Relations among restricted and repetitive behaviours, anxiety and sensory features in children with autism spectrum disorders

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Abstract

The purpose of this study was to explore how atypical reactions to sensory stimuli contribute to the relation between restricted and repetitive behaviours and anxiety in children with Autism Spectrum Disorders (ASD). In Study 1, factor analysis of restricted and repetitive behaviours was carried out using the Repetitive Behaviour Questionnaire-2 (RBQ-2), completed by 120 parents of 2- to 17-year-olds with ASD. Two subtypes resulted: repetitive sensory and motor behaviours, and insistence on sameness, accounting for 40% of the variance. This two-factor solution was retained even when the sensory items of the RBQ-2 were removed. In Study 2, 49 of the same parents also completed the Spence Anxiety Scales and the Sensory Profile. The insistence on sameness factor was significantly associated with anxiety while the repetitive motor behaviours factor was not. The relation between anxiety and insistence on sameness was mediated by sensory avoiding and to a lesser extent by sensory sensitivity. Implications for arousal explanations of ASD and for clinical practice are discussed.

Keywords:

Arousal, repetitive behaviours, anxiety, sensory features, insistence on sameness, autism spectrum disorders

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1. General Introduction

Restricted and repetitive behaviours (RRBs) are part of the core criteria for autism spectrum disorders (ASD). They form a heterogeneous class of behaviours that are characterised by invariant repetition and desire for sameness in the environment (Kanner, 1943). Factor analytic studies using the Autism Diagnostic Interview-Revised (ADI-R) consistently indicate a division into two subclasses: (a) *repetitive motor and sensory (RSM) behaviours* such as repetitive hand or finger movements and (b) *insistence on sameness (IS)*, including narrow interests, rigid routines, and rituals (Cuccaro et al., 2003; Honey, Rodgers, & McConachie, 2012; Richler, Bishop, Kleinke, & Lord, 2007; Szatmari et al., 2006). Subgroups of RRB have been proposed to represent different neural pathways (Langen, Durston, Kas, Van Engeland, & Staal, 2011) and show different presentations in early typical development (Arnott et al., 2010).

Early theoretical accounts considered the use of RRBs to be a coping mechanism for maintaining a homeostatic state of arousal, with RRBs helping to increase sensory stimulation when an individual is under-aroused and reduce stimulation or soothe when over-aroused (Kinsbourne, 1980; Ornitz & Ritvo, 1968; Zentall & Zentall, 1983). Consistent with this explanation, RRBs are often considered to be a marker for anxiety, forming a buffer to alleviate anxiety and distress in a similar way to the role of RRBs in young children (Evans et al., 1997) and compulsions in obsessive compulsive disorder (Zandt, Prior, & Kyrios, 2007).

To date, evidence supports the claim that RRBs are associated with anxiety in individuals with ASD (Joosten, Bundy, & Einfeld, 2009; Kamp-Becker, Ghahreman, Smidt, & Remschmidt, 2009; Rodgers, Glod, Connolly, & McConachie, 2012; Spiker, Lin, van Dyke, &Wood, 2012; Sukhodolsky et al., 2008; Tantam, 2003). RRBs are also associated with sensory features (Boyd, McBee, Holtzclaw, Barenek, & Bodfish, 2009; Gabriels et al., 2008; Chen, Rodgers, & McConachie, 2009), even after controlling for age and IQ (Boyd et al., 2010; Gabriels et al., 2008). There is also evidence of an association between atypical sensory features and anxiety in ASD (Ben-Sasson et al., 2009; Green & Ben-Sasson, 2010; Green, Ben-Sasson, Soto, & Carter, 2012; Pfeiffer, Kinnealey, Reed, & Hertzberg, 2005). However, little is known about the particular way in which atypical reactions to sensory stimuli contribute to the relation between anxiety and RRBs in children with ASD. Evidence from toddlers with non-specific ASD (PPD-NOS) suggests that the onset of sensory features developmentally precedes the onset of symptoms of anxiety (Green et al., 2012), but as yet the nature of the three way relation between sensory features, anxiety and RRBs remains to be characterised.

One proposal is that different subclasses of RRBs may function in different ways to either increase or reduce sensory stimulation and anxiety (Leekam, Prior, & Uljarevic, 2011). To date only one published study has examined the relation between anxiety and each of the RSM and IS subclasses of RRBs. Using the 38-item Spence Children's Anxiety Scale-Parent version (SCAS-P; Spence, 1998) and the 33-item Repetitive Behaviour Questionnaire (RBQ; Turner, 1995) with 8- to 16-year-olds with ASD, Rodgers et al. (2012) found a significant association between total anxiety score and IS. This association was especially strong among children meeting the cut-off score indicating clinical levels of anxiety. However RSM behaviours did not significantly relate to anxiety. More recently, Gotham et al. (2013) also explored the relationship between anxiety and IS in a very large sample of 5- to 18-year-olds, using the anxiety problems score of the Child Behaviour Checklist, derived from six items

based on DSM-IV criteria for anxiety disorders (Achenbach & Rescorla, 2001), and an IS score based on six items from the ADI-R (Rutter, Le Couteur, & Lord, 2003). Although this study found a weaker relation between IS and anxiety than shown in the Rodgers et al. study, nevertheless a modest and statistically significant relation existed. In this study, the relation between anxiety and RSM was not reported. Similarly, while a study of repetitive motor behaviours in ASD found that an elevated RRB score is associated with both sensory under-responsiveness and over-responsiveness (Gal, Dyck, & Passmore, 2002), this study did not include measures of either anxiety or IS.

