiety and RRBs. Intolerance of uncertainty is currently an increasing part of psychological treatment protocols targeting anxiety in typically developing cohorts (McEvoy & Mahoney, 2012) in which cognitions related to IU are addressed and more flexible behavioural

repertoires are developed. This approach would need adaptation for use with an ASC child population but its development would be worth exploring given the potential role of intolerance of uncertainty in developing and maintaining anxiety and RRBs.

Given the findings that the RRB-anxiety relationship and the influence of mediating factors on this appears to be specific to ASC may also have theoretical implications in that the development of a model of anxiety specific to ASC may be warranted as well as the development of anxiety measures standardised for use with children with ASC.

Future recommendations

Future studies would benefit from including anxiety subscales and RRB subtypes within their outcome measures instead of relying on 'total' scores, as doing so means the differential relationships between the subtypes are missed. Future research focusing on clarifying the RRB subtypes would be of value in making more specific conclusions about possible associations and mediating factors.

Longitudinal research is needed to establish the direction of relation between subtypes of RRB and anxiety as well as to test more complex mediation models and experimental research designs. RRB, anxiety, sensory features and cognitive mechanisms (such as intolerance of uncertainty) each represent targets for intervention and so understanding the complex and dynamic inter-relationships between these constructs is important in order to develop more specific and effective interventions as well as more sensitive outcome measures.

Conclusions

In summary, the current review suggests a moderate relationship between the higher order RRB of IS and anxiety, with initial evidence for sensory processing differences and intolerance of uncertainty being mediating factors in this relationship. Currently, Boulter et al (2014) have tentatively proposed an integrated cognitive model of anxiety for ASC which appears promising in explaining these interrelationships.

The association between the lower order RRBs and anxiety remains questionable with mixed findings in this area. However, future studies including observational measures and incorporating stratification of participants according to cognitive and adaptive functioning would be beneficial in clarifying this potential relationship.

There is currently a paucity of longitudinal and experimental research within this area, which is compounded by the heterogeneity of the samples used within research, the range of measures used to assess repetitive behaviours and the lack of an anxiety measure standardised for use within ASC populations.

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Appendix. Quality checklist scoring for papers included in the review (n=16).

Authors (year)	Reporting							External Validity			Bias			Confounding			Power	Total (%)
	1	2	3	5	6	7	10	11	12	16	18	20	21	22	25	27		
Black et al. (2017)	1	1	1	1	1	1	1	0	0	1	1	1	1	0	1	0	12/17 (71)	
Dempsey et al. (2016)	1	1	1	1	1	1	1	1	1	1	1	1	n/a	n/a	1	0	13/15 (87)	
Factor et al. (2016)	1	1	1	1	1	1	0	1	0	1	1	1	n/a	n/a	1	1	12/15 (80)	
Gotham et al. (2013)	1	1	1	2	1	1	1	0	0	1	1	1	n/a	n/a	1	0	12/15 (80)	
Joyce et al. (2017)	1	1	0	0	1	1	1	0	0	1	1	1	n/a	n/a	0	0	8/15 (53)	
Lidstone et al. (2014)	1	1	1	1	1	1	1	0	0	1	1	1	n/a	n/a	1	0	11/15 (73)	
Magiati et al. (2016)	1	1	1	2	1	1	1	0	0	1	1	1	n/a	n/a	1	0	12/15 (80)	
Rodgers et al. (2012a)	1	1	1	2	1	1	1	0	0	1	1	1	1	0	1	0	13/17 (76)	
Rodgers et al. (2012b)	1	1	1	2	1	1	1	0	0	1	1	1	1	0	1	0	13/17 (76)	
Spiker et al. (2012)	1	1	1	0	1	1	0	0	0	1	1	1	n/a	n/a	0	0	8/15 (53)	
Stratis & Lecavalier (2013)	1	1	1	1	1	1	1	0	0	1	1	1	n/a	n/a	1	0	11/15 (73)	
Sukhodolsky et al. (2008)	1	1	1	1	1	1	1	0	0	1	1	1	n/a	n/a	0	0	10/15 (67)	
Teh et al. (2017)	1	1	1	1	1	1	0	0	0	1	1	1	n/a	n/a	1	0	10/15 (67)	
Uljarevic et al. (2017)	1	1	0	0	1	1	1	0	0	1	1	1	n/a	n/a	1	0	9/15 (60)	
Wigham et al. (2015)	1	1	1	1	1	1	1	0	0	1	1	1	n/a	n/a	1	0	11/15 (73)	
Williams et al. (2015)	1	1	1	1	1	1	0	0	0	1	1	1	n/a	n/a	1	0	10/15 (67)	

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Section 2: Research Report

Development and preliminary psychometric evaluation of a questionnaire
to identify autism subgroups: an exploratory study.

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Abstract

Objectives. The aims of the current study were to 1.) develop a brief and acceptable autism subgrouping measure that could be completed by parents of children with autism spectrum condition (ASC), 2.) carry out preliminary psychometric evaluation of this measure and 3.) explore whether the developed measure could be used to derive autism subgroups with distinct ASC symptom profiles within our sample.

Design. The study employed a cross sectional, quantitative online-survey design.

Method. Phase one of the study involved survey development (including item generation, pre field testing, item refinement and online testing), resulting in the Brief Autism Subgrouping Questionnaire (BASQ). Phase two comprised of data collection. Parents of a child/young person with a diagnosis of ASC (n=260) completed the BASQ at time one and then a smaller subset (n=177) completed the measure at time two. Additionally, time one participants were asked to complete a validated measure as a test of convergent validity (n=225).

Results. The BASQ demonstrated acceptable internal consistency, test-retest reliability and convergent validity. Latent class cluster analysis revealed three subgroups within our sample with distinct ASC symptom profiles across the BASQ subscales.

Conclusions. The BASQ shows promise as a valid and reliable tool for identifying subgroups within the autism spectrum. However, further

psychometric testing is required to further explore construct validity and establish inter-rater reliability.

Practitioner Points

- In future, the BASQ may be used for research purposes to collect data relevant to stratifying children with ASC into more homogenous groups based on ASC symptom profiles.
- The BASQ has the potential for furthering our understanding of subgroups within ASC and to be used in the testing of hypotheses related to the different etiologies, developmental trajectories and responses to interventions of subgroups within ASC.

Autism Spectrum Condition (ASC)¹ is a complex and multifaceted neurodevelopmental syndrome that is characterized by impairments in social communication and social interaction, alongside the presence of restricted, repetitive behavioural patterns, interests or activities (American Psychiatric Association, 2013). Within this unified definition, the severity of clinical presentation is highly variable (Beglinger & Smith, 2001; Castelloe & Dawson, 1993; Persico & Bourgeron, 2006) with individuals with autism showing great variance in terms of behaviour, language ability, cognitive profile and biological mechanisms (Lai, Lombardo, Chakrabarti & Baron-Cohen, 2013). Additionally, the extent to which ASC is accompanied by co-occurring features such as intellectual disability, medical conditions (such as epilepsy) and psychiatric conditions (such as anxiety and mood disorders) varies greatly between individuals (Anagnostou et al., 2014; Cuccaro et al., 2012). The onset, developmental course and response to intervention among individuals with a diagnosis of autism are also factors that are subject to significant individual difference (Beglinger & Smith, 2001; Geschwind & Levitt, 2007; Ousley & Cermak, 2014).

The diverse manifestation of ASC is considered an obstacle to advancements in ASC research and the translation of research into clinical practice (Newsschaffer, Fallin & Lee, 2002). Some authors have proposed that due to the clinical and genetic heterogeneity seen in ASC it would be more constructive to view this condition as ‘the autisms,’ encompassing a

¹ Throughout this report I will use the term ‘Autism Spectrum Condition’ when referring to a diagnosis of ‘Autism Spectrum Disorder,’ in line with the view that ‘condition’ is a less stigmatizing, more neutral and respectful term (Lai & Baron-Cohen, 2015).

range of partially distinct sub-disorders rather than a unitary disorder (Geschwind & Levitt, 2007). Other authors recommend the need to abandon the search for a 'single entity' of autism (Happé, Ronald & Plomin, 2006) and instead work towards identifying subgroups within the autism spectrum (Lai, Lombardo, Chakrabarti & Baron-Cohen, 2013). The identification of more clinically meaningful and homogenous subgroups would potentially allow for improved interpretations of empirical research, the development of more focused interventions and to further progress genotyping studies in this field (Dawson et al., 2002; Deboth & Reynolds, 2017.) Consequently, a current priority for autism research is to establish whether more homogeneous subtypes of ASC can be reliably identified and defined.

ASC and subgroups

A number of research efforts have been aimed at reducing the heterogeneity in the sample populations used in autism research studies by identifying potential subgroups within ASC (Castelloe & Dawson, 1993; Eaves, Ho & Eaves, 1994; Miles et al., 2005; Prior et al 1998; Stevens et al, 2000; Veatch, Veenstra-Vanderweele, Potter, Pericak-Vance & Haines, 2014; Wing & Gould, 1979).

One method of subtyping that has proved popular is the use of cluster analysis (Cuccaro et al., 2012; Hu & Steinberg 2009) in which multiple variables are subjected to exploratory analysis to identify subgroups. This method allows for the analysis of a diverse set of phenotypic variables in order to reveal previously unidentified associations and underlying latent constructs (Cuccaro et al., 2012).

Cuccaro et al. (2012) employed latent class analysis with an ASC data set (n=557) that included 64 individuals with epilepsy. A range of indicator variables were selected based on their relevance to co-occurring autism-epilepsy and included selected items from the Autism Diagnostic Interview- Revised (ADI-R; Lord, Rutter & LeCouteur, 1994), as well as an adaptive behavior composite score taken from The Vineland Adaptive Behavioural Scales – Second Edition (VABS – II; Sparrow, Cicchetti & Balla, 2005). The analysis identified five clusters differentiated by relative difference in scores across the three ADI-R domains (nonverbal communication, reciprocal social interaction and repetitive behavior). These clusters were further differentiated on the basis of other developmental and autism specific indicators (such as age at recognition and developmental milestones). One of the clusters was defined by a high rate of epilepsy (29%), earlier age of diagnosis and high rates of repetitive object use and unusual sensory interests.

In another study, Hu & Steinberg (2009) carried out cluster analysis on 123 item scores from the ADI-R for a large sample of children with ASC (n=1954). They identified four phenotypic clusters based on similarities in severity indicated by scores across items. Subsequent gene expression profiling using the same dataset revealed that these subgroups were also associated with distinct gene expression profiles, providing support to the idea that the identification of subgroups is relevant to the study of genetic etiology (Hu et al., 2009). Veatch et al. (2009) also reported evidence for genetic contributions to subgroups. In their study they used multiple sources of behavioural and biomarker data to subgroup individuals with ASC and found significant familial clustering and more

similar genotypes within subgroups when compared to the non subgrouped data set in their study.

To date, a growing number of studies have used cluster analytic methods to identify potential subclasses of ASC based on multivariate phenotypic variables. Whilst the majority of these studies report between two to four subclasses of autism (Eaves et al., 1994; Fein et al., 1999; Prior et al., 1998) others have proposed up to fifteen potential subclasses (Veatch et al., 2014). Cluster analyses and their outputs are dependent upon the type and number of variables included in the analyses and the characteristics of the sample used in the study (Beglinger & Smith, 2001) and so these factors are likely to account for the range of findings in the literature. One general and consistent finding has been that when developmental level is explored, intellectual functioning (IQ) is found to be one of the strongest indicators of subclass (Fein et al., 1999; Spiker, Lotspeich, Dimiceli, Myers & Risch, 2002).

