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### Abstract

In order to investigate the experience of anxiety and restricted and repetitive behaviours (RRB) in young people with ASD, nineteen families with young people with ASD aged between 13 and 20 years completed questionnaire measures of RRB, anxiety, and intolerance of uncertainty. Ten young people also completed a novel semi-structured interview exploring an individualised example of an RRB. Findings demonstrated that young people with ASD can self-report and show insight in to their RRB, and replicated previous findings based on parent report showing a significant positive relationship between RRB and anxiety. This is the first evidence of young person self-report using both quantitative and qualitative data and indicates a range of reasons why young people may engage in RRB.

**Key words:** ASD, anxiety, restricted and repetitive behaviours, intolerance of uncertainty, self-report

Restricted and repetitive behaviours (RRB) are a central feature of ASD (APA, 2013). The term RRB covers a large range of behaviours which can be separated in to two subtypes; repetitive sensory motor (RSM) such as excessive smelling or touching of objects, or repetitive hand mannerisms and insistence on sameness (IS) behaviours such as inflexible adherence to routines, or ritualized patterns. The two subtypes are also sometimes referred to as lower order RRB and higher order RRB respectively (Bishop et al., 2013). Although there may be different explanations for the cause of RRBs (see Leekam, Prior & Uljarevic, 2011 for review), in recent years research attention has focused on the association between RRB and anxiety.

Further understanding is needed about the experience of anxiety in young people with ASD and how this may relate to RRB. Although we are gaining increasing insights about subjective experiences of anxiety in ASD, our knowledge of RRBs to date has relied almost exclusively on parent report. Parent report of RRB is likely to be influenced by observable behaviours, yet a range of personal experiences, either positive or negative are an intrinsic part of the insistence on sameness and sensory and motor behaviours and parents may not be aware of all the RRBs experienced by their children. Understanding further the experiential nature of RRBs has the potential for a positive impact on young people's lives through the development of effective interventions.

Raised levels of anxiety in ASD have been associated with a greater presence of RRB in a number of studies (Gotham et al. 2013; Lidstone et al. 2014; Rodgers, Riby, Janes, Connolly, & McConachie, 2012). This relationship is yet to be well understood and there may be different explanations for the link (Leekam et al. 2011; Wood & Gadow, 2010). For example findings suggest that RRB may potentially act as a coping response to reduce anxiety (Joosten, Bundy, & Einfeld, 2009; Spiker, Lin, Van Dyke, & Wood, 2012), the

behaviours may potentially elicit anxiety themselves (Sofronoff, Attwood, & Hinton, 2005) and/or anxiety may potentially act as a moderator exacerbating RRB (Wood & Gadow, 2010).

Some studies have shown that anxiety in children with ASD is selectively associated with a particular type of RRB. Rodgers, Glod, Connolly, & McConachie (2012) for example found a specific association between higher levels of anxiety and a subtype of RRBs often called insistence on sameness (IS), which includes routines, rituals and unusual interests. These results suggest that anxiety may underpin engagement in RRB, supporting the idea that IS RRB may act as a coping response to anxiety. A study of parent reported anxiety in 2-17year olds with ASD, also found anxiety to be significantly and selectively associated with IS behaviours, but not with repetitive sensory and motor behaviours (RSM) such as fiddling, pacing and spinning (Lidstone, Uljarevic et al., 2014). The authors also found that the relation between anxiety and IS behaviours was mediated by sensory avoiding and to a lesser extent by sensory sensitivity.

A key factor in the RRB-anxiety-sensory responsiveness relationship is the presence of intolerance of uncertainty. Intolerance of uncertainty (IU) refers to an individual's negative perception of uncertain situations, resulting in high levels of stress, worry and avoidance (Dugas, Gagnon, Ladouceur, & Freeston, 1998). Given evidence of an association between IU and anxiety in the typically developing population, researchers have been investigating this association in young people with ASD. IU has been found to be a recognisable construct in children with ASD, as identified by parents (Hodgson, Freeston, Honey & Rodgers, 2016). Evidence suggests that it may act as a mediator between ASD symptoms and anxiety with higher levels of IU in ASD accounting for the greater rates of anxiety associated with the disorder (Boulter, Freeston, South & Rodgers., 2014).