The purpose of the current study was to explore the potential contribution made by atypical sensory features to the anxiety-RRB relation in children with ASD. Standard RRB questionnaire measures traditionally include a number of sensory items as these are part of the category of RRBs within diagnostic criteria for DSM-IV-TR (American Psychiatric Association, 1994). To enable comparison with previous research, in Study 1 we tested the structure of a RRB questionnaire, the Repetitive Behaviour Questionnaire-2 (RBQ-2; Leekam et al., 2007), which includes a set of sensory items. We then removed these items in a second factor analysis. Study 1 also analysed associations with age given longitudinal evidence of age changes in childhood using the ADI-R (Richler, Huerta, Bishop, & Lord, 2010). In Study 2, to avoid artificially inflating relations with other sensory measures, we studied the relation between these non-sensory RRB items and anxiety, and also an independent measure of sensory atypicality taken from the Sensory Profile (Dunn, 1999).

2. Study 1

Study 1 examined the factor structure of the RBQ-2 questionnaire, using this measure for the first time with a sample of children with ASD. To date, published data on the factor analytic structure and psychometric properties of the RBQ-2 has been confined to a typically developing sample at 15 and 24 months of age (Arnott et al., 2010; Leekam et al., 2007), with results showing that the items group into two factors (RSM and IS). Study 1 aimed to replicate this finding with an ASD sample, firstly using the full RBQ-2 and secondly using the RBQ-2 with sensory items removed.

2.1. Method

2.1.1. Participants

Parents of 120 children with ASD (110 males, 10 females), with ages ranging from 2 years 5 months (2;5) to 17;9 (*M* 7;7, *SD* 3;10), participated as part of their involvement in a research study being carried out in two different parts of the UK (South Wales, $n = 59$, and the South East of England, $n = 61$) investigating the association between RRB and other factors. The South Wales sample was aged 2;5 to 17;9 (*M* 9;11, *SD* 4;4, 54 males, 5 females) and the South East England sample was aged 2;9 to 8;5 (*M* 5;4, *SD* 1;2, 56 males; 5 females). All had a community multidisciplinary team assessment leading to a best estimate clinical diagnosis of an ASD (including autism and Asperger syndrome) according to DSM-IV-TR (American Psychiatric Association, 1994) and ICD-10 (World Health Organization, 1993) criteria. Children with a recognised medical condition such as epilepsy, brain injury, cerebral palsy or a known genetic condition (e.g., Fragile X, Down syndrome) were excluded. Both were opportunity samples studied via a set of parental postal questionnaires (described below).

2.1.2. Procedure and Measures

Ethical approval for the study was provided by Research Ethics Committees of Cardiff and Cambridge Universities. Both samples were recruited through local schools and parent support groups. Parents were contacted via email and telephone by the researcher. Those whose children met the inclusion criteria were then sent a set of questionnaires including the RBQ-2. Most parents completed and returned the questionnaires by post. A small proportion (16%) chose to visit the university and complete the questionnaire as part of the visit, but without assistance from a researcher.

2.1.2.1. Repetitive Behaviour Questionnaire 2. The RBQ-2 (Leekam et al., 2007) is a 20-item questionnaire that is a modified form of the 33-item RBQ (Honey, McConachie, Turner, & Rodgers, 2012; Turner, 1995). It includes 13 items found in both the RBQ and the Diagnostic Interview for Social and Communication Disorders (DISCO; Wing, Leekam, Libby, Gould, & Larcombe, 2002), plus five DISCO-only items and two RBQ-only items. In a study of typically developing children studied at ages 15 months and 2 years, there was good internal consistency for the total scale and for each subscale (Arnott et al., 2010; Leekam et al., 2007). The items are shown in Table 1. Response choices, based on current behaviour (in the last month), are combined into three alternatives for each item (1: *never/rarely*; 2: *mild/occasional*; 3: *marked/notable*). Following the scoring of previous published studies, a Total score (mean score 1.00 to 3.00) is calculated for each child by adding the responses for each item completed in the questionnaire and dividing by the number of questions completed by the respondent.

Initial analysis using *t* tests revealed that the two samples did not differ in their Total score (South Wales: *M* 1.99, *SD* 0.38; South East England: *M* 1.92, *SD* 0.44; $t(118) = 0.99$, $p = .33$). Furthermore, although the two groups differed in age, the Total RBQ-2 score was not associated with age, $r = -.02$, $p = .84$. Factor analysis was then carried out using the combined dataset.[1] All questionnaires were fully completed with no missing data.