In contrast to the multivariate approaches other studies have focused solely on identifying subclasses using variables that represent the core symptom domains of ASC, e.g. social and communicative impairment and restricted interests and repetitive behaviour. Georgiades et al. (2013) used a factor mixture modelling approach to derive subgroups from item scores on the ADI-R for a sample of 391 children with autism. They found that the two symptom dimensions of social communication deficits (SCD) and fixed interests and repetitive behaviours (FIRB) could be used to stratify children with ASC into three homogeneous subclasses. Two of their classes represented a severity gradient in that both of the classes showed

greater impairment in SCD than FIRB, with one class more severely affected across both symptom domains than the other. However, a third class demonstrated a reverse profile, with the lowest scores on the SCD relative to higher scores on the FIRB domain.

Greaves-Lord et al. (2013) carried out latent profile analysis on the six subscale scores of the Children's Social Behaviour Questionnaire (CSBQ; Hartman, Luteijn, Serra & Minderra, 2006) using data obtained from a sample (n=949) of individuals with a diagnosis of Pervasive Developmental Disorder Not Otherwise Specified (PDD-NOS). Six subclasses were identified which they were able to discriminate on the basis of scores on social communication and stereotyped behaviours. These classes differed in both symptom severity and in the relative pattern of subscale scores indicating that they represented distinct phenotypic profiles and not just differences in severity of presentation within the spectrum.

Elevated levels of restricted and repetitive behaviours (RRBs) relative to other ASC symptom domains, have been reported as one potential subgroup marker within subtyping studies (Georgiades et al., 2013; Greaves-Lord et al., 2013). Additionally, a number of studies have reported significant correlations between the ASC symptom domain of RRBs with atypical sensory responses (Boyd, McBee, Holtzclaw, Baranek & Bodfish, 2009; Chen, Rodgers & McConachie, 2009; Gabriels et al., 2008; Gal, Dyk & Passmore, 2010). Gabriels et al. (2008) examined the relationship between RRBs and sensory responses in a sample of 70 children and adolescents with ASC and reported that RRBs significantly co-occur with atypical sensory responses (as measured by the Repetitive

Behaviour Scale-Revised; Bodfish, Symons, Parker & Lewis, 2000 and the Sensory Profile 2; Dunn, 1999) and that these associations remained significant even after overlapping items were removed from measures. Gabriels et al. (2008) suggested that a subgroup of children with ASC appeared to exist in their sample (n=70) with consistently high rates of difficulties with both RRBs and sensory processing.

Overall these studies provide evidence suggesting that ASC can be partitioned into empirically meaningful groups based on similarities across a wide range of features and that the search for genetic etiology is improved when working with more homogeneous samples (Hu & Steinberg, 2009, Veatch et al., 2014). The results also suggest that different subclasses of ASC are represented by distinct patterns of relative ASC symptom severity in the areas of social communication/interaction and RRBs. These findings highlight the potential for children with ASC to be stratified into more homogenous subgroups, based on their relative levels of symptom severity across the symptom dimensions of ASC.

Current Measures

To date the only validated measure designed for the purposes of subgrouping children with autism is the Wing Subgroups Questionnaire (WSQ; Castelloe & Dawson, 1993). This questionnaire was developed to classify children into one of Wing's three hypothesized social subgroups, named 'aloof', 'passive' and 'active-but-odd' (Wing & Gould, 1979). However, limitations of this subgrouping measure include its reliance on clinical observation for subgroups (rather than empirical evidence), its focus solely on quality of impairments in social interaction as a subtyping

scheme (Borden & Ollendick, 1994) and the mixed findings in terms of the validity of Wing's social subtypes on which it was based (Volkmar, Cohen, Bregman, Hooks & Stevenson, 1989).

More recently, due to the wide range of indicator variables and the lack of a validated measure designed specifically for the purpose of subtyping, studies tend to use a combination or variation of diagnostic/screening measures to collect the range of information needed in order to stratify children into more homogeneous groups. These measures typically include any combination of the following: the Autism Diagnostic Interview-Revised (ADI-R; Lord et al., 1994), the Repetitive Behaviour Scale (RBS; Bodifsh, Symons & Lewis, 1999), the RBS-Revised (RBS-R; Bodfish et al., 2000), the Vinelands Adaptive Behaviours Scale (Sparrow, Balla & Cicchetti, 1984), the Autism Diagnostic Observation Schedule (Lord et al., 1989), the Social Communication Questionnaire (Rutter, LeCouteur & Lord, 2003) or the Social Responsiveness Scale (Constantino & Gruber, 2005).

Scores from the ADI-R (Lord et al., 1994) have been used most consistently within studies that have attempted to identify potential ASC subgroups but the number of items and types of items included has varied between studies, with some studies using subscales in the analyses and others including some or all of the individual items.

Limitations of using this current battery of measures are that they are long, time consuming and in some cases require a trained professional to administer them. Since the content of items included in the analyses of studies can impact on the results obtained, the lack of a standardized

measure is an important source of variability amongst research outcomes into potential autism subgroups.

Given the need for future large-scale studies using both larger study samples and longitudinal repeated measures methodology it seems pertinent that a reliable and valid measure is developed that can capture information on the wide range of identified indicator variables in a brief, acceptable and standardized format.

Aims

The aims of the current study were to 1.) develop a brief and acceptable autism subgrouping measure (the Brief Autism Subgrouping Questionnaire – the ‘BASQ’) that could be completed by parents of children with ASC and would capture information on the core ASC symptom domains as well as on potential subgrouping indicator variables, 2.) carry out preliminary psychometric evaluation of the BASQ and 3.) to explore whether the BASQ can be used to stratify children with ASC into more homogeneous subgroups based on symptom severity and if so, to then define and characterize these classes.

Specific hypotheses were:

H¹ Consistent with findings in the literature of a significant correlation between RRBs and sensory processing difficulties (Gabriels et al., 2008), it was hypothesized that the BASQ scores for items assessing the ASC symptom domain of RRBs would correlate positively with scores on a validated measure of sensory processing (thus demonstrating convergent validity).

H² Conversely, it was hypothesized that the BASQ scores for items assessing social interaction and communication would correlate negatively with scores on a validated measure of sensory processing (thus demonstrating divergent validity).

H² In line with current research (Georgiades et al., 2013) we hypothesized that we would be able to discriminate subgroups with distinct ASC symptom profiles using the BASQ.

Further information on how these aims were addressed and the specific hypotheses tested is described in more detail in the method section below.

Clinical and Theoretical Implications

The existence of a measure which is brief yet comprehensive and can be completed by the parent at home/online in a reasonable amount of time will allow not only screening for large scale and longitudinal studies to be carried out with ease but has the potential to provide a simple and standardized way of stratifying children with ASC into more homogeneous groups allowing more meaningful research into the etiology of autism and more focused interventions for ASC to take place.

Method

Design

The study employed a cross sectional, quantitative online-survey design that consisted of two phases. In phase one an online survey (the Brief Autism Subgrouping Questionnaire - 'BASQ') was developed through a process of item generation, face to face pre field testing (in the form of

cognitive interviews), item refinement and online testing. In phase two data was collected from a clinical sample using online survey software (Qualtrics, 2005) in order to validate the BASQ.

Phase 1: Brief Autism Subgrouping Questionnaire (BASQ)

Development

Item generation. The objective of the BASQ was to accurately capture information on the wide range of potential subgrouping indicator variables currently identified in the research literature.

Item generation consisted of a comprehensive review of published literature on autism subgrouping. This review identified the following potential subgroup indicator variables: presence of non-febrile seizures/epilepsy; language acquisition, type of onset (regression history), age of autism diagnosis, gastrointestinal dysfunction, family history of autism, and presence of psychiatric disorders (Cuccaro et al., 2012; Doshi-Velez, Yaorong & Kohane, 2014; Georgiades et al., 2013; Greaves-Lord et al., 2013; Hrdlicka et al., 2005; Ingram, Takahashi & Miles, 2008; Miles et al., 2005; Ming, Brimacombe, Chaaban, Zimmerman-Bier & Wagner 2008). These findings informed the creation of a demographic and medical/developmental history section to the questionnaire (Section 1) to collate information covering these potential subgrouping markers as well as potential confounding variables.

Distinct profiles of autistic traits are reported in the literature as a potential method of stratifying children with ASC into homogenous subgroups (Georgiades et al., 2013; Greaves-Lord et al., 2013), with relative levels of functioning in social interaction, social communication

and restricted and repetitive behaviour domains effectively discriminating potential subgroups. Analyses of an existing comprehensive data set were carried out to identify which measures, and more specifically which items, were most reliable at discriminating potential subgroups. These analyses, combined with findings from the literature on autism subgroups, identified key autistic traits from each symptom domain to be included in the measure.

Existing validated measures used to capture information on performance in these domains were then consulted (the ADI-R: Lord et al., 1994 and the RBS: Bodifsh et al., 1999) and were used as a guide to develop items aimed at capturing information on the core ASC symptom domains. This resulted in a 12-item behaviour and skills section to the questionnaire (BASQ: Section 2). A list of items from validated measures which were adapted and used to guide the development of the BASQ items can be found in Appendix A.

BASQ content coverage. The resulting questionnaire comprised of two subsections designed to be completed by the parent/caregiver of a child with an ASC diagnosis.

Section one: covering demographic information (child's age, gender, ethnicity, socioeconomic status by postcode), child's medical and developmental history (seizure history, regression history, language delay/loss, current medication, information on visual/hearing impairments, presence of associated developmental and mental health conditions) and family history (diagnosis in blood relatives of ASC and associated

developmental and mental health conditions). This section comprises of a mixed format of multiple-choice questions and open text responses.

Section two: 12 items targeting the two domains of ASC symptoms (social/communication deficits and fixed interests and repetitive behaviours) as proposed in the DSM-V (APA, 2013). Items 1-6 cover skills in non-verbal communication and social interaction and items 7-12 cover difficulties relating to restricted and repetitive interests and behaviours. Respondents were asked to score items using a Likert ordinal scale indicating the frequency in which these skills/behaviours are observed in the child. The scale ranged from 1 ('My child never does this') to 5 ('My child always does this'). Total score was the sum of the 12 items (after first 6 items have been reverse scored), with a range 12-60. A Likert scale was chosen in order to adequately capture variance in the domains being assessed.

Cognitive Interviews. To assess the content validity of the BASQ a cognitive interviewing approach was employed with both a clinical parent sample (n=4) and a non-clinical parent sample (n=5). The clinical sample were recruited through an existing database of families in the Sheffield area who had registered their interest in taking part in research into autism. The non-clinical sample (n=5) were recruited via an advert in a local community centre in the Leeds area. Demographic data for these samples can be found in Appendix B.

A cognitive approach known as the Three-Step Test-Interview (TSTI) method (Hak, van der Veer & Jansen, 2008) was used that comprised of the following steps: 1.) observing the response behaviour and

concurrent verbalization of thought processes (“thinking aloud”) of respondent whilst completing the BASQ questionnaire; 2.) carrying out some follow up probing with the respondent (aimed at clarifying actions or thoughts observed during the response process) and 3.) completing a debriefing interview with respondent (aimed at eliciting experiences and opinions on completing the BASQ). The interviews were conducted either at the participants’ homes or within a room at the University of Sheffield (decided by each participant). Each participant was interviewed once and field notes were made during every interview to document the participant’s verbalizations, observed behaviour and responses to follow up probing and debriefing. The interviews lasted approximately 60-90 minutes. Analysis was performed at item level with comments, behaviours and problems labelled and grouped into categories (e.g. ‘problem with response options’, ‘problem with question wording’ etc.).