Furthermore, Wigham, Rodgers, South, McConachie, and Freeston (2014) investigating the

interplay between sensory processing abnormalities, IU, anxiety and RRB in ASD, found evidence for pathways involving IU and anxiety from both over and under sensory responsivity to both types of RRB. This analysis is supported by findings by Uljarevic, Carrington and Leekam (2015) who found evidence for a mediating effect of IU in the relation between sensory sensitivity and anxiety in mothers of children with ASD, and more recently has been supported by findings with children with ASD (Neil, Olsson & Pellicano, 2016). RRB may function as an attempt to cope with sensory-linked anxiety but this evidence is preliminary, using limited methods of investigation. Together these research studies suggest that IU is likely to be a key factor in understanding the relationship between RRB and anxiety.

Much research investigating anxiety and RRB in young people with ASD to date has relied on parent report questionnaires. Yet the methodology used to study RRB in ASD is crucially important to research outcomes (Honey, Rodgers, & McConachie, 2012; Leekam, et al., 2011). We know from research with typically developing children that parent and child reports often differ when reporting the child's emotional functioning (Boughton & Lumley, 2011; Comer & Kendall, 2004; Krain & Kendall, 2000). Thus researchers have suggested a multi-informant consensus between both parent and child report as a useful strategy and indicative of best practice (Cantwell, Lewinsohn, Rohde, & Seeley, 1997; Silverman & Ollendick, 2005). Given that RRBs may also involve internal experiences, that are less observable by others it therefore also appears important that the same multi-informant approach is taken to collecting RRB data.

It is possible that the previous reliance by researchers on parent report is due to the unique communication needs and cognitive style of young people with ASD, casting some doubt on whether young people can accurately report their own emotional state and behaviours. There have also been mixed findings in terms of levels of agreement between

parent and self-report in ASD (Farrugia & Hudson, 2006; Hurtig et al., 2009; Jepsen, Gray, & Taffe, 2012; Russell & Sofronoff, 2005). However a recent meta-analysis by Stratis and Lecavalier (2015) investigating informant agreement on emotional and behaviour problems in youth with ASD concluded that informant agreement in children with ASD is generally comparable to that reported in typically developing children. Therefore there is no reason why the difficulties associated with the use of self-report in young people with ASD would be any different to those with typically developing self-report.

Despite concerns about the ability of young people with ASD to report their own emotional states, thoughts and behaviours, it has been shown that young people with ASD are capable of engaging with cognitive behavioural therapy (CBT), a therapy that involves such skills (Lickel, MacLean, Blakeley-Smith, & Hepburn, 2012), especially when more concrete, structured, tangible approaches are used such as the use of visual aids (Moree & Davis, 2010). Taking note of such modifications can help us design effective approaches to engage young people with ASD in research where we are enquiring about their thoughts, feelings and behaviour.

In summary, a number of questions remain about the nature of anxiety in young people with ASD and the relationship between anxiety, RRB and IU. Furthermore what we do know has relied on parent report and is yet to be explored from a self-report perspective.

In order to meet the research aims of helping to shed light on these gaps in knowledge the current study used a novel mixed method design that included self-report questionnaires as well as parent-report questionnaires in order to gain quantitative evidence of the relation between anxiety, IU and RRBs. We also conducted interviews with young people to gain detailed qualitative evidence of their experiences. Specifically the study aimed to replicate previous findings on the relation between RRB, anxiety and IOU demonstrated using parent report, and to establish whether similar findings are found using self-report. The study also

aimed to systematically describe the relationship between RRB and anxiety using semi-structured interviews exploring real life RRB examples with young people with ASD.

## Method

### *Participants*

Participants were thirteen young people aged 13-20 years with a confirmed diagnosis of ASD, without an intellectual disability, and their parents. Parents of a further six young people who did not wish to take part themselves, completed questionnaires and their parent report data was used in the analysis with the young person's consent, giving information from nineteen families in total.

### *Recruitment*

Families were recruited either via local mainstream and specialist schools, through social media or the Database of Children with Autism Spectrum Disorder Living in the North East (Dasl<sup>ne</sup>). Local schools which had agreed to be involved in recruitment were asked to identify eligible young people in their school and information packs were then given to these families who could then return consent forms to the researcher to indicate their interest in taking part. An advertisement about the research was placed on a local ASD support group on social media website Facebook. All families on the Dasl<sup>ne</sup> database have indicated an interest in taking part in research and all those eligible were invited to take part.