2.2. Results

For every RBQ-2 item, the *mild/occasional* to *marked/notable* response range was endorsed for at least 10% of the sample. Factor analysis was carried out using Principal Components Analysis (PCA) with varimax rotation. Item 20, which asks about the child's flexibility in self-chosen activities, was excluded so that the items in the factor analysis were identical to those used in the previous factor analysis (Leekam et al., 2007). Initial screening showed that assumptions of non-multicollinearity, sampling adequacy, and factorability were all met. An initial analysis was run to obtain Eigen values for each component in the data. Horn's (1965) parallel analysis (PA) was used using the Parallel Analysis Program (http://www.statstools.net/Parellel_Pgm) which is based on the Monte Carlo simulation of random production of Eigen values to determine the number of components. Results showed that factors 3 onwards had Eigen values less than that of those from simulations and a two-factor solution should be retained in the final analysis. The PCA with varimax rotation was rerun specifying a two-factor solution. Factor loadings for items were set at .38 as previously (Leekam et al., 2007). Table 1 shows the final two-factor solution for the present study, which accounted for 40.1% of the variance (RSM 11.1% and IS 29.2%).

[1]Footnote. As in Leekam et al. (2007) and Arnott et al. (2010), data were positively skewed and logarithmic transformation was applied before comparing scores for each sample by *t* test.

No item loaded on both factors. Only two items (*arranging objects* and *carrying around objects*) did not reach a factor loading of .38 for either factor. A further two items loaded in the opposite way than they had for typically developing children in previous research (Leekam et al., 2007). Of these, one item that loaded on the RSM factor for typically developing children (*interest in smell*), loaded on the IS factor for the ASD sample, and another item (*eating same/small range of foods*), which loaded onto the IS factor for typically developing children, loaded onto the RSM factor for ASD children. Apart from these four items, all items that loaded for the original typically developing child sample (Leekam et al., 2007), loaded in exactly the same way for children with ASD. There were two additional RBQ-2 items, that had not loaded for the original sample of typically developing children (*hoarding objects* and *fascination with specific objects*), both of which loaded for the ASD children onto the IS factor and were retained.

Cronbach's alpha (α) showed that internal consistency was high for the Total RBQ-2 scale (α =.86 for 20 items; α = .86 for 19 items) and for each RSM and IS subscale separately (RSM α = .79; IS α = .83). For the RSM factor the mean item-correlation was .63, *SD* .05, range .56 to .71, and for the IS factor the mean item-correlation was .67, *SD* .11, range .50 to .82 (Appendix, Table 5).

The mean Total score (which, like the subscales, has a possible range of 1.00 to 3.00) was 1.96 (*SD* 0.41); the mean RSM subscale score was 1.89 (*SD* 0.51) and the mean IS subscale score was 2.02 (*SD* 0.51). These mean scores were higher than shown in the published results for typically developing children (Leekam et al., 2007) which were, for 15-month olds, Total 1.62 (*SD* 0.30); RSM 1.83 (*SD* 0.43); IS 1.37 (*SD* 0.32), and for 24 month-olds, Total 1.55 (*SD* 0.33), RSM 1.52 (*SD* 0.40), IS 1.54 (*SD* 0.42).

In order to examine the structure of the questionnaire without the sensory items, a further factor analysis was carried out, excluding the sensory-related items shown in Table 1 from the RSM subscale. The sensory items were item number 8 (*angles*), 9 (*smell*), 10 (*feel*), 18 (*clothes*) and also item 7 (*fascination*) and 19 (*foods*) described above. Results showed that when sensory items were excluded, the two-factor solution was retained with the Repetitive Motor Behaviours (RMB) component explaining 17.1% of the variance and the IS factor explaining 36.9% of the variance (Table 2). The internal consistency (α) was .76 for the RMB subscale, .83 for the modified IS subscale and .77 for the total score.

2.3. Discussion

The results indicate that the modified form of RBQ, the 20-item RBQ-2, provides a suitable measure of RRB not only for typically developing children (Arnott et al., 2010; Leekam et al., 2007) but also for children with ASD aged from 2 to 17 years, with good internal consistency. Total repetitiveness scores exceeded the published RBQ-2 repetitiveness scores of typically developing 15- and 24-month-olds (Arnott et al., 2010; Leekam et al., 2007). However, the factor structure of RRBs for the two groups was very similar, resulting in two factors of RSM and IS. Of the 19 items entered in the factor analysis, only a minority did not load in the same way as for the original factor analysis carried out with typically developing children (Leekam et al., 2007). Some of the differences might be explained in terms of the developmental level of each sample, given that the ASD sample was older than the original published sample. For example, two items (*carrying around objects* and *arranging objects*), commonly seen in typical toddlers and infants (59% and 64% of children respectively in Leekam et al., 2007), did not load sufficiently highly to be included for the ASD sample, while two other items (*hoarding objects* and *fascination with specific objects*), which are common in older children, loaded in the factor analysis for ASD children but not in the original published study. Apart from these four items that might be explained by their