Item refinement. Data and feedback from these cognitive interviews identified problem items and prompted revisions to be made to the first version of the BASQ. Across sections one and two of the BASQ, one item was removed due to yielding inconsistent and unreliable data (savant skills item), five items had alterations made to their formatting, five items had additional response options incorporated and four items were refined in terms of the wording and examples provided to improve clarity and understanding.

Online development. Once the final refined versions of the BASQ section’s one and two (Appendices C & D) had been agreed, an online survey version was created using the survey software Qualtrics (Qualtrics,

2005). Static pages incorporating study information (Appendix E) participant information sheet (Appendix F) and consent form (Appendix G) were created in addition to active web pages used to capture individualized data from respondents. 'Skip logic' was employed within the survey so that certain responses would lead to respondents skipping questions that were not applicable to them and to also ensure that the respondents completed the age appropriate version of the sensory profile. The completed online survey was then piloted by a small group of parents (n=4) to identify any difficulties with technical aspects of the survey, such as errors in the flow. Necessary amendments to the programming of the survey were made based on this feedback.

Phase 2: BASQ piloting

Recruitment. Participants were recruited over a six-month period (between October 2016 and April 2017) through a variety of online methods. A statement detailing the purpose and nature of the study, contact details for the researcher and an active URL link to the survey were distributed via: (a) emails out to volunteers that had signed up to the Sheffield Autism Research Lab database (b) emails out to relevant third sector/charity organisations across the UK (specifically targeting parent groups/support) and (c) sharing on specific autism related (non-NHS) social media platforms (relevant Facebook group pages and Twitter accounts). On accessing the survey link, participants were provided with participant information followed by a consent declaration page with tick boxes to indicate consent.

A snowballing approach (Goodman, 1961) was used to maximise recruitment via the social networks of respondents. A large number of specialist third sector charities agreed to share the study information and survey link on their web pages and to distribute it via e-newsletters and emails to parents/carers. An online specialist autism parenting magazine agreed to share the study within their e-publication and the study was promoted on a web page dedicated to autism research.

All participants at T1 were asked to provide their email address if they were happy to be contacted at T2 to complete the BASQ again. At T2 (3 months after T1) an email containing a link to the T2 survey was circulated amongst those that had consented and two follow up reminder emails (a week apart) were also scheduled to send should the T2 survey not be completed within two weeks of the original email. The T2 survey comprises of section 2 of the BASQ ('Behaviours and Skills' section) alongside some basic demographic questions for identification purposes only.

Validation Measure

The validity of the BASQ was assessed by looking at whether it correlated with another measure to the degree expected based on theory and related empirical research. Previous studies have described the co-occurrence of restricted and repetitive behaviours with atypical sensory responses (Boyd et al., 2009; Boyd et al., 2010; Chen et al., 2009; Gabriels et al., 2008; Gal et al., 2010) and significant correlations have been found between scores on the Sensory Profile with the Repetitive Behaviour Scale-Revised (Gabriels et al., 2008; Inada et al., 2015). The BASQ utilizes

questions developed from the RBS-R scale to form 6 items covering the ASC symptom domain of RRB and so we hypothesized that these items would correlate with scores on the Sensory Profile 2 (Dunn, 1999).

Therefore, in order to assess validity of the BASQ, participants were also asked to complete the Sensory Profile 2 (Dunn, 1999) which was incorporated as an additional online measure that participants were directed to, upon completion of the BASQ.

Sensory Profile 2 (SP2)

The Sensory Profile 2 (Dunn, 1999) is a 125-item, standard caregiver questionnaire of the effect of sensory processing on the child's ability to function in daily life. Caregivers are asked about the frequency in which the child engages in a list of behaviours in response to sensory events. Item responses occur on a five-point Likert rating scale (from 1 corresponding to 'almost never' to 5 corresponding to 'almost always'). Normative data for the sensory profile were obtained from 1037 typically developing children ages 3-10 years. The developers of this measure have collected data with children with ASC ages 3–17years (Dunn, 1999). The Sensory Profile provides two sets of standard scores depending on how the items are clustered: (1) domain scores (Sensory Processing, Sensory Modulation, Behavior and Emotional Response) and (2) factor scores (nine empirically derived factors). This study used total raw score (sum of the domain scores) in all analyses.

Sample size

Many recommendations regarding sample size in factor analysis have been made, but none are founded on a strict theoretical or empirical

basis. The most widely accepted rule uses the ratio of the number of subjects (N) to the number of items (p). A 10 to 1 ratio for each item has been recommended in the literature (Floyd & Widaman, 1995; Nunnally, 1978). Therefore, in order to validate our 12 item behaviour and skills scale (section 2 of the BASQ) we aimed to recruit a minimum of 120 participants to be sufficient in ensuring a robust factor structure.

Currently, there is no consensus regarding the minimum sample size required for latent class cluster analysis (LCCA). Power in LCCA is thought to be a balance between the number and quality of indicators, the structure of latent classes and the existence of covariates (Wurpts & Geiser, 2014). Larger samples ($n > 500$) have been recommended by some authors (Finch & Bronk, 2011) and small samples ($n < 100$) are recommended to be interpreted with caution (Wurpts & Geiser, 2014).

Participants

A total of 260 parents/caregivers of children completed the BASQ in full in respect to their child at time point one (T1). Core inclusion criteria were that the parents/caregivers had English as a main language and that their child was: (a) aged between 4 years and 15 years, 11 months at time of T1 completion and (b) had a diagnosis of an autism spectrum condition (inclusive of atypical autism, pervasive developmental disorder-not otherwise specified, Asperger's syndrome and childhood disintegrative disorder diagnoses). Participants who completed the measure were Mother ($n=234/90\%$), Father ($n=9$), Foster/Adoptive Mother ($n=8$) and other primary caregiver ($n=9$). The age range of children were 4 – 15 years (mean age 9.7yrs) and 70% were male. The ethnicity of the sample of

children was 240 (92%) identified as white British and 20 (8%) identified as other (including Indian, Pakistani, black African, white and black Caribbean, and white Asian). Fifty-two (20%) of the 260 participants were diagnosed with co-morbid mental health difficulties including anxiety, depression, attachment difficulties and OCD. Ninety-eight (38%) of the participants had a blood relative with a diagnosis of ASC, six (2%) had a diagnosis of epilepsy and seventy (27%) had a history of language regression. An index of multiple deprivation (IMD) was calculated using the English Indices of Deprivation (Ministry of Housing Communities and Local Government, 2015) for participants who had provided their postcode (n=234). This showed 21% of the sample with an IMD within deciles 1-3, 38% with an IMD within deciles 4-7 and 41% with an IMD within deciles 8-10 (with decile 1 being the most deprived and decile 10 being the least deprived).

Of the 260 participants who completed the BASQ at T1, 225 also completed a validated sensory processing measure; The Sensory Profile 2 (Dunn, 1999). Three months after T1, the participants were also invited to complete the BASQ measure again for test-retest reliability purposes. A subset (68%) of the original sample (n=177) responded and completed the measure at T2. This sub-set of participants had similar characteristics to the sample in T1 as the sample was predominantly White British (93%), 70% of children were male and the age range was 4-15 years 11 months (mean age 9.4 years).

Flow of participants into final sample. Figure 1 illustrates the flow of participants into the final sample (n=260) and at what point exclusions were made. Participants were required to meet the eligibility criteria (of being aged between 4yrs-15 yrs 11 mths, having a diagnosis of ASC and having English as a main language) and to have completed the BASQ in full to be included in the final sample.

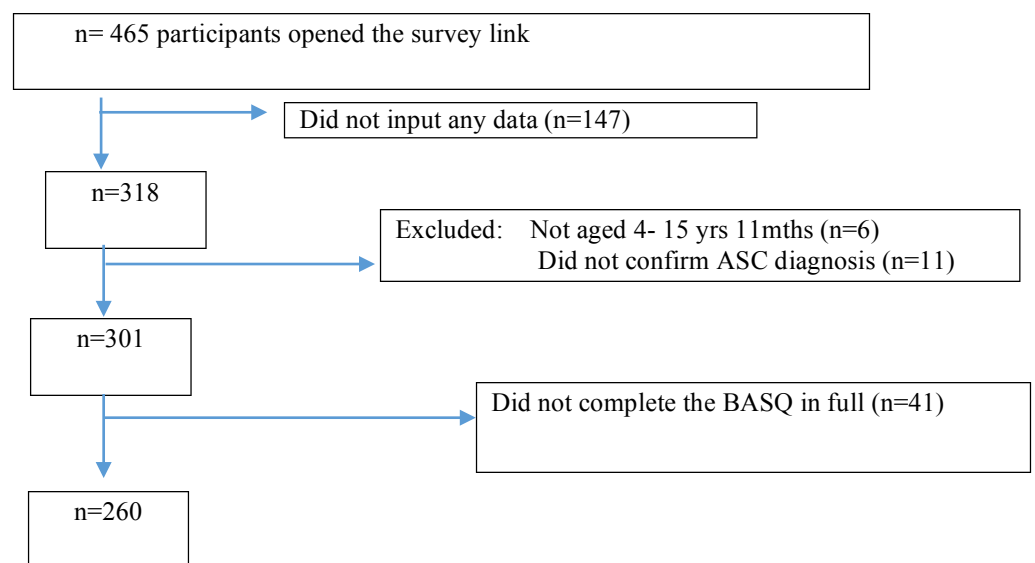


Figure 1. Diagram showing the flow of participants into the final sample.

Ethical considerations

The University of Sheffield's Department of Psychology Research Ethics Committee granted ethical approval of the project (Appendix H) and The British Psychological Societies (BPS) ethics guidelines for internet-mediated research were adhered to (BPS, 2017). All participants were provided with participant information detailing the purpose of the study, the nature of the questions, how data would be stored/handled and informing them of their right to withdraw at any time. A consent

declaration page with tick boxes to indicate consent was included but consent was also assumed if a participant completed the survey in full. Anonymity of questionnaire responses was ensured through the use of participant identification (ID) numbers. When data was extracted from Qualtrics (survey software), participant identifiable data (names, email and IP addressed) were removed and names and email details were then stored separately to non-identifiable data but linked by a unique ID number. All data was maintained by the researcher and was saved on a password-protected secure computer drive.

Statistical Analysis

Data analysis took place in three stages. All data analysis used data from the behavior and skills section of the BASQ (section 2). The first stage of the analysis involved assessing the structure and internal consistency of the BASQ, using confirmatory factor analysis and Cronbach's alpha respectively. The second stage assessed the psychometric of the BASQ and its subscales including exploring test-retest reliability and convergent validity. The third stage of the analysis involved exploring whether the data set provided evidence for ASC subgroups using latent class cluster analysis (LCCA) and defining the characteristics of these subgroups using a combination of ANOVAs, chi square tabulation and paired samples t-tests.

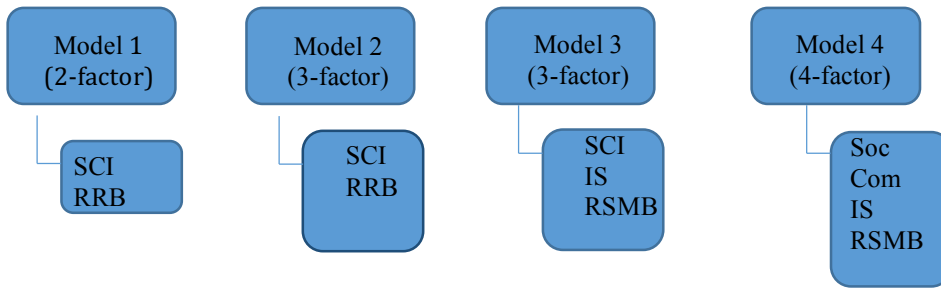
Stage one: BASQ factor structure and internal consistency

Confirmatory factor analysis. Data from the 'behaviours and skills section' of the BASQ were used in a confirmatory factor analysis (CFA) to describe the factor structure. CFA was applied to test four models

based on the variants in core symptom structure proposed in the literature (APA, 2000; 2013 and Szatmari et al. 2006). The specific models ranged from two to four factors (see figure two).