The young people's level of ability was determined through information held by Dasl<sup>ne</sup> and schooling information. ASD diagnosis was confirmed outside of the study. Young

people on the Dasl<sup>n</sup>e database are required to have received a clinical diagnosis in order to be on the database. Young people recruited through other routes were identified to have an ASD diagnosis by teachers or self-referral to the study. All participants also completed a screening measure of ASD symptoms as part of the study.

### *Questionnaire measures and scoring*

*Spence Children's Anxiety Scale (SCAS-P, SCAS-C) (Spence, 1998).* This measure assesses symptoms of anxiety, providing a total score with indicators of clinical caseness (37 for boys and 44 for girls) and six subscale scores for different types of anxiety e.g. separation anxiety. In this study, only the total score is used. This is calculated by totalling the 38 anxiety items, removing the positive filler items. The scale has previously been used in ASD research (Rodgers et al., 2012; Sofronoff et al., 2005) and has been shown to have acceptable reliability and validity for use with children with ASD (Wigham & McConachie, 2014).

*Intolerance of Uncertainty Scale - Child and Parent (Walker, 2009).* Walker's IUS-C and IUS-P are 12-item questionnaires adapted from the 12-item version of the Intolerance of Uncertainty Scale (Carleton, Norton, & Asmundson, 2007), a short form of the original 27-item Intolerance of Uncertainty Scale (Freeston, Rhéaume, Letarte, Dugas, & Ladouceur, 1994). The Walker (2009) scale requires participants to rate, on a five point Likert scale, the extent to which statements relating to emotional, cognitive and behavioural responses to uncertainty are representative of them or their child. Both the parent and child versions of these measures have been used in previous ASD research (Boulter et al., 2014; Wigham et al., 2014). A total score on this scale was used for analysis and is calculated by totalling all items.

*Repetitive Behaviour Questionnaire-2 (RBQ-2), (Leekam et al., 2007).* The RBQ-2 is a 20 item questionnaire developed using items from the Diagnostic Interview for Social and



Communication Disorders and the original Repetitive Behaviour Questionnaire. Parents rate behaviours for severity or frequency on a three or four point Likert scale. The RBQ-2 has been shown to have good internal consistency for a sample of children aged 2-17 years with ASD (Lidstone, Uljarevic et al., 2014). There are currently no self-report measures for examining RRB in children and young people with ASD. Therefore a recently developed self-report version of the RBQ-2 for adults was used, the RBQ-2A (Barrett et al., 2015) which includes no change to item content and just very minor changes to the wording of response choices to aid understanding. The self-report RBQ-2A has been effectively tested with adults with ASD and shown to have good psychometric properties (Barrett et al., 2015). To our knowledge this is the first time the measure has been used with young people with ASD.

*Parent report RBQ-2 scoring* (Leekam et al., 2007). An average total score is calculated for each participant by equalising all the sections into a 3-point Likert scale, adding together the responses for each item and dividing by the number of items. This produces an average score, indicating the child's level of repetitiveness on a scale of 1–3. In the same way, an average score for a 'Motor/Sensory Behaviours' (items 1-6, 8-10) and 'Rigidity/Routines/Preoccupation with Restricted Interests' (items 11, 13-19) subscales are calculated.

*Self-report RBQ-2A scoring* (Barrett et al., 2015). An average total score is calculated as described above. The RBQ-2A however has different subscales following Barrett et al's (2015) factor analysis study with adults. The 'Repetitive Motor Behaviour' subscale is calculated by equalising into a 3-point Likert score as above and totalling the scores on items 1-6, and then dividing by the number of items. The 'Insistence of Sameness' subscale is calculated in a similar manner with items 11-17 and 19.

*Social Responsiveness Scale 2 (Constantino, 2012).* Parents also completed the Social Responsiveness Scale 2 (SRS2) (Constantino, 2012). This is a sensitive and valid measure of social responsiveness, indicative of autistic traits in children age 4-18 years (Constantino et al., 2003). For participants over the age of 18 years, parents completed the relative report version of the adult SRS2. Total scores on the measure were calculated and used for this study.