developmental appropriateness, only two further items (*interest in smell* and *eating the same/small range of foods*) loaded onto different factors for the two samples, although both might be considered to have a mixed interpretation relevant to both sensory and restricted interests. Furthermore, even when these two mixed items and all other sensory-related items were removed from the analysis, the two-factor structure was retained, producing an RMB factor and an IS factor. This two-factor structure is similar to that found with studies of the ADI-R (Bishop, Richler & Lord, 2006; Cuccaro et al,, 2003; Richler et al., 2007), and RRB studies using questionnaire methods (Honey, Rodgers, & McConachie, 2012). Further research could investigate the utility of questionnaire measures of this kind within the diagnostic process; for example, they may provide a supplement to clinical interviews such as the DISCO which also asks about specific separate behaviours.

3. Study 2

Study 2 examined the association between the two subtypes of RRB and anxiety and the contribution made by sensory features to this association using the Sensory Profile (Dunn, 1999).

3.1. Method

3.1.1. Participants

As this study focused on the associations among RRB, anxiety, and sensory features, the sample was restricted to a subset of participants from Study 1 who had completed the RBQ-2 and had also completed the Sensory Profile, a Spence Anxiety Scale and a language questionnaire. All were from the South Wales sample. Only parents whose children were aged 3 years or more were included (as the Sensory Profile is not valid for 2-year-olds). The sample comprised 49 children and adolescents (45 males, 4 females) aged 3 years 0 months (3;0) to 17;9 (*M* 10;7, *SD* 3;10). As described above, all had a clinical diagnosis of an ASD, established according to DSM-IV-TR and ICD-10 criteria. Scores for the Social Communication Questionnaire (SCQ; Rutter, Bailey & Lord, 2003), an autism screening questionnaire for parents, were available for 43 children who had a developmental level in the appropriate range for this scale. The mean score was 27.63 (SD=5.82), range was 14-37. Three children scored 14, one scored 16 and 39 scored 21 or above. A score of 11 has been used in previous validity studies of the SCQ (e.g. Allen, Silove, Williams & Hutchins, 2007) as a cut-point to indicate ASD. Exclusion criteria were as for Study 1. A language questionnaire provided an estimate of expressive language level. The questionnaire is based on language items taken from the DISCO (Wing et al., 2002). It has been used in a previous questionnaire study on RRBs by Honey, Leekam, Turner, and McConachie (2007). Parents are asked to report if their child has (in their expressive language) no words, single words, 2- to 3-word phrases, longer phrases, spontaneous sentences, or complex sentences with past, present and future tense, with these six categories corresponding to scores of 0 (no words) to 5 (complex sentences with past, present and future tense). Of the 48 parents who completed this questionnaire, 36 (75%) reported that their child used complex grammatical speech or spontaneous sentences, 8 (17%) that their child used phrase speech and 4 that their child used single words or no speech (8%).

3.1.2. Procedure and Measures

The procedure was the same as for Study 1. Ethical approval for the study was given by Cardiff University School of Psychology Research Ethics Committee.

3.1.2.1. Repetitive Behaviour Questionnaire-2 (RBQ-2). Parents of all children completed the full RBQ-2. To avoid sensory items within the RBQ-2 artificially inflating relations with sensory features, the sensory items were excluded in the analysis. The RMB score was the mean of items 2 to 6 (*fiddle*, *spin*, *rock*, *pace*, *flap*), and the IS score was the mean of items 12 to 17 (*hoard items*, *sameness at home*, *upset by minor changes*, *daily routines*, *just right*, *same activity*) (items shown in Table 2). The Total score (excluding sensory items) was used as described in Study 1.

3.1.2.2. Sensory Profile. All parents completed the Sensory Profile (Dunn, 1999), a 125-item caregiver-report measure of an individual's reactions to everyday sensory experiences. The Sensory Profile has established reliability and validity (Dunn, 1999). Caregivers indicate the frequency of their child's response to particular sensory experiences on a 5 point Likert scale ranging from *never* to *always*. Sensory features are measured using Dunn's (1997) model of sensory processing, in which an individual's sensory profile is defined by their scores on four quadrants which are determined by two dimensions; a *neurological threshold* (low or high) and a *behavioural response* (passive or active). A high threshold combined with a passive response is described as *low registration*, and a high threshold with an active response is described as *sensation seeking*. In contrast, a low threshold combined with passive response is described as *sensory sensitivity* and a low threshold with an active response is described as *sensation avoiding*. These quadrants reflect an individual's pattern of responding across modalities. The Sensory Profile is scored so that higher scores indicate typical performance and lower scores indicate atypical performance. Preliminary analysis established that the excluded sensory items from RBQ-2 correlated with each sensory quadrant (low registration: $r = -.37$, $p = .01$; sensation seeking: $r = -.70$, $p < .001$; sensory sensitivity: $r = -.49$, $p < .001$; sensation avoiding: $r = -.26$, $p = .07$), justifying their exclusion from subsequent analysis, although the Cronbach's alpha (α) for the sensory item set of RBQ-2 was low (.58).