Model specification:

- A two-factor model based on the two domain DSM-V model (APA, 2013), consisting of social/communication (items 1-6) and RRB (items 7-12)
- A three-factor model based on the three domain DSM-IV model (APA, 2000), consisting of communication impairments (items 1-3), social interaction impairments (items 4-6) and RRB (items 7-12)
- A three factor model similar to the DSM-V model consisting of a social/communication factor (items 1-6) but then with RRB separated into two further factors of insistence on sameness (IS; items 7-9) and repetitive sensory motor behaviours (RSMB; factors 10-12) based upon previous research supporting this distinction (Szatmari et al., 2006).
- A four-factor model similar to the DSM-IV model consisting of a communication impairment factor (items 1-3), a social impairment factor (items 4-6) and then with RRB separated into two further factors of IS (items 7-9) and RSMB (items 10-12) based upon previous research supporting this distinction (Szatmari et al., 2006).



Note. SCI = Social communication impairment, RRB = Restricted and repetitive behaviours, IS = Insistence on sameness, Soc = Social interaction, Com = Communication, RSMB = Repetitive sensory and motor behaviours.

Figure 2. Four competing hypothesized models of the BASQ factor structure

Five statistical indices, including the model chi-square/degrees of freedom (χ^2/df), Comparative Fit Index (CFI) Tucker-Lewis Index (TLI), the Root Mean Square Error of Approximation (RMSEA) and the Weighted Root Mean Square Residual (WRMR) were used to evaluate the goodness of fit of each model to the data. Mplus8 (Muthén & Muthén, 1998-2017) software was used to compute all confirmatory factor analyses. In confirmatory factor analysis (CFA) a χ^2/df value less than 3.0, CFI and TLI values between 0.9-1.0 and RMSEA values of 0.06 or less indicate a good model fit (Browne & Cudeck 1993, Hu & Bentler, 1999, Briggs & Cheek, 1986).

Internal consistency. The internal consistency for each item and factor was then assessed using Cronbach's alpha (α). George & Mallery (2003) provide the following guidelines for interpretation of Cronbach's alpha: $>.9$ (excellent), $>.8$ (good), $>.7$ (acceptable), $>.6$ (questionable), $>.5$ (poor) and $<.5$ (unacceptable).

Stage Two: Establishing BASQ psychometric properties

Reliability. Participants were invited to complete the BASQ a second time (3 months later) after the initial administration and test retest reliability for section 2 of the BASQ (behavior and skills section) was assessed by calculating Pearson correlation coefficients for subscales and total scores.

Validity. Pearson correlation coefficients were used to assess the convergent validity of the BASQ scale with the Sensory Profile 2. The BASQ subscales of ‘insistence on sameness’ and ‘repetitive sensory and motor behaviours’ measure RRBs which have been reported to correlate with scores on the Sensory Profile (Gabriels et al., 2008; Inada et al., 2015). We hypothesized that BASQ subscale scores for the domains of IS and RSMB would demonstrate convergent validity by correlating with raw total scores on the Sensory Profile 2. We also hypothesized that BASQ subscale scores for the domains of communication and social interaction would not correlate with raw total scores on the Sensory Profile 2 and thus would demonstrate divergent validity.

Stage Three: Exploring subgroups (classes)

Latent Class Cluster Analysis (LCCA). To determine whether classes with distinct profiles of autistic traits could be identified, LCCA was performed using Mplus version 8 (Muthén & Muthén, 1998-2017). LCCA is a model based cluster analysis method used to identify subtypes of related cases (latent classes) from categorical, ordinal and continuous multivariate data (Muthén & Muthén, 1998-2017). The number of

competing models to evaluate was based on previous literature in this field (Cuccaro et al., 2012; Doshi-Velez et al., 2014; Georgiades et al., 2013; Greaves-Lord et al., 2013; Hu & Steinberg, 2009; Stevens et al., 2000) which has reported identifying between two to six ASC subclasses within their data sets. In our study we tested models that ranged from 1-6 classes to incorporate this range. As the confirmatory factor analyses indicated that a four-factor solution best fitted the data (see results section below), all models were tested using the four subscale scores (derived from the 12 item BASQ).

To estimate the number of classes underlying the sample, we compared the fit of six consecutive models using a combination of criteria. First values of the Akaike information criteria (AIC), Bayesian information criteria (BIC) and the Sample Size Adjusted BIC were used to estimate the optimal number of classes. AIC and BIC values are measures of model selection based on goodness of fit. They both give penalties to models with more classes in order to protect against the potential to ‘overfit’ models by including too many parameters (Geiser, 2013). Lower AIC, BIC and Adj.BIC values suggest better fitting models with BIC considered the superior information criteria (Nylund, Asparouhov & Muthén, 2007). Then, the results of the Vuong-Lo-Mendell Rubin Likelihood ratio tests were evaluated to compare the improvement of fit between neighbouring class models. This test provides a p value that can be used to determine if there is a statistically significant improvement in fit for the inclusion of one more class (Nylund, Asparouhov & Muthén, 2007).

Next, we looked at relative entropy, an overall measure of how well a model predicts class membership, which ranges from 0 (no predictive power) to 1 (perfect prediction). Finally, we looked at the mean posterior probability of a case belonging to each class. A good fitting model would have high individual probabilities for each case belonging to just one class (Muthén & Muthén, 1998-2017).

Currently there is no common agreement on the best criteria overall for determining the number of classes and so it is recommended that in addition to analyzing the aforementioned fit criteria that the interpretability of a solution (Geiser, 2013) and its agreement with substantive theory is also considered (Nylund, Asparouhov & Muthén, 2007).

Characterisation of classes. For the best fitting model class assignment of participants was carried out by placing participants in the class with the highest posterior class probability. A repeated measures ANOVA and Post hoc analyses were then carried out to describe the derived classes. Classes were described in relation to the child's current age, child's age at diagnosis as well as by their subscale scores on the BASQ measure (e.g. communication, social interaction, insistence on sameness and repetitive sensory and motor behaviour factor scores.) Cross-tabulation (chi-square analysis) was used to compare the proportion of children across classes by gender, history of language regression, language delay, history of seizures, presence of gastrointestinal problems, psychiatric comorbidities and presence of ASC in blood relatives.

Results

Each stage of the data analysis will now be summarised in turn.

Stage one: BASQ factor structure and internal consistency

Scale characteristics. The mean BASQ total score for the sample (n=260) based on the original 12 item version was 38.25 (SD=7.07) and scores ranged from 16-59 (possible range being 12-60) with no participants scoring at floor or ceiling level. Individual item distributions revealed a range from 2.3%-42.7% of participants scoring the same on any item and therefore did not identify any items as being a cause for concern.

Model fit. Table 1 presents goodness of fit indices from CFAs for each of the four models evaluated. RMSEA's ranged from 0.059 (model 4) - 0.119 (model 1) with model 4 meeting the criteria for 'good fit' (with a value <0.06). When other measures of fit were considered model 4 was also statistically superior to model 1 and 2 in terms of CFI and TLI values (0.972 and 0.961 respectively) and chi-square/degrees of freedom (1.90). Model 3 demonstrated an acceptable fit in three of the fit indices (chi-square/degrees of freedom, CFI and TLI values) however in comparison model 4 demonstrated a superior fit across all five fit indices reported. Based on these results the four factor model provided the best fit for the data and was therefore selected to describe the structure of the BASQ.

Table 1

Fit indices from confirmatory analyses for each model (based on 12 item BASQ).

Model	χ^2 (df)	χ^2/df	CFI	TLI	RMSEA
1 (2 factors)	249.205* (53)	4.70	0.873	0.841	0.119
2 (3 factors)	207.664* (51)	4.07	0.898	0.868	0.109
3 (3 factors)	146.811* (51)	2.88	0.938	0.920	0.085
4 (4 factors)	91.418* (48)	1.90	0.972	0.961	0.059

Notes. Italics denotes best fitting model for each fit statistic. * $p < .001$

Model 1 = two factors (social communication) & (RRB)

Model 2 = three factors (social), (communication) and (RRB)

Model 3 = (social communication), (IS) and (RSMB)

Model 4 = (social), (communication), (RSMB) and (IS).

Factor loadings. For the four factor model the average factor loadings were 0.76 for the communication (COM) subscale, 0.66 for the social interaction (SOC) subscale, 0.78 for the insistence on sameness (IS) subscale and 0.74 for the repetitive sensory motor behaviours (RSMB) subscale. See table two for the standardized factor loadings of individual items within subscales. All item loadings exceeded .40 and differed reliably from zero ($p < .01$), indicating that each of the four factors were well defined by their items.

Table 2

Standardized factor loadings of BASQ items

BASQ Item	Subscales			
	COM	SOC	IS	RSMB
1	.79			
2	.78			
3	.72			
4		.82		
5		.48		
6		.68		
7			.80	
8			.87	
9			.68	
10				.71
11				.80
12				.70

Internal consistency. Reliability statistics on the 12 items revealed the BASQ has acceptable internal consistency Cronbach's Alpha (α) = .73 with an average inter item correlation of 0.182 (Clark & Watson 1995; Nunnally, 1978). However, when a scale is measuring more than one construct it is recommended to calculate alpha for each of the constructs measured within the scale rather than for the entire scale (Nunnally, 1978). Using the model with the best fit (four factor) to define the dimensionality of the scale, the four subscales (each containing three items) of the BASQ were therefore assessed for internal consistency. Cronbach's α was found to be acceptable for three out of the four subscales (COM subscale α = .77, RSMB subscale α = .73 and IS subscale α = .75). The subscale of social interaction was within the questionable range (SOC subscale α = .65) and therefore warranted further consideration. A low alpha value could be caused by low number of questions, poor interrelatedness between items or heterogeneous constructs (Tavakol & Dennick, 2011). Inter-item correlation scores are not influenced by scale length like alpha (Briggs & Cheek, 1986) and guidelines about their interpretation take into account

how broad/narrow the construct being measured is (Clark & Watson, 1995). Therefore, individual inter-item correlations ($r=.25$ and $r=.35$) and the mean inter-item correlation for the SOC subscale ($r=0.3$) were inspected and all fell within the acceptable range (0.15 - 0.50) (Briggs & Cheek, 1986).

Scale structure. Based on CFA and reliability statistics the BASQ scale's structure was defined as a four factor scale consisting of 12 items, with three items loading onto each of the factors. The factors included: communication (COM) social interaction (SOC); insistence on sameness (IS) and repetitive sensory and motor behaviours (RSMB). This final 12 item, four factor scale forms the basis of the second stage of the analysis.

Stage 2: Establishing BASQ psychometric properties

Scale characteristics. The possible score range for each subscale of the BASQ was 3-15 with a possible total score range of 12-60. The first 6 items of the BASQ require reverse scoring. The COM subscale comprised items 1-3, the SOC subscale items 4-6, the IS subscale, items 7-9 and the RSMB subscale comprised items 10-12. See table 3 for means and standard deviations for all participants ($n=260$). A higher score is indicative of more severe difficulties.

Table 3

Means and standard deviations (SD) for the BASQ by subscale.