### *Semi-structured interview*

Using a semi-structured interview script, detailed and systematic descriptions of real life examples of RRB were gained with the aim of providing triangulation and complementarity of the quantitative results. Triangulation refers to seeking convergence and corroboration of results from different methods studying the same phenomena. Complementarity refers to seeking elaboration, enhancement, illustration, clarification of results from one method with results from the other methods (Greene, Caracelli, and Graham, 1989).

The structure of the interview was designed to take into consideration the thinking style and communication difficulties of young people with ASD using a visual structure similar to a comic strip conversation to explore their report of what was happening just prior to the RRB commencing, during the RRB and afterwards. Comic strip conversations provide visual representations of the different levels of communication that take place in a conversation or situation. The visual structure, similar to a cartoon strip explores what happens in a given social situation including the thoughts and feelings of the people involved (Gray, 1998).

The interview had a mix of open-ended questions and ratings using visual analogue scales personalised to the young person and designed to elicit information about a specific

RRB example. Two 1-5 rating scales were used, 'not strong at all' to 'very strong', and 'not at all' to 'very much'. The young people gave their own labels to points 2, 3, and 4 on the scale. They also had a choice of a plain horizontal line scale, a scale with blocks of increasing height, a scale of very pale blue to dark blue and a scale from red through to green. These scales were used to rate intensity of emotion, how much the young person was enjoying what they were doing and how much they might have preferred to do something else. The interview began by getting a good behavioural description of the RRB, what was happening prior to the young person starting the RRB and what happened afterwards. This was then drawn in to the comic strip, with a box for each time point. Each box was then explored guided by questions enquiring about the young person's thoughts and feelings.

The semi-structured interview was developed drawing on current practices for working with young people with ASD and consultancy with professionals in the field. The interview was piloted with a typically developing thirteen year old.

### *Procedure*

Parent questionnaires were sent out to parents in the post. Completed questionnaires were either collected during a visit to the family home to meet with the young person or returned in the post. When the researcher met with the young people to complete their questionnaires, information about the project was recapped and consent to participate was confirmed verbally.

*Interviews.* Once the young person had completed the questionnaires, information about the interview stage of the research was discussed with the families who had indicated an interest. The interview was carried out according to the semi-structured script. As part of the interview, participants chose from a choice of four different visually presented analogue scales, and personalised the anchor points on their chosen scales. An example of a RRB from

the participants' responses on the RBQ-2A was chosen to be the subject of the interview. The choice was made jointly between the researcher and the participant. Criteria for selection included behaviours that were very frequent, a behaviour the person could remember a specific example of clearly and a behaviour that they felt comfortable to discuss. Participants were given the choice of whether the researcher drew in the pictures for the comic strip representation or they did, all but one chose for the researcher to draw. All data was collected, and transcribed by the first author.

A favourable ethical opinion was provided by NHS Research Ethics Committee, NRES Committee North East – Newcastle & North Tyneside 1.

### *Analysis*

*Design.* A single group mixed methods design was used including quantitative questionnaire measures and a semi-structured interview. Greene, Caracelli, and Graham (1989) identify five broad purposes or rationales for the use of mixed methods, of which triangulation and complementarity are applicable to this study.

*Questionnaire data.* Intraclass correlations were carried out to assess the level of agreement between parent and young person report. Pearson correlations were used to examine the relationships between constructs of interest.

*Interview data.* All interviews were transcribed verbatim. Data provided from rating scales were examined using descriptive statistics. Guidance in analysing the qualitative elements of the SSI using thematic analysis were taken from Braun and Clarke (2006). A theoretical analysis was carried out, coding for a specific question, driven by the researcher's theoretical interest in the interview topic. This is consistent with the aim of the semi-structured interview to provide triangulation and complementarity to the quantitative questionnaire data.

Coding of the data was a systematic iterative process, and data extracts could be assigned multiple codes. A code identifies a feature of the data that is interesting or relevant to the research questions and represents the data in its most basic form (Braun & Clarke, 2006). Each time a new code was applied, previously coded data was re-read for the new code. Initial codes were then reviewed and refined before themes were identified. Themes capture something important about the data and represent some level of patterned response or meaning (Braun & Clarke, 2006).

*Validity of qualitative analysis.* Validity of the results was considered in the completion of the analysis. A reflective journal was kept so that researcher bias could be considered and identified as well as allowing for ideas to be developed as part of the iterative process. Findings that would be expected from existing theory in the field of anxiety in ASD were not exclusively looked for and other ideas and perspectives were given equal attention.