3.1.2.3. Spence Anxiety Scales. Parents of 7- to 17-year-olds ($n = 34$) completed the Spence Children's Anxiety Scale-Parent Version (SCAS-P; Spence, 1998) and parents of younger children ($n = 15$), the preschool version (Preschool Anxiety Scale, PAS; Spence, Rapee, McDonald, & Ingram, 2001). These questionnaires assess a child's anxiety across the main DSM-IV-TR categories of anxiety disorder. The SCAS-P has established convergent and divergent validity: Nauta et al. (2004) found that the SCAS-P correlated well with parent-reported internalising symptoms and more weakly with externalising symptoms. The measure differentiated between children with anxiety disorders and controls. Parents are asked to rate on a Likert scale how often each of the symptoms happens for their child. Each version generates a total score which reflects the frequency of anxiety symptoms occurring across the DSM-IV-TR categories. SCAS-P scores ($\alpha = .91$) and PAS scores ($\alpha = .76$) were converted into standardised T scores (see www.scaswebsite.com).

3.1.3 Analysis

Given the wide age range of this study and inconsistency in the literature regarding the link between age and both anxiety (van Steensel, Bögels, & Perrin, 2011) and RRB (Esbensen, Seltzer, Lam, & Bodfish, 2009; Richler et al., 2010), preliminary analyses were first carried out to examine associations with age. A series of analyses was then carried out to investigate relations between RRBs, anxiety, and sensory features. First we replicated the approach taken by Rodgers et al. (2012): Children were first allocated into one of two groups according to possible clinical caseness based on their anxiety T score and then group

differences in RRB and sensory features were assessed. Finally, we examined relations among RRB, anxiety, and sensory features using a series of partial correlations and mediation analyses using Baron & Kenny's (1986) procedure.

3.2. Results

3.2.1. Age

Age was not significantly related to anxiety, r= .16, p =. 28, nor to RMB, r= -.17, p = .25, IS, r= .06, p = .68, or RBQ-2 Total score, r= -.16, p = .26 (sensory items excluded). Nor was it significantly associated with the Sensory Profile quadrants (low registration*:* r= .06, p = .71; sensory sensitivity: r= .07, p = .64; sensation avoiding: r= -.19, p = .18) except for sensation seeking, r= .38, p = .007, with higher ages related to lower levels of sensation seeking.

3.2.2. Restricted and Repetitive Behaviours and Anxiety

Anxiety was positively correlated with RBQ-2 Total score, r= .41, p = .004, and IS (without sensory items), r= .46, p < .001, but not with RMB, $r(49)$ = .24, p = .10.

Twenty-four children (49%) scored above indicative clinical cut-off for anxiety (a proportion equivalent to that found by Rodgers et al., 2012, n = 33, 49%). The two groups were compared in terms of age, language level, RRB, and sensory scores (Table 3). As found in Rodgers et al., the groups did not differ in age or language level. The anxious group had significantly higher scores for the IS factor than the non-anxious group. However there was no significant difference between anxiety groups in RMB.

3.2.3. Restricted and Repetitive Behaviours, Anxiety, and Sensory Features

Table 3 shows that the anxious and non-anxious groups differed markedly on sensory sensitivity and sensation avoiding, the two sensory quadrants reflecting low neurological thresholds. Effects were smaller for the other two quadrants and the difference was nonsignificant in the case of sensation seeking. Table 4 shows the three-way associations between anxiety, RRBs and sensory quadrant scores using continuous score ranges. Alpha level was adjusted to .01 to reduce the probability of a Type I error. RMB did not correlate with anxiety but did correlate with sensation seeking and with sensation avoiding. IS was associated with all sensory quadrants and with anxiety. Anxiety was associated with sensory sensitivity, sensation avoiding, and low registration, but not with sensation seeking.

A series of partial correlations explored which sensory quadrants contributed to the significant relation between anxiety and IS. When either sensory sensitivity or sensation avoiding was controlled for, the significant association between anxiety and IS disappeared,. However, when either low registration or sensation seeking were controlled, the association between anxiety and insistence on sameness was relatively unaffected (r= .37, p = .01 after controlling for low registration; r= .36, p =.01 after controlling for sensation seeking) and the correlation remained moderately strong even when both were entered simultaneously, r= .32, p = .02.