BASQ scale	Mean (SD)
COM (items 1-3)	9.27 (2.98)
SOC (items 4-6)	8.90 (2.68)
IS (items 7-9)	10.81 (2.72)
RSMB (items 10-12)	9.26 (3.02)
BASQ Total	38.25 (7.07)

A repeated measures ANOVA showed that participants' mean scores for the four subscales were statistically different $F(3,771) = 23.783$, $p < 0.0005$ in the sample ($n=260$). Paired samples t-tests revealed that scores on the IS subscale ($m=10.82$, $sd=2.715$) were significantly higher ($p=.000$) than for the COM ($m=9.27$, $sd=2.978$), SOC ($m=8.90$, $sd = 2.677$) or RSMB ($m=9.26$, $sd = 3.023$) subscales. No other pairwise comparisons of subscale scores were significant.

Retest reliability. Data for the second administration of the BASQ was available for 68% ($n=177$) of the original sample ($n=260$). The retest sample represented similar demographic characteristics as the original full sample from which it was drawn in terms of age, gender, ethnicity. Mean length of time between initial administration and retest was 3 months (range: 3-6 months, mode: 3 months, SD: 0.517). Total BASQ scores at both testing times were significantly correlated ($r=.73$, $p < 0.01$, two-tailed), indicating good stability in the BASQ over time (see table four). The test-retest reliability coefficients for the subscales of communication ($r=.68$, $p < 0.01$), insistence on sameness ($r=.75$, $p < 0.01$) and repetitive sensory and

motor behaviours ($r=.81, p<.01$) showed strong and significant correlations between administrations. However, the test retest reliability coefficients for the subscale of social interaction ($r=.37, p<.01$) showed only a weak but significant correlation between the two administrations time points. Overall the scale showed acceptable stability over time in the areas of total score and for three out of the four subscales but with the social interaction subscale showing a relative weakness in this area.

Table 4

Means, standard deviations and correlation coefficients for subset of sample (n=177) who completed the BASQ at both time points (T1 & T2).

Subscale	Score					r
	T1		T2			
	M	SD	M	SD		
COM	9.16	2.911	9.25	2.704	.68*	
SOC	8.91	2.661	9.10	1.442	.37*	
IS	10.82	2.713	11.07	2.509	.75*	
RSMB	9.17	3.020	9.32	2.908	.81*	
Total score	38.06	7.113	38.74	6.048	.73*	

**Correlation is significant at the 0.01 level (two-tailed).*

Validity. Convergent validity was examined using a subset (n=225) of the original sample (n=260) whom had completed the Sensory Profile 2 (in full) at the same time point (T1) as completing the BASQ (see table five). This sample represented similar demographic characteristics as the original full sample from which it was drawn. Pearson product moment correlations were conducted to examine the strength of the relationship between the BASQ total and subscales scores with the Sensory Profile 2 total scores.

Table 5

Pearson product correlations between the BASQ and the Sensory Profile 2.

	BASQ				
	Total Score	COM	SOC	IS	RSMB
SP2: Total Raw score	.486*	.055	.106	.443*	.542*

* $p < .01$ (two tailed)

Note: BASQ= Brief Autism Subtyping questionnaire, SP2 = Sensory Profile 2.

BASQ Total Score was found to be moderately and significantly correlated with total raw scores on the Sensory Profile 2 (Pearson's $r = 0.49$, $p < 0.01$). Both the IS subscale and the RSMB subscale showed positive moderate correlations with the total raw score on the Sensory Profile ($r = 0.44$ and $r = 0.54$ respectively) that were significant ($p < 0.01$). Whilst the SOC and COM subscales showed very weak and non-significant positive correlations with the sensory profile total raw scores. These findings are in line with our predictions about associations between the BASQ and Sensory Profile 2 measure and demonstrate acceptable convergent and divergent validity.

Age and gender differences. A Pearson product moment correlation was conducted to examine whether BASQ total scores demonstrated a significant relationship to child age. Results revealed a very weak negative correlation ($r = -.156$, $p < .05$).

An independent samples t-test indicated that there was a statistically significant difference in mean BASQ total score for males ($m = 38.81$, $SD = 7.435$) and females ($m = 36.94$, $SD = 5.955$), $t = 2.149$, $p = .033$.

Stage Three: Exploring subgroups (classes)

Latent class cluster analysis (LCCA). Table six reports all fit indices of the latent class analysis of the dataset (n=260). The BIC, adjusted BIC, and AIC parameters were all within a similar range but pointed to different class solutions. The AIC score favoured the 6 class model, the BIC favoured the 3 class model and the adjusted BIC favoured a 5 class model. The adjusted BIC and BIC are considered the superior indices and so we were left with a possible three or five class solution to consider.

In the absence of a conclusive result across the AIC, adjusted BIC and BIC indices, the vPLMR result and average latent class probabilities were also consulted. Overall the three class model provided the best fit to the data, demonstrated by its superior (lowest) BIC score and a pVLMR that approached significance ($p=0.05$) demonstrating that a third class improved fit for the model over a two class solution. Subsequent pVLMR scores demonstrated no improved model fit over the three class solution.

A careful examination of both the three- and five- class model solution profile plots showed the three class model to be more distinctly defined compared to the five- class model. On inspection of these profile plots, the larger class model appeared to be dividing two of the classes from the three class solution into higher or lower symptom severity and to not actually be representing additional 'distinct' profiles. For the three class model, average latent class probabilities were 0.82 for class 1, 0.83 for class 2 and 0.84 for class 3 and so indicated good latent profile distinctiveness.

According to this LCCA result, ASC can be described in this sample using data from the four subscale dimensions of the BASQ measure (COM, SOC, RSMB and IS) to stratify children into three relatively homogenous classes (class 1: 22%, class 2: 33% and class 3 45%).

Table 6

Goodness of fit indices for four-factor LCCA's specifying 1-6 classes

No of classes	Loglikelihood	No. of pars	AIC	BIC	Adj BIC	S	pVLMR
1	-2560.69		5137.38	5165.87	5140.50	-	-
2	-2525.85	13	5077.69	5123.98	5082.77	0.650	0.001
3*	-2501.54	18	5039.08	5103.18	5046.11	0.635	0.052
4	-2493.89	23	5033.78	5115.68	5042.76	0.679	0.242
5	-2486.91	28	5029.82	5129.52	5040.74	0.693	0.619
6	-2476.14	33	5028.96	5146.47	5041.84	0.711	0.150

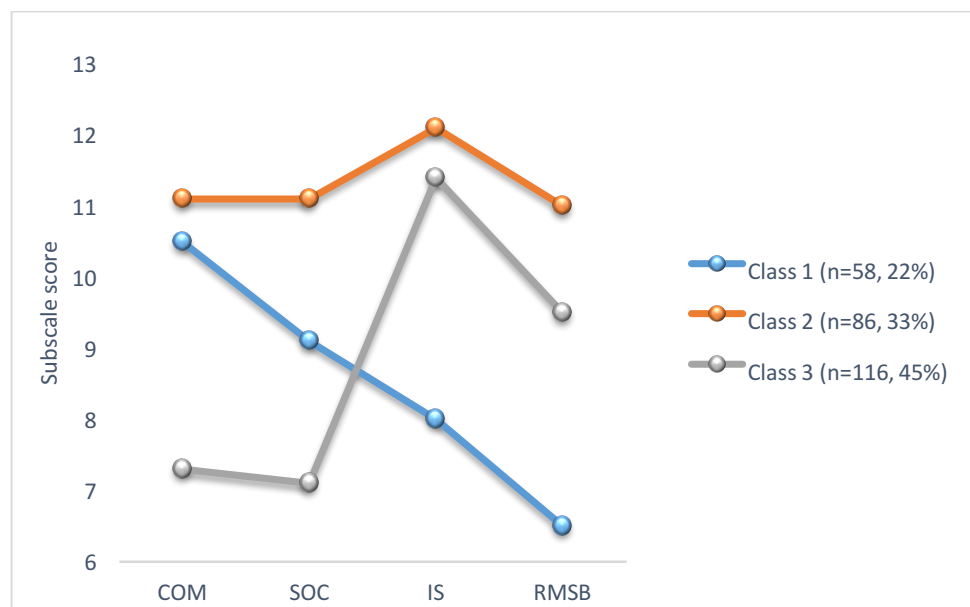
**selected model*

Note: BIC Bayesian Information Criterion, Adj. BIC Sample Size Adjusted BIC, AIC Akaike Information Criterion, S Entropy, p VLMR significance level from the Vuong-Lo-Mendell-Rubin Likelihood ratio test

Characterisation of classes based on BASQ profiles

A repeated measures ANOVA showed that there was a significant main effect of class membership; $F(6, 771) = 61.445, p < 0.001$ on total BASQ score, meaning that the three classes scored significantly different on the BASQ measure.

Figure three shows the average BASQ subscale/factor scores for each of the three subclasses and table seven displays the means and SDs for all continuous variables. High subscale scores indicate more severe symptom severity within a domain.



Notes: COM=Communication, SOC =Social Interaction, IS =Insistence on sameness and RMSB = Repetitive sensory motor behaviours.

Figure 3. Three class model - class profiles using mean scores on COM, SOC, IS and RMSB symptom dimensions (n=260).

Table 7

Means and standard deviations for the three classes

	Class 1	Mean (SD) Class 2	Class 3
Current age (in years)	10.28 (3.0)	9.28 (3.6)	9.72 (2.8)
Age at diagnosis (in years)	6.58 (3.04)	6.23 (3.30)	6.89 (2.7)
COM subscale	10.66 (2.36)	11.20 (2.26)	7.15(2.27)
SOC subscale	9.22 (2.14)	11.26 (2.04)	6.98 (1.70)
IS subscale	7.67 (2.12)	12.23 (2.10)	11.34 (2.09)
RSMB subscale	6.07 (2.22)	11.13 (2.31)	9.47(2.54)

Class 1 (n=58) showed a profile of increased severity in SOC and COM domains relative to RSMB domain scores. Paired-samples *t* tests confirmed significantly higher scores in the COM domain compared to SOC $t(57) = 4.062, p=.000$, RSMB $t(57) = 10.539, p=.000$ and IS $t(57) = 8.186, p=.000$, with the greatest difference being between the COM and RSMB domains. Scores in the SOC domain were also found to be significantly higher than scores in the IS domain; $t(57) = 4.575, p=.000$ and the RSMB domain; $t(57) = 7.522, p=.000$ within this class. Finally, the RSMB score was significantly lower than the IS score within this class; $t(57) = -3.605, p=.001$.

Class 2 (n=86) represented the most severely affected subgroup with the highest scores across all ASC symptom domains and a significantly elevated insistence on sameness score relative to the other subscales. Paired-samples *t* tests showed a significant difference between the score on the IS subscale and all other subscales, with the COM subscale $t(85) = 2.857, p=.005$, with RSMB subscale $t(85) = -3.417, p=.001$ and also with the SOC subscale $t(85) = -3.078, p=.003$. No significant differences were found between the COM and SOC subscale scores $t(85) = -.178, p=.859$, the SOC and RSMB subscale scores $t(85) = .389, p=.698$ or the COM and RSMB subscale scores $t(85) = .204, p=.839$.

Class 3 (n=116) showed the lowest scores in the communication and social interaction domains than any of the other classes and a significantly elevated score in the insistence on sameness domain. Paired-samples *t* tests showed the IS subscale score to be significantly higher than all the other subscale scores; with COM; $t(115) = .14.766, p=0.000$, with

SOC; $t(115) = 17.564$, $p = 0.000$ and with RSMB; $t(115) = 9.050$, $p = 0.000$.

The difference between the COM and SOC subscale scores within this class were not significant; $t(115) = .641$, $p = .523$.

These results show that the three classes represent significantly distinct profiles in terms of ASC symptoms that do not simply reflect symptom severity gradients within the sample.

Further characterisation of classes

Table eight presents the results of the cross tabulation (chi square analysis). Class one showed the lowest rates of mental health diagnoses than any of the other two classes but this result was not at a statistically significant level.