Initial codings were discussed with two authors (JR and EH) and the final codings were given to an independent researcher with knowledge of ASD and RRB to group in to themes. These themes were then compared to the author's themes looking for areas of overlap and disagreement. Any disagreements were taken back to be discussed with JR and EH.

## Results

Nineteen families took part in the study. There were 13 parent and young person dyads and six families where only the parent took part. The mean age of the young person was 16.81 years (range: 13.19 – 20.77, SD 2.39) including those where only the parent took part. Sixteen of the 19 young people were male. Eighteen mothers took part and 1 father.

All parents completed the SRS-2 as a screening measure of social responsiveness, indicative of ASD symptoms. The mean score was 104.11 (range: 45-144). Two participants scored below the indicative cut off of 60, scoring 45 and 49. It was decided to keep these families in the study and analysis as both of these participants were recruited through Dasl<sup>n</sup>e. All young people on the Dasl<sup>n</sup>e database have received their ASD diagnosis through stringent clinical assessment using standardised ASD diagnostic measures such as the ADOS. This is a requirement to be on the database.

### *Questionnaire Data*

*Scale Reliability.* Cronbach's alpha coefficients were calculated for the child and parent report versions of the measures and their relevant subscales used in the study analyses. All measures showed acceptable levels of internal consistency ( $\alpha=0.75-0.96$ ), including the self-report RBQ-2A ( $\alpha=0.89$ ) used for the first time in this population. Internal consistency for the parent and child versions of the SCAS and IUS were very similar to those reported by Boulter et al. (2014). Internal consistency of the parent RBQ-2 was similar in terms of overall score and factors to Lidstone et al. (2014).

(Insert table one here)

Based on parent reported SCAS scores, seven young people were above the clinically indicative cut off for anxiety. Using young person self-report, six young people were above the cut-off. This is not a diagnostic measure but scoring above the cut-off indicates that there are likely to be significant difficulties with anxiety.

*Parent and Young Person Agreement.* Intraclass correlation coefficient (ICC) analyses were used to examine the consistency and absolute agreement between dyads ( $N=13$ ) on each

of the measures. For the total SCAS scores, findings were substantial for absolute agreement ( $ICC=0.66$ ; 0.17-0.88) and consistency ( $ICC=0.64$ ; 0.16-0.87).

Agreement between dyads ( $N=13$ ) was poor on the repetitive behaviour measures and IUS, compared to the SCAS. For the repetitive behaviour measures, findings were poor for absolute agreement ( $ICC=0.21$ ; -0.38-0.67) and consistency ( $ICC=0.20$ ; -0.37-0.66). Follow up analysis was carried out comparing only items that load similarly on to the two types of RRB on both the parent-report and self-report questionnaires. Results showed some improvement in absolute agreement ( $ICC=0.32$ ; -0.25-0.72) and consistency ( $ICC=0.31$ ; -0.26-0.73), however remained poor. Findings for the IUS were also poor on absolute agreement ( $ICC=0.12$ ; -0.45-0.61) and consistency ( $ICC=0.12$ ; -0.44-0.61). These findings suggest that parent and young person pairs were generally responding differently to the questionnaire items.

(Insert Table 2 here)

A positive association between parent reported levels of young person's RRB and anxiety was found, replicating previous findings. The same positive association was seen in young person self-reported RRB and anxiety for the first time.

Pearson correlation revealed a positive association between parent reported levels of young person's intolerance of uncertainty and anxiety, also replicated in self-report.

Parent reported levels of young person's intolerance of uncertainty and the different types of RRB were shown to be positively associated. Positive associations were also seen in self-report however did not reach significance.

The relationship between anxiety and the two different types of RRB were also examined for both parent and self-report. A significant positive association was seen between

parent reported anxiety and both types of RRB; RSM and IS. In self report a positive association was seen between anxiety and both types of RRB however these did not reach significance.

### *Interview Data*

Ten young people took part in an interview. One participant provided two examples of RRB, so in total 11 real life examples of RRB were described using a comic strip conversation format. A range of RRB were discussed as can be seen in Table 3.

(Insert Table 3 here)

As seen in Table 3 there was some disagreement between parent and self-report on the items chosen for the semi-structured interview (SSI). There are cases where the parent report suggests the parent was either unaware of the RRB or rated it as infrequent or mild however the young person reported the RRB as frequent or severe. Conversely, there are also cases where the parent is reporting a greater frequency and severity compared to the young person. This emphasises the importance of gaining both reports as using parent report alone some RRB experienced by the young person may not be reported.