To examine in more detail the contribution of sensory sensitivity and sensation avoiding to the relation between anxiety and insistence on sameness, mediation models were tested. Data screening revealed no skewness or outliers. Sensory sensitivity and sensation avoiding quadrant scores were analysed separately due to collinearity. For the first analysis focusing on sensory sensitivity, the first two regressions showed that both IS (F(1,47) = 12.70, R^2 = .196, p = .001) and sensory sensitivity (F(1,47) = 28.17, R^2 = .361, p < .001) were

predicted by anxiety. IS was also predicted by sensory sensitivity ($F(1,47) = 10.50$, $R^2 = .183$, $p = .002$).When the IS-anxiety relation controlled for sensory sensitivity, the IS-anxiety relation weakened and became only marginally significant ($F(2,46) = 7.52$, $R^2 = .214$, R^2 change = .064, $p = .06$). For the second analysis focusing on sensation avoiding, the first two regressions also showed that both IS ($F(1,47) = 12.70$, $R^2 = .196$, $p = .001$) and sensation avoiding ($F(1,47) = 46.81$, $R^2 = .488$, $p < .001$) were predicted by anxiety. IS was also predicted by sensation avoiding ($F(1,47) = 15.23$, $R^2 = .229$, $p < .001$). However, the significant IS-anxiety relation disappeared when sensation avoiding was entered ($F(2,46) = 8.49$, $R^2 = .238$, R^2 change = .025, $p = .22$).

Finally, the analyses above were rerun, removing any items in the Sensory Profile that could be interpreted as measures of RRB or anxiety (item numbers: 26, 27, 28, 93, 103, 104 and 114). The pattern of results was unchanged (Appendix, Tables 6 and 7).

3.3. Discussion

Study 2 showed that anxiety was significantly associated with IS behaviours such as routines, rituals, hoarding and dislike of change, but not with RMBs such as fiddling, pacing and spinning, replicating the pattern of results found by Rodgers et al. (2012) in a different sample of children in a different geographical region. As found by Rodgers et al., this pattern of relationships was not explained by age or language level.

Further replication of the differential association between anxiety and the two types of RRB is needed using other measures, given that Rodgers et al. (2012) used a very similar questionnaire measures. Also the sample size in both this study and in Rodgers et al. was small, and in both groups the majority of children were verbal and/or had higher levels of functioning. It is possible that a larger sample may yield a stronger correlation between RMB and anxiety. Nevertheless, the same results were found even though, in the current study, the RMB subscale included only motor behaviours with sensory items removed, and even though the present study included some children who were younger and less able than those in Rodgers et al.'s study. The partial correlations and mediation analyses gave insights into the influence of sensory features on the IS-anxiety relation. Low registration and sensation seeking were related to both IS and anxiety but partial correlation analyses indicated that these sensory features did not explain the relation between IS and anxiety; that is, when these variables were entered into a partial correlation, the anxiety-IS relation did not substantially change. In contrast, sensation avoiding in particular (and, to a lesser extent, sensory sensitivity) appeared to play a mediating role in the association between anxiety and IS. These results indicate that different sensory features contribute in different ways to the association between anxiety and RRB.

4. General Discussion

Recent explanations of RRBs have described their function as a marker for anxiety, forming a buffer to alleviate distress. It has also been proposed that different subclasses of RRBs may function in different ways to either increase sensory stimulation or to reduce both anxiety and sensory stimulation. This research investigated the potential relationships between different types of RRBs, anxiety and sensory features in order to identify the contribution made by sensory features to the anxiety-RRB relation. Using the RBQ-2, for the first time in a sample of children with ASD, a two-factor structure of RRBs was identified, a finding previously reported using other measures of RRBs in ASD and using the RBQ-2 in non-ASD samples. Results also supported previous findings (Rodgers et al., 2012) indicating that anxiety is related to particular types of RRB. Anxiety is related to IS behaviours, such as

routines and rituals and narrowly focused interests, but not to RMB, such as hand flapping and spinning.

Is it possible to conclude that the relation between IS and anxiety is explained by sensory reactivity? This would support the original view of Ornitz & Ritvo (1976) and Ornitz (1974) of ASD being associated with fluctuating states of under-arousal and over-arousal, and RRB representing ongoing attempt to correct imbalances in arousal to achieve optimal stimulation (Zentall & Zentall, 1983). Correlational analyses prevent any conclusions being made about direction of causality, but the mediation analysis results using the Sensory Profile do indicate a positive answer to this question. It might therefore be speculated that different types of repetitive behaviours serve different functions for modulating arousal with varying levels of success. IS behaviours, which are linked to arousal by sensory sensitivity and sensation avoiding, may function to narrow sensory input. This is consistent with findings by Joosten & Bundy (2010) that sensory sensitivity and the tendency to avoid sensory stimuli help to explain anxiety in ASD. However the link between these IS behaviours and anxiety indicates that these behaviours may not provide an optimal strategy for regulating arousal and they may even serve to create and maintain anxiety. Alternatively, RMB may serve a more effective function for regulating arousal without creating or maintaining anxiety, by providing stimulation through sensation seeking in the case of underarousal and through soothing and avoidance outcomes in the case of overarousal.