Class two stood out for differing significantly ($p < 0.05$) from the other two classes in terms of children in this class having a different type of onset to the other two groups (regression) as well in terms of the number of children who had a history of language delay. Additionally, class two differed from the other classes in terms of having a significantly ($p < 0.05$) higher prevalence of OCD diagnosis amongst the children in this class.

Class 3 differed from the other classes by having the highest reported history of non- febrile seizures and was the only group where diagnosis of epilepsy was reported, however these differences were not at a significant level. Within the total sample only 5% reported a diagnosis of epilepsy in their child, this small proportion may have limited our ability to detect whether this result is significant.

There were no significant differences in distribution across the three classes for any of the following diagnoses; ADHD, anxiety, attachment difficulties, depression, conduct disorder, oppositional defiant disorder or pathological demand avoidance. Groups also did not differ significantly by presence of genetic conditions, diagnoses of comorbid developmental disorders or in rates of diagnosis with intellectual disability. Neither did groups differ significantly by gender or ethnicity. A one-way between subjects ANOVA showed that current age and age when diagnosed did not differ significantly between classes.

Table 8

Further characteristics of the three classes

Class	Mean Age (SD)	% Female	%ASC:AA: Asp:PDD-NOS	% Seizure (non febrile) history	% Epilepsy diagnosis	% Language regression	% Other regression	% Language delay	% with ASC diagnosed in a blood relative	% with GI problems	% MH diagnosis	% OCD
1 (n=58)	10.28 (3.1)	31	72:2:26:0	7	0	19	19	45	42	48	12	0
2 (n=86)	9.28 (3.6)	21	83:4:12:1	7	0	44*	56*	65*	36	59	22	9*
3 (n=116)	9.72 (2.8)	36	82:1:16:1	13	5	18	29	49	38	64	23	2

* denotes a subset category whose proportion differs significantly (at the 0.5 level) from the others

Note: %ASC:AA:Asp:PDD-NOS = percentage of children with Autism Spectrum Condition (ASC), Atypical autism (AA), Asperger's syndrome (Asp) and Pervasive Developmental Disorder-Not Otherwise Specified (PDD-NOS) based on care giver reported specific diagnosis. GI problems = gastrointestinal problems, MH diagnosis = mental health diagnosis, OCD = Obsessive Compulsive Disorder.

Discussion

The primary purpose of this study was to develop and carry out preliminary psychometric evaluation of a brief and acceptable measure to identify autism subgroups. The outcome was the production of the Brief Autism Subgrouping Questionnaire (BASQ). Additionally, we wanted to examine whether the BASQ could then be used to stratify children with ASC into more homogenous subclasses based on symptom severity and to then further characterize these classes.

Summary of main findings

Confirmatory factor analysis (CFA) was employed to verify the factor structure of the BASQ. Four hypothesised models were examined which ranged from two to four factors and which were based on variants in ASC core symptom structure as proposed in the literature (refs). The results of these analyses indicated that a four factor structure provided the best fit to the data and outperformed the three other models across all five fit indices that were examined (chi-square, degrees of freedom, CFI and RMSEA scores). This suggests that the BASQ consists of four subscales that we refer to as (1) Communication; (2) Social interaction; (3) Insistence on sameness and (4) Repetitive motor and sensory behaviours. The distinction between communication and social interaction factors suggested by our results aligns in part with the DSM-IV classification system for autism (APA, 2000), in terms of its consideration of social interaction and communication as separate symptom domains.

The suggestion by our results of a two-dimensional nature to the repetitive behaviour domain is in line with previous studies into restricted and repetitive behaviours in autism (Bishop, Richler & Lord, 2006; Cuccaro et al., 2003; Shao et al., 2003; Szatmari et al., 2006) which have reported the same two factor structure to this domain based on analyses of ADI-R data. These studies have similarly identified the two factors to comprise of a 'lower-order category', the RSMB domain and a 'higher-order' category, the IS domain. Our findings are likely to be influenced by the original ADI-R item pool used in this study as a basis for the development of the BASQ question items and so we would expect to see these similar results. The emergence of the four-factor model as a representation of the BASQ is in contrast to the recent changes in the classification system for autism (DSM-V: APA, 2013) which has seen a move to a two-factor model of autism (social communication and RRB).

Initial evaluation of the BASQ showed promising psychometric properties, with acceptable internal consistency ($\alpha = .73$) and test-retest reliability ($r = .73$). Our hypothesis (H^1) that the BASQ items pertaining to RRBs would correlate positively with scores on the SP2 was supported. Convergent validity of the measure was therefore demonstrated by significant but moderate positive correlations between SP2 total scores and the IS ($r = .44$, $p < .01$) and RSMB ($r = .54$, $p < .01$) subscales of the BASQ. Our second hypothesis (H^2) that the BASQ scores for assessing social interaction and communication would not show any significant correlation with scores on the SP2 was also supported and suggestive of divergent validity of the measure. The BASQ communication and social interaction subscales showed very weak and non-significant correlations with the

sensory profile total raw scores ($r=.06$ and $r=.11$ respectively). The finding of significant relations between the RSMB and IS subscales with SP2 scores is in line with reports of a direct relation between RRBs and atypical sensory responses amongst individuals with ASC (Gabriels et al., 2008) and with the current inclusion of abnormal sensory behaviours within the DSM-V (APA, 2013) RRB symptom domain.

Finally, our third hypothesis (H^3) that the BASQ would be able to discriminate subclasses with distinct ASC symptom profiles was supported by the results of the LCCA, ANOVA and subsequent post hoc analyses. LCCA of the data set identified a set of three classes differentiated by significantly distinct profiles of autistic traits. Class one (22%) showed a relative increased severity in the areas of communication and social interaction compared to significantly lower scores in the IS and RSMB domains. Class two (33%) showed an overall 'severe' presentation across all four symptom domains in comparison to the other classes but with a significantly higher score in the IS domain relative to the other three symptom domains. Class three (45%) showed the reverse profile of class one, with low scores in both communication and social interaction compared to higher scores in both RRB domains, with IS score being significantly higher than all other subscale scores. These findings are in line with and support those of Georgiades et al. (2013) who also found evidence of three classes similarly defined by differential severity gradients on social communication deficits and fixated interests and repetitive behaviours

Clinical & Theoretical Implications

Development of the BASQ. The study provides preliminary evidence that the BASQ is a valid and reliable measure for identifying distinct ASC symptom profiles. The BASQ can be completed as either an online or paper version and is relatively brief in nature; comprising a demographic section (including medical and developmental history) and a 12 item behaviour and skills section. This briefer, standardised subtyping measure has the potential to be a useful tool for future studies wanting to further explore, evidence and define potential autism subclasses. Having a reliable measure, which collects the relevant data for stratifying individuals with ASC into more homogeneous groups, means that the larger scale longitudinal studies currently recommended within the literature will be more achievable. The use of a standardized measure within subtyping research will also reduce some of the variability across studies introduced from the different items/measures used.

Symptom structure of ASC domains. Although the ASC symptom domain of RRBs is diagnostically considered a unitary domain of behaviour, the results of this study add some support to a growing body of evidence suggesting a two-factor structure to RRBs in autism, comprised of RSMB and IS (Cuccaro et al., 2003; Bishop, Richler & Lord, 2006; Szatmari et al., 2006). The theoretical and clinical significance of the identification of distinct factors within RRB is that it demonstrates that there are potential RRB subtypes which may represent different etiological mechanisms. The differentiation of etiologies would then be of particular significance for research into intervention outcomes and the development of more tailored clinical interventions. These findings are consistent with the

work of Berkson and Tupa (2000) who comment that to further our understanding of repetitive behaviours they should not be grouped together in a single class. However, our study results were limited by the pool of items we used in our measure in that they were similar to the items previously identified as representing these two RRB constructs. Questions measuring ‘circumscribed interests’ were not included in our measure, which has been identified as a possible third factor to this domain (Lam, Bodfish & Piven, 2008).

Relation between RSMB, IS and sensory processing. Our finding of a significant positive correlation between the BASQ’s RSMB and IS subscales with the Sensory Profile 2 (SP2) is in line with the current evidence base (Boyd et al., 2009, 2010; Chen et al., 2009; Gabriels et al., 2008; Gal et al., 2010) suggesting that more frequent/severe restricted, repetitive and stereotyped behaviours and interests are associated with atypical sensory responses. The BASQ did not include any overlapping items with the SP and so this correlation was not artificially inflated due to item overlap.

Clinically, it would be of benefit for professionals to be aware of the potential associations between RRBs and sensory differences in order to inform more comprehensive assessments and clinical interventions when individuals present with RRB difficulties. Theoretically, further studies are required to support/refute these findings of an RRB-sensory differences association and the factors that influence this relationship in order to further our understanding of the underlying mechanisms and possible targets for intervention.

Evidence for subclasses differentiated by distinct symptom

profiles. Previous research has provided evidence for subclasses of children with ASC that differ in relative severity of symptoms rather than absolute symptom severity. The findings of our exploratory study add further support to studies which claim to be able to distinguish subgroups with distinct profiles of autistic traits. Our theoretical findings could be used to generate hypotheses related to the three derived ASC subgroups (in terms of their different developmental trajectories and responses to intervention) which could then be examined through longitudinal follow up studies, using the BASQ to stratify children into subgroups. Additionally, further exploration using additional measures and testing would help to characterize these potential classes further, for example carrying out cognitive testing or completing measures of adaptive functioning would help to establish the relationship between these classes and IQ/functioning level.

The current results also have implications for genetic studies. Being able to organize children with autism into more homogeneous ASC subclasses could allow for children to be stratified into more meaningful groups for genetic studies and studies looking for biological markers of ASC.

It is important that these subclass findings of the present study are interpreted in the context of this being an exploratory study, using what might be considered a relatively small sample size for LCCA. It would be prudent to see if these subclass findings were replicated using the BASQ with a much larger sample of $n > 500$ in order to be more confident of these findings.

Strengths and limitations

A strength of this study was the design of a specific measure to be used in autism subtyping research. To date, no such measure exists despite growing evidence in support of the presence of subtypes within the autism phenotype. The BASQ collects targeted data relevant to potential subclasses in a more concise and standardised format than current methods. The online format of this measure also allows for initial screening/recruitment on a larger scale with limited costs incurred and with less burden to the participant.

The use of a cognitive interview approach in pre-field testing of the BASQ (pilot version) was another strength of the study. Cognitive interviews captured parents' response behaviour whilst completing the measure and allowed identification of potential problem items prior to distribution. This methodology enabled a valuable insight into the response process and allowed for unique parent/caregiver input into the acceptability, comprehensiveness, relevance and clarity of items, examples and instructions.

The study recruited an adequate sample size ($n=260$) to validate the 12-item behavior and skills section of the BASQ, greatly exceeding our recruitment target of $n=120$ which was chosen to ensure a robust factor structure.

Nevertheless, the present study was of an exploratory nature and as such had a number of limitations. The sample was 70% male ($n=182$) and 92% white British meaning that females and children from other ethnic

backgrounds were under represented. However, studies looking at the prevalence rates for ASC with respect to gender report gender ratios of between 2:1 to 5:1 with male dominance (Adak & Halder, 2017) and so it would be expected to see a significantly higher proportion of males in a sample of individuals with ASC.

The presence of sample bias in relation to parents who responded to the study recruitment advertisement must also be considered. It could be that parents who responded were more concerned about their children or were parents with better access to the internet and so possibly more knowledgeable about autism and/or from a higher socioeconomic background.

Another limitation of our sample was the wide age range (4-15 years 11 months) of the sample. However, the age differences between the three identified subclasses were non-significant and so we were able to assume that age factors did not interfere directly or indirectly with the clustering process.