*Thematic analysis results.* Refinement of 30 initial codes and initial themes, resulted in five final themes outlined below. A description of each of the themes can be seen in Table 4 and Figure 1 represents how the themes are linked.

(Insert Figure 1 here)

(Insert Table 4 here)



*Young person's insight into RRB.* This theme relates to the level of insight young people demonstrated within the interviews. Level of insight was mixed; some young people had clear ideas on the function of their behaviour while others were unsure why they engaged in a particular behaviour.

P08

**(Interviewer) So when you stopped pacing you felt calmer. What do you think helped you feel calmer?**

Well, it's the actual pacing, it gets rid of a lot of energy

**Yeh**

so um I'm a lot more, a little bit tired, that helps calm me down

**Yeh, so getting rid of some of the energy**

And also it helps me organise my thoughts so

**Uhhh**

So I'm a lot better organised mentally

Some young people actively sought clarification of whether something counted as an RRB or demonstrated an understanding of trying to distinguish between a RRB and any other type of behaviour. Other aspects of insight covered within this theme include the young person's sense that the RRB was a necessity, related to insight into function of the behaviour, and also difficulties or experiences of reporting thoughts and emotions in relation to the RRB were included in this theme. Many of the young people were able to report thoughts and feelings, however data extracts from four participants indicated some difficulties.

*Experience of RRB - Enjoyment.* Nine young people reported enjoying the RRB and scores of the associated feelings on the individualised visual analogues scales ranged from 2 to 5 in strength of feeling. However two of these participants also talked about not enjoying the behaviour or a wish to be doing something else instead. Both of these participants, when asked how much they were enjoying the RRB, reported the behaviour as being enjoyable as it was helping them in some way e.g. to manage anxiety but that they would have a strong preference to be doing something else.

*Experience of RRB - Attention.* The attention subtheme within the theme ‘experience of RRB’ relates to a number of codes identified in the data that speak to an attentional aspect of the experience of RRB. This includes two young people talking about the level of awareness they have of doing the RRB and also one young person talking about being distracted by the RRB from what they had intended to do. Two participants talked in some detail about a sense of ‘zoning out’. They both talk about what appears to be the RRB bringing about a ‘zoned out’ state or sense of non-reality. One of them also talked about the way they do the RRB behaviour to keep them from being too ‘zoned out’.

P09

(Young person) If I’m feeling quite like angry, or wound up you know, like it’s just sort of like a calming mechanism for that

**Uhhh**

like it just like, takes us in to like that alternate world where like you know nothing’s the matter and it’s just like you and what you’re doing

**Uhhh**

There’s like no-one else

**So...**

So it’s like a bubble

P08

(Young person) I’m not really, not exactly here if you know what I mean? The reason why I don’t put shoes on is because I need to feel the gravel to not just end up sitting there, standing there in a daydream because then I’ll come back in as hyperactive as before

Conversely to a sense of zoning out, four participants talked about the RRB providing them with focus, when they need to be paying attention.

The final aspect of attention identified within this theme is that of two young people getting ‘stuck’ in an attentional sense with a RRB, for example getting stuck listening to the same song over and over again.

*Motivating factors - Initiating RRB.* A number of factors were present in the data that can be understood as motivating factors for initiating RRB. This may not have been explicitly stated by the young person as a motivating factor but present in their description of their experience in a way that suggests it impacted on their choice to do the behaviour. The factors described were varied amongst participants and behaviours. A number of the motivating factors captured under initiating RRB appear to be somewhat related to anxiety or uncertainty. One participant described feeling hyperactive or restless, five participants talked about wanting to know more detail or interest in the details of something, RRB as a way to pass time were described by three participants, boredom was talked about by four participants. Other factors included perfection, with two participants suggesting a seeking out of perfection, one participant talked about a feeling of anger and agitation if his plans and routines are not kept the same suggesting a desire to not feel angry may be driving his restricted behaviours. Finally four participants talked about being alone or avoiding people in relation to a range of RRB, either using the RRB to be alone and avoid people as in the example below, or a preference to be alone when doing their RRB.