Further research is needed to test the direction of relation between RRB subtypes and sensory features. It will be important to develop and test alternative theoretical models using more complex mediation models and experimental research designs. For example it is possible that IS is either a reaction to anxiety or co-occurs simultaneously with anxiety. These alternatives also need to be considered in future longitudinal studies. It has been proposed that the developmental trajectory for anxiety is one in which sensory features developmentally precede anxiety (Green et al., 2012). An important consideration is that the developmental trajectory of anxiety may be influenced by the onset and maintenance of different types of RRB. The function of RRB subtypes and their effect on anxiety will also be important aspects to consider when developing models of atypical arousal regulation and attention (Keehn, Müller, & Townsend, 2013) and when developing clinical interventions. Given that sensory features, RRB, and anxiety each represent targets for intervention, understanding of the complex inter-relationships between these constructs may serve to further enhance the specificity of interventions as well as the sensitivity of outcome measures.

The findings from this study also have important clinical implications for diagnostic assessment. The results of the RBQ-2 factor analysis results identified the RBQ-2 as a suitable measure of RRB not only for very young children with typical development but also for children and adolescents with ASD across a wide age range. The work indicates the potential of the RBQ-2 as a useful questionnaire for clinical practice. Eighteen of its 20 items come from a semi-structured clinical diagnostic tool, the DISCO, and these RRB items are included within the DISCO diagnostic algorithms for ICD-10 and DSM-5 (Kent, Carrington et al., 2013). It is possible that these items, already drawn from a clinical interview tool, could function as a stand-alone supplement to a diagnostic interview with parents completing the questionnaire before coming to the clinic for a full developmental history and interview assessment.

Further research is needed before a recommendation could be made to use the RBQ-2 in clinical settings. First, research is needed with different participant groups, including girls with ASD instead of the predominantly male sample included here, that are compared with other clinical comparison groups. Second, all developmental ability levels, including intellectual disability should be represented. A convergent validity analysis is also needed, comparing different measures, both of a questionnaire format (e.g., RBS-R) and across parent

interviews (DISCO and ADI-R). Meanwhile, however, the results indicate the potential value of collecting RRB information through parent questionnaires.

Studies using questionnaires have some definite limitations. Questionnaires cannot investigate causal predictions about the role of sensory features, and the complex interactions of co-occurring problems of anxiety and RRBs in individuals with ASD, are difficult to disentangle. The interpretation of the results is also constrained by the small sample size and verbal abilities of the children. Despite these limitations, the findings suggest directions for future work in order to progress our understanding of these exploratory results. Data from future longitudinal designs that trace development from the early years will be informative in identifying the pathway of emergence of sensory atypicality in relation to different types of RRBs and anxiety in children with ASD.

4.1. Conclusion

This research using a new questionnaire measure, the RBQ-2, supports previous research suggesting that anxiety in children with ASD is related to a particular type of RRB, insistence on sameness, and suggests that sensory features explain this relation.

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Highlights

- Factor analysis using the RBQ-2 resulted in two subtypes of repetitive behaviour
- One subtype, insistence on sameness (IS), was found to be associated to anxiety
- The association between IS and anxiety was mediated by sensory features

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Table 1

Factor Structure of RBQ-2 Questionnaire Items

Questionnaire items within each factor	Item factor loading
Factor 1: Repetitive sensory and motor behaviours – 11.1% variance	
2. Repetitively fiddle with toys etc.	.683
3. Spin self around and around	.639
4. Rock backwards and forwards	.617
5. Pace/move around repetitively	.697
6. Repetitive hand/finger movements	.660
8. Looks at objects from particular/unusual angles	.571
10. Special interest in feel of different surfaces	.565
19. Insists on eating same foods or small range of foods	.447
Factor 2: Insistence on sameness – 29.2% variance	
7. Fascination with specific objects	.390
9. Special interest in smell of people/objects	.484
12. Collect or hoard items of any sort	.579
13. Insists on things (e.g. in house) remaining the same	.777
14. Gets upset about minor changes to objects	.716
15. Insists on aspects of routine remaining the same	.741
16. Insists on doing or re-doing things in a certain way	.829
17. Plays same music, game, video, book repeatedly	.594
18. Insists on wearing same clothes/refuses new clothes	.569

Note. Item 1 (arranging objects into patterns or rows) and item 11 (has special objects that likes to carry around) had loadings on both factors of less than .360.