The BASQ is a parent/caregiver report measure and subsequently all of our findings can be subject to reporting biases such as selective recall, social desirability and the influence of informant characteristics (personality, parental knowledge and understanding about autism). Some aspects of the demographics section of the questionnaire require participants to provide historical information with regards to the child's early development which might be subject to recall errors. In terms of the parent/caregiver that responded, 90% of completed BASQs were based on

maternal reports. The BASQ would benefit from testing using multiple informants to look at interrater reliability.

One limitation shared by other postal or internet- based questionnaires is that there is no independent validation that respondents actually do have a child with an autism diagnosis; however, as there was no incentive for participating there would be little motive to enter false data.

Future recommendations

Further validation of the BASQ with more diverse samples will be needed and it is important that the results of this study are interpreted with these limitations in mind. Further psychometric testing in relation to the construct validity and interrater reliability of the BASQ would be beneficial.

The current study was limited by the lack of confirmation of a formal diagnosis of ASC. Future studies into the validation of the BASQ would benefit from incorporating a means by which the diagnostic status of participants can be confirmed, either by requesting the child's original diagnostic report or including the administration of valid autism screening/diagnostic measures.

The development of the BASQ for future use will hopefully stimulate further research into autism subgroups and further our understanding in this area.

It is important that future studies consider whether subgroups might represent different developmental stages. The completion of larger scale longitudinal studies to address disputes around whether subgroups are a

result of developmental or cognitive differences would therefore be a useful next step in this field.

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Appendix A. Items used to guide the development of the BASQ alongside final version of BASQ items. Removed due to copyright restrictions

Appendix A (continued). Removed due to copyright restrictions

Appendix A (continued). Removed due to copyright restrictions.

Appendix A (continued). Removed due to copyright restrictions

Appendix B. Demographic data for samples in piloting phase of BASQ development.

Table B1

Demographic details for clinical sample (n=4) in piloting phase of BASQ development.

Respondent	75% (n=3) birth mother 25% (n=1) foster mother
<u>Child's details</u>	
Gender	75% (n=3) male
Age	Mean= 9 years (range=7-12 years)
Clinical status	100% (n=4) have an ASC diagnosis

Table B2

Demographic details for non-clinical sample (n=5) in piloting phase of BASQ development.

Respondent	80% (n=4) birth mother 20% (n=1) birth father
<u>Child's details</u>	
Gender	80% (n=4) male
Age	Mean= 7 years (range=4-9 years)
Clinical diagnosis	100% = No ASC diagnosis

Appendix C. Section 1 of BASQ

Please note, the format of the BASQ looks slightly different online to this paper version (due to skip logic and drop down menu features) but it has the exact same content.

Brief Autism Subtyping Questionnaire**Instructions:**

This questionnaire should be completed by the primary caregiver of a child/young person between the ages of 4 years-15 years 11 months old with a diagnosis of an autism spectrum condition (ASC).

Additional instructions for online version:

You have up to one week in which to complete the questionnaire and can save/close the browser and return to the questionnaire via the link at any point in that time. However, after one week has passed your questionnaire will close and the data entered so far will be recorded.

BASQ: Section 1**About You**

This form is filled out by...

- Mother
- Father
- Foster Mother
- Foster Father
- Adoptive Mother
- Adoptive Father
- Other primary caregiver (please state)

Please provide your postcode: _____

What is the main language used by the family at home? (please state) _____

There is a second part to this research study that involves you completing a significantly shorter version of this questionnaire in 3 months-time. Please supply your email address below so that we can email you the link to this once this time period has passed:

Appendix C. (continued)

About Your Child

What is the child's date of birth? ___/___/___

Please provide the child/young person's current age

- 4 years - 4 years 11 months
- 5 years - 5 years 11 months
- 6 years - 6 years 11 months
- 7 years - 7 years 11 months
- 8 years - 8 years 11 months
- 9 years - 9 years 11 months
- 10 years - 10 years 11 months
- 11 years - 11 years 11 months
- 12 years - 12 years 11 months
- 13 years - 13 years 11 months
- 14 years - 14 years 11 months
- 15 years - 15 years 11 months

Please provide the child's/young person's gender:

- Male
- Female

Appendix C. (continued)

Please provide the child/young person's ethnicity:

- White British
- White Irish
- White Gypsy or Irish Traveller
- Any other white background, please describe _____
- White and Black Caribbean
- White and Black African
- White and Asian
- Any other mixed/multiple ethnic background, please describe__
- Indian
- Pakistani
- Bangladeshi
- Chinese
- Any other Asian background, please describe _____
- Black African
- Black Caribbean
- Any other Black/African/Caribbean background, please describe _____
- Arab
- Any other ethnic group, please describe _____

Diagnosis Information

1.) Does your child have a diagnosis of an autism spectrum condition (ASC)?

- Yes
- No

Please indicate the *specific* diagnosis given by choosing one of the options below:

- Autism spectrum disorder (ASD)/condition (ASC)
- Atypical autism
- Asperger's syndrome
- Childhood disintegrative disorder (CDD)
- Pervasive developmental disorder - not otherwise specified (PDD-NOS)
- Other (Please state) _____

If possible, please provide the name of the service where this diagnosis was given? _____

Appendix C. (continued)

If possible, please provide the age of the child/young person when they were first given this diagnosis?

2.) Does your child have a diagnosis of any of the following developmental disorders?

Tick all that apply

- Pathological demand avoidance (PDA) syndrome
- Rett syndrome
- Fragile X
- Prader-Willi or Angelman syndrome
- Turner syndrome
- Cohen syndrome
- Tuberous sclerosis
- Down's syndrome
- Attention deficit hyperactivity disorder (ADHD)
- Tourette's syndrome
- Conduct disorder
- Oppositional defiant disorder (ODD)
- Developmental co-ordination disorder (DCD) or dyspraxia
- Sensory processing disorder/ Sensory integration dysfunction
- Learning/intellectual disability
- Social (pragmatic) communication disorder
- Other (please state) _____
- No diagnosis of any of the above

Appendix C. (continued)

3.) Does your child have a diagnosis of any mental health condition?

- Anxiety or generalised anxiety disorder (GAD)
- Attachment disorder
- Depression
- Obsessive compulsive disorder (OCD)
- Other (Please state) _____
- No diagnosis of any of the above

4.) Does your child have any current uncorrected or severe visual/hearing impairments?

- No
- Yes, uncorrected/severe hearing impairment
- Yes, uncorrected/severe visual impairment
- Other (please state) _____

Family History

Below is a list of various conditions. We are interested in knowing if any of these have been diagnosed in the child's blood relatives (siblings, mother, father, grandparents, aunts, uncles and/or cousins)?

Appendix C. (continued)

Please tick all that apply and indicate which blood relative in the space provided.

- Autism spectrum disorder(ASD)/condition (ASC)

- Atypical autism _____
- Asperger's syndrome _____
- Childhood disintegrative disorder (CDD) _____
- Pervasive developmental disorder - not otherwise specified (PDD-NOS) _____
- Pathological demand avoidance (PDA) syndrome

- Rett syndrome _____
- Fragile X, Prader-Willi or Angelman syndrome _____
- Turner syndrome _____
- Cohen syndrome _____
- Tuberous sclerosis _____
- Down's syndrome _____
- Attention deficit hyperactivity disorder (ADHD)

- Tourette's syndrome _____
- Conduct disorder _____
- Oppositional defiant disorder (ODD) _____
- Developmental co-ordination disorder or dyspraxia

- Sensory processing disorder/Sensory integration
dysfunction _____
- Other (please state) _____
- Not known _____
- No blood relatives of the child known to have any of the
above _____

Appendix C. (continued)

Medical Information

5.) Has your child ever suffered from non-febrile (without a fever) seizures?

- My child has not had a seizure
- My child has had a non-febrile (without a fever) seizure but has not been diagnosed with epilepsy
- My child has been diagnosed with epilepsy

6.) Has your child experienced any of the following gastro intestinal problems that are chronic, persistent, recurrent, frequent or excessive in nature, with no clear anatomic, metabolic or pathological cause?

- Chronic diarrhea
- Chronic constipation
- Faecal incontinence
- Encopresis (faecal soiling)
- Abdominal pain/discomfort/irritability/bloating
- Persistent/excessive vomiting
- Persistent/excessive nausea
- Gastroesophageal reflux (GER) Changes to bowel habit
- Other (please state)
- No gastro intestinal problems of this type

8.) Is your child taking any medication currently or have they been within the last six months?

- Yes
- No

If yes, please elaborate on the medication that your child is taking/has taken in the last 6 months
(e.g. what and what for?)

Appendix C. (continued)

Developmental History

- 9.) Did/does your child show a delay in the area of language development? (with delay defined as failure to develop speech to the level where they could combine two words or know more than 30 words by 24 months).
- No delay in language development
 - Yes, they did/do show a delay in language development
 - Not known

- 10.) Between the ages of 4-5 years old, did/does your child show an understanding of simple words/phrases without requiring gestures or pointing to indicate their meaning?
e.g. if you ask him/her to get something you don't have to point at the object to indicate what it is, or if offering him/her something they don't require a visual cue/prompt/gesture to know what you are talking about?

- Yes, they did/do show understanding without gesture or pointing
- No, they didn't/don't show understanding without gestures or pointing
- Not known

- 11.) Did you ever notice that your child regressed in the area of language development, e.g. developed speech typically and then lost certain words from their vocabulary or stopped using language that had previously been used?

- Yes
- No
- Not known

If your child did show a delay or regression in the area of language development was he/she referred for Speech and Language therapy either for assessment or intervention?

- Yes
- No
- Not known

Appendix C. (continued)

If yes, at what age was your child referred to speech and language therapy?
Please provide as much information as possible.

12.) Have you ever felt that your child regressed in other areas of their ability? e.g. making eye contact, showing you how they feel (facial expressions), using gestures/pointing, showing you their toys etc? N.B. by regression we mean your child seemed to develop skills in a normal way but later lost these skills. Please note that this is different from a child who never developed these skills.

- Yes
- No
- Not known

If yes, at what age(s) did your child's regression(s) in other areas of ability occur?

Please provide as much information as possible. _____

Appendix D. BASQ Section 2 – ‘Behaviour and Skills’ section

Please note, the format of the BASQ looks slightly different online to this paper version (due to skip logic and drop down menu features) but it has the exact same content.

BASQ: Section 2**Behaviours and Skills****Instructions**

The pages that follow contain statements that describe behaviours and skills that children might exhibit. For each of the following twelve descriptions please indicate an option on the scale that best describes your child's current skills/behaviour in that area (e.g. in the last six months).

Examples of the types of behaviours or skills we are looking for are provided as a guide but are not an exhaustive list.

1. Spontaneously uses appropriate hand gestures to communicate a message with others (either alongside or independently from vocalizations).

Some examples might include: clapping to congratulate; waving to say hello/goodbye; giving a thumbs up to show approval and/or tapping the space next to them to signal for you to sit there.

1 _____ 2 _____ 3 _____ 4 _____ 5

My child
never
does this

My
child
rarely
does

My child
sometimes
does this

My child
often does
this

My child
always does
this

2. Points to things in order to draw your attention to something of interest in the environment (this is done for the purpose of showing/sharing and not to request that you pass them the object). This can be done either alongside or independent from speech.

Some examples might include: pointing out an animal in the street/park or pointing out something of interest at a show/fair. When they do so they will look at the object, then look to you, get your attention, point and then look back at the object.

1 _____ 2 _____ 3 _____ 4 _____ 5

My child
never
does this

My
child
rarely
does

My child
sometimes
does this

My child
often does
this

My child
always
does this

Appendix D. (continued)

3. Uses the following gestures spontaneously (without prompting) either alongside or independently from vocalisations; Nods head to communicate yes, shakes head to say no and/or shrugs shoulders to say 'don't know/mind or not sure.'