Table 2

Factor Structure of RBQ-2 Questionnaire Items Resulting From Analysis With Sensory Items Excluded

Questionnaire items within each factor	Item factor loading
Factor 1: Repetitive behaviours – 17.1% variance	
2. Repetitively fiddle with toys etc.	.687
3. Spin self around and around	.607
4. Rock backwards and forwards	.721
5. Pace/move around repetitively	.785
6. Repetitive hand/finger movements	.706
Factor 2: Insistence on sameness – 36.9% variance	
12. Collect or hoard items of any sort	.550
13. Insists on things (e.g. in house) remaining the same	.816
14. Gets upset about minor changes to objects	.730
15. Insists on aspects of routine remaining the same	.785
16. Insists on doing or re-doing things in a certain way	.860
17. Plays same music, game, video, book repeatedly	.616

Table 3

Comparison of Anxious and Non-Anxious Groups[a]

	Anxious (n = 24)	Non-anxious (n = 25)	t test		Cohen's d
	M (SD)	M (SD)	t	p	
Background variables					
Age (months)	135.7 (43.0)	120.6 (49.4)	-1.14	.26	0.32
Language level	5.4 (0.9)	4.6 (2.1)	-1.72	.10	0.49
Repetitive behaviours					
Repetitive motor behaviours	1.86 (0.55)	1.83 (0.54)	-0.17	.87	0.05
Insistence on sameness	2.37 (0.43)	2.03 (0.42)	-2.79	.008	0.80
RBQ-2 Total	2.15 (0.42)	1.95 (0.41)	-1.71	.09	0.48
Sensory quadrants					
Low registration	46.3 (11.6)	52.6 (10.0)	2.02	.05	0.57
Sensation seeking	86.4 (19.1)	93.2 (14.9)	1.40	.17	0.40
Sensory sensitivity	57.4 (15.5)	70.1 (11.3)	3.28	.002	0.94
Sensation avoiding	77.9 (14.5)	94.2 (12.5)	4.24	<.001	1.21

Note. [a]Groups defined by the Spence Anxiety Scale cutoffs for possible clinical caseness.

Table 4

Correlations Among RBQ-2, Sensory Profile Quadrants, and Anxiety

	Repetitive motor behaviours	Insistence on sameness	Anxiety
Low registration	-.21	-.38**	-.40**
Sensation seeking	-.42**	-.49**	-.34
Sensory sensitivity	-.31	-.43**	-.61**
Sensation avoiding	-.42**	-.49**	-.71**
Anxiety	.24	.46**	-

Note. Alpha level adjusted to .01 to reduce the probability of Type I error.
** $p < .01$, *** $p < .001$

APPENDIX

Table 5
Correlated Item-Total Correlations for the Two Factors of the RBQ-2

RBQ-2 Items	Factors	
	RSM	IS
Repetitive sensory and motor (RSM) behaviours		
2. Repetitively fiddle with toys or other items	.677***	.252**
3. Spin self around and around	.627***	.193*
4. Rock backwards and forwards	.606***	.235*
5. Pace/move around repetitively	.709***	.279**
6. Repetitive hand/finger movements	.676***	.291**
8. Looks at objects from particular/unusual angles	.573***	.226*
10. Special interest in feel of different surfaces	.635***	.407***
19. Insists on eating same foods or small range of foods	.563***	.336***
Insistence on sameness (IS)		
7. Fascination with specific objects	.379***	.503***
12. Collect or hoard items of any sort	.153	.606***
13. Insists on things (e.g. in house) remaining the same	.372***	.779***
14. Gets upset about minor changes to objects	.318***	.716***
15. Insists on aspects of routine remaining the same	.312**	.739***
16. Insists on doing or re-doing things in a certain way	.325***	.820***
17. Plays same music, game, video, book repeatedly	.330***	.610***
18. Insists on wearing same clothes/refuses new clothes	.231*	.584***

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

Table 6
Differences Between Anxious and Non-Anxious Groups[a] *on the Modified Sensory Profile Quadrants*[b]

	Anxious (*n* = 24)	Non-anxious (*n* = 25)	*t* test		Cohen's *d*
	M (*SD*)	*M* (*SD*)	*t*	*p*	
Low registration	46.3 (11.6)	52.6 (10.0)	2.02	.05	0.57
Sensation seeking	75.9 (17.5)	82.0 (13.2)	1.38	.18	0.41
Sensory sensitivity	57.4 (15.5)	70.1 (11.3)	3.28	.002	0.94
Sensation avoiding	69.5 (12.7)	82.9 (10.7)	4.01	<.001	1.17

Note. [a]Groups defined by the Spence Anxiety Scale cutoffs for possible clinical caseness. [b]Item numbers 26, 27, 28, 93, 103, 104, and 114 were excluded as they could be interpreted as including some measure of RRB or anxiety.

Table 7

Correlations Between RBQ-2, Modified Sensory Profile Quadrants,[a] and Anxiety

	Repetitive motor behaviours	Insistence on sameness	Anxiety
Low registration	-.21	-.38**	-.40**
Sensation seeking	-.31	-.43**	-.33
Sensory sensitivity	-.31	-.43**	-.61**
Sensation avoiding	-.39**	-.45**	-.69**
Anxiety	.24	.46**	-

Note. Alpha level adjusted to .01 to reduce the probability of Type I error.
[a]Item numbers 26, 27, 28, 93, 103, 104, and 114 were excluded as they could be interpreted as including some measure of RRB or anxiety.
** $p < .01$, *** $p < .001$