1 _____ 2 _____ 3 _____ 4 _____ 5

My child never does this	My child rarely does	My child sometimes does this	My child often does this	My child always does this
--------------------------------	-------------------------------	------------------------------------	--------------------------------	---------------------------------

4. Tries to show you or direct your attention towards toys, objects or activities that they are interested in or achievements that they are proud of? (this is NOT done because they want you to do something for them with the object, such as open it). This can be done either alongside or independent from speech.

Some examples might include: bringing you a picture that they have drawn to show you; calling/pulling you over to look at a jigsaw that they have completed or inviting you to watch them doing something.

1 _____ 2 _____ 3 _____ 4 _____ 5

My child never does this	My child rarely does	My child sometimes does this	My child often does this	My child always does this
--------------------------------	-------------------------------	------------------------------------	--------------------------------	---------------------------------

5. Shows concern or tries to comfort you or others (such as older or younger children) when you/they are sad, ill, hurt or in visible distress? e.g. hugging someone who is crying or if they are verbal asking if you/they are ok.

1 _____ 2 _____ 3 _____ 4 _____ 5

My child never does this	My child rarely does	My child sometimes does this	My child often does this	My child always does this
--------------------------------	-------------------------------	------------------------------------	--------------------------------	---------------------------------

6. Wants to share their enjoyment or pleasure in an activity with you (or others) in a way where they invite you (or others) to join in and share the experience with them?

Some examples might include: inviting you to join in with a hobby; showing you something funny on TV or initiating for you to join in playing a game they enjoy. This can be done by verbal or nonverbal communication.

1 _____ 2 _____ 3 _____ 4 _____ 5

My child never does this	My child rarely does	My child sometimes does this	My child often does this	My child always does this
--------------------------------	-------------------------------	------------------------------------	--------------------------------	---------------------------------

Appendix D. (continued)

7. Shows resistance to and is distressed by small changes in their routine or environment.

Some examples of this might include becoming upset if there is a change to the time or order in which things happen, becoming upset at the change from winter to summer clothing, wanting things to be kept in the same place, listening to/watching the same part of a song/movie repetitively and/or becoming upset when others try to stop/interrupt what they are doing.

Please think of the most commonly occurring behaviour of this type in your child when answering this question.

1 _____ 2 _____ 3 _____ 4 _____ 5

My child never does this	My child rarely does	My child sometimes does this	My child often does this	My child always does this
--------------------------------	-------------------------------	------------------------------------	--------------------------------	---------------------------------

8. Strongly insists on performing activities of daily life in a set way.

Some examples of this might include: insisting on certain routines/orders during mealtimes or when washing or dressing; insisting that certain items are used/worn (same cup/socks etc.); insisting on certain routes when travelling and/or wanting others to perform activities or say/respond to things in a certain way.

Please think of the most commonly occurring behaviour of this type in your child when answering this question.

1 _____ 2 _____ 3 _____ 4 _____ 5

My child never does this	My child rarely does	My child sometimes does this	My child often does this	My child always does this
--------------------------------	-------------------------------	------------------------------------	--------------------------------	---------------------------------

9. Needs things/actions to be repeated or done according to a rule/until 'just right.'

Some examples of this might include: repeating actions such as going in/out of a room; placing or arranging items in a particular way/order; having all doors/lids closed; having to count to a certain number and/or touch/tap items a set number of times.

Please think of the most commonly occurring behaviour of this type in your child when answering this question.

1 _____ 2 _____ 3 _____ 4 _____ 5

My child never does this	My child rarely does	My child sometimes does this	My child often does this	My child always does this
--------------------------------	-------------------------------	------------------------------------	--------------------------------	---------------------------------

Appendix D. (continued)

10. Focuses on certain parts of a toy or object and uses them in a way that is repetitive and different to their intended use/function.

Some examples of this might include: spinning the wheels of toy car/pram/bike but not playing with the toy in any other way or pressing buttons without an interest in their function.

1 _____ 2 _____ 3 _____ 4 _____ 5

My child
never does
this

My
child
rarely
does

My child
sometimes
does this

My child
often does
this

My child
always does
this

11. Moves his/her fingers, hands or arms in a quick, deliberate and repetitive manner.

Some examples of this might include: flapping, waving or shaking their fingers, hands or arms quickly and repetitively. These movements might usually (but not always) be done within their own line of vision with the child watching the movements out of the corner of their eye.

1 _____ 2 _____ 3 _____ 4 _____ 5

My child
never
does this

My
child
rarely
does

My child
sometimes
does this

My child
often does
this

My child
always does
this

12. Displays repetitive movements of his/her whole body.

Some examples might include: spinning; jumping up and down; crossing or uncrossing of legs; foot to foot swaying and/or dipping of body. These behaviours may or may not be carried out on tip toes.

1 _____ 2 _____ 3 _____ 4 _____ 5

My child
never
does this

My
child
rarely
does

My child
sometimes
does this

My child
often does
this

My child
always
does this

Appendix E. Study information page

Autism Subtyping Questionnaire

Title of Research Project: Developing a questionnaire to identify autism subtypes.

Researchers: Kirsty Howell (Trainee Clinical Psychologist) & Dr Elizabeth Milne (Supervisor/Director of the Sheffield Autism Research Lab).

What is the research about?

This research aims to develop a better understanding of autism by developing a new questionnaire looking at the differences and similarities across certain characteristics and behaviours in children and young people with autism. We believe that there may be different sub-types of autism and we hope to be able to define these sub-types with this new questionnaire. By taking part you will be contributing to developing this understanding and contributing to the progression of research in this field.

Who can take part?

To take part in the research you must be the parent/caregiver of a child/young person aged 4 years -15 years 11 months old with a diagnosis of an autism spectrum condition (ASC).

What does it involve?

The research involves completing two online questionnaires, at two different time points, about your child's development and behaviour. The first questionnaire we are asking you to complete now and takes approximately 30-45 minutes to do. The second questionnaire will be emailed to you in three months time and if you choose to complete it, should take between 10-15 minutes to complete.

Please read the [participant information sheet](#) for full details about the study.

If after reading the information sheet you have any questions about the research you can contact the researcher by email on khowell1@sheffield.ac.uk

Appendix F. Participant Information Sheet

Participant Information Sheet

Designing a questionnaire to identify subtypes in autistic spectrum conditions.

You are invited to take part in a research project. Before you decide if you would like to take part it is important for you to understand why the research is being done and what it will involve. Please take time to read the following information carefully and discuss it with others if you wish. Ask us if there is anything that is not clear or if you would like more information. Take time to decide whether or not you wish to take part. Thank you for reading this.

What is the purpose of the research?

We are developing a new questionnaire looking at the differences and similarities across certain characteristics and behaviours in children and young people with autism. We believe that there may be different sub-types of autism and we hope to be able to define these sub-types with this new questionnaire. At this stage we are inviting people to complete the questionnaire so that we can test whether it will be useful and reliable to use in research. This study is being undertaken as part of a Doctorate in Clinical Psychology at the University of Sheffield.

Who can take part?

Parents or primary care givers of children aged 4 years -15 years 11 months old with a diagnosis of an autism spectrum condition (including childhood disintegrative disorder and pervasive developmental disorder – not otherwise specified).

Do I have to take part?

It is up to you to decide whether or not to take part. If you agree to take part now, you can withdraw at any time without giving reason.

What will happen to me if I do take part?

We are asking everyone who takes part to complete two questionnaires about his or her child's development and behaviour. The first questionnaire should take no longer than approximately 30-45 minutes to complete. The questionnaires can be accessed and completed via an online link or a paper copy can be made available. You will receive an email with a link asking you to complete a shorter version of the questionnaire after 3 months. This second questionnaire would take approximately 10-15 minutes to complete.

What are the possible disadvantages and risks of taking part?

There are no potential disadvantages or risks involved in taking part in this study.

What are the possible benefits of taking part?

Whilst there are no direct or immediate benefits for those people participating in this research, it is hoped that this work will contribute to a greater understanding of autism spectrum disorders and help to inform future research.

Will I receive individual feedback about my child?

No, unfortunately, due to the size of the study we won't be able to provide you with individual feedback after taking part in this study. However we will be able to send out information about the overall results of the study once it has been completed. If you would like to receive this information then please indicate this as your preference when asked in the questionnaire.

Appendix F (continued)

What if there is a problem?

If you have any questions or concerns you should speak to the researcher (Kirsty Howell) who will do her best to answer your questions. If at any time you are unhappy about the treatment that you receive from the researcher and wish to make a complaint or raise a concern about the research process you can contact Dr Elizabeth Milne (Project Supervisor) on (0114) 222 6558. If you feel that your complaint has not been handled to your satisfaction, then you can contact the University's Registrar and Secretary.

Will my taking part in this project be kept confidential?

All the information that we collect about you during the course of the research will be kept strictly confidential. We will allocate you a participant ID number so that your answers will be anonymous and stored separately to your contact details. Any paper records will be stored securely within the Department of Psychology. Only members of the research team will view the information gathered. You will not be able to be identified in any reports or publications.

What will happen to the results of the research project?

The results of the study will be analysed by Kirsty Howell and Dr Elizabeth Milne. You will not be individually informed about the final results however you may request a research summary from the researcher. The results are likely to be published in scientific journals and written up as a doctoral-level thesis. No information about any individuals will be available from this report.

Who is organising and funding the research?

The University of Sheffield is carrying out this research study.

Who has ethically reviewed the project?

All research is looked at by an independent group of people, called a Research Ethics Committee to protect your safety, rights, well-being and dignity. This study has been approved by the Research Ethics Committee at the University of Sheffield.

Researcher Contact Details;

You can contact the researcher Kirsty Howell (Trainee Clinical Psychologist) by phone, email or post. Please state that you are calling about 'Developing a measure of autism subtypes' and leave your contact details so that they can return your call as soon as possible.

Telephone: 0114 222 6610

Address:

Email (K.Howell1@sheffield.ac.uk)

Mrs Kirsty Howell, Clinical Psychology Unit,
Dept of Psychology, University of Sheffield,
Western Bank, Sheffield, S10 2TN, UK.

Appendix G. Parent consent form

Parent/Carer Consent Form

If you wish to take part in the study please read the following statements carefully.

- I confirm that I have read and understand the participant information sheet dated 20.09.16 explaining the above research project and I have had the opportunity to ask questions about the project.
- I understand that my participation is voluntary and that I am free to withdraw at any time without giving any reason and without there being any negative consequences. In addition, should I not wish to answer any particular question or questions, I am free to leave it/them out.
- I understand that my responses will be kept strictly confidential.
- I give permission for members of the research team to have access to my anonymised responses.
- I understand that neither my name nor my child's name will be linked with the research materials, and we will not be identified or identifiable in the report or reports that result from the research.
- I agree for the data collected from me to be kept and used anonymously in future research.
- I agree to take part in the above research project.

Please confirm that you agree to the above statements to continue.

If you do not agree to any of the above statements, we appreciate your time and you may now close the browser.

Appendix H. University Ethical Approval

From: **Psychology Research Ethics Application Management System** <no_reply@psychologyresearchethicsapplicationmanagementsystem>

Date: 1 March 2015 at 18:02

Subject: Approval of your research proposal

Your submission to the Department of Psychology Ethics Sub-Committee (DESC) entitled "An Exploratory Study: Development and preliminary psychometric evaluation of a questionnaire to identify autism subtypes." has now been reviewed. The committee believed that your methods and procedures conformed to University and BPS Guidelines.

I am therefore pleased to inform you that the ethics of your research are approved. You may now commence the empirical work.

Yours sincerely,
Prof Paul Norman