P16 (talking about liking the feel of different surfaces, particularly walls)

**(Interviewer)...this sounds like maybe something you do when you feel worried?**

Yeh I usually do it when I'm in like busy places like if I'm going to town and stuff ... I like to kind of go to the side and stuff, try and feel along the wall 'til where I'm going which is usually the game shop or something

**So keeping to the edges, kind of away from people?**

Yeh. It just helps us like, cool down, like figure out where I am and stuff like that.

**Yeh and this is a bit of a difficult question, but do you know why that helps you kind of calm down?**

Ermm I don't know actually, erm...

**Yeh**

But I've done it since I was small so I guess like, just like I guess knowing that there's not people all around us I guess

*Motivating factors - Ending RRB.* Motivating factors to end RRB were also noted in the interviews. Four participants talked about external influences on them stopping the behaviour. Two of them talked about a sense that to continue would be inappropriate, for example continuing to rock on a chair at school once a class has started.

Participants also talked about internal influences to stop their behaviours. These included losing interest, wanting to get back to what they were doing previously or to do something else. Two young people also talked about a sense that they had done something enough or completed it, or that it had been done right.

One participant who talked about using pacing to calm down, could say that he stopped pacing when he was calmer, suggesting internal emotional regulation as a potential motivator to stop the behaviour.

*Anxiety.* The theme of anxiety includes participants talking about anxiety in relation to their example of a RRB, participants talking about calmness and also participants specifically talking about how they cope or manage with anxiety.

Six of the ten participants talked about anxiety in their interviews while talking through their example RRB. Three of the examples were focusing specifically on a situation that the young person identified as making them feel particularly anxious including, being in a new class at college, worry over performance on a school test and anxiety related to a large group of people being present. These young people were able to relate their anxiety to their RRB, although to varying levels.

P07 (talking about rocking on a chair)

**(Interviewer) Can you remember how you felt, what feelings or emotions you had?**

Probably anxiety

**Yeh**

Because in the middle of the class

**Yeh**

Yeh

**So you were feeling anxious?**

yep

**How strong was that feeling of anxiety?**

Probably a strong

**So a number 4?**

3...3 to 4 I would say

P16

(Young Person) Ah yeh, when I get nervous I just fiddle with my fingers and stuff like that ermm...I'll flick my little finger and stuff like that or just kind of like rub them together and stuff, I only do that if I'm feeling nervous.

P08

**(Interviewer) Can you remember it clearly yesterday, how it happened, what was going on?**

Yes. I was erm quite stressed yesterday because of school work

**Ah right**

I'd had a history test erm just a bit stresses so I went for a bit of a pace to relieve that stress

Participants identified a range of ways of coping with anxiety including, 'just swallow it down', talking to someone about it, finding out more information, touching a cold wall to cool down, avoiding people, focusing on something else or distracting yourself, pacing, exercising, listening to music, a taught problem solving technique and fiddling with things. Some of these related to the RRB the young person had discussed as their example in the interview. In connection with this, four participants talked about calmness related to their RRB.

P09

**(Interviewer) Can you remember what you were thinking about when you ticked that one?**

Probably music, I like that's like something that like, it's a calming mechanism so it's like something I do every day

**Ah ok**

I use that as like, me specific thing

For some of the young people there appeared to be a strong relationship between anxiety and motivation for a RRB. This was verbalised in some interviews, in others it could be understood this way, bearing in mind the assumptions that could be made with researcher bias.

*Uncertainty.* Seven participants talked about an element of uncertainty in a situation or thinking about what may or may not happen in the future while describing their example RRB. This is illustrated in the example below.

P19

(Young person) Er...I don't know if there's a word that can describe someone who really thinks about what might happen in the future, like predicting maybe  
**So you felt that you were kind of thinking a lot about the future?**  
 Thinking about what could happen if I....if I either did this or if I did that and what the results might be of that

The data from this theme appears to relate closely to the construct of IU. Some examples of this in the interviews were seen as anxiety provoking and some were seen as a positive or exciting thing, for example one participant described feeling excited that he may find 'a really good song that would be the best one yet'. One participant talked about the need for routine, to stop uncertainty in the form of a surprise, suggesting a potential link between uncertainty and motivation for RRB.

In summary, thematic analysis of the interviews identified five main themes. These themes provide further useful information towards the aim of better understanding the relationship between anxiety and RRB in young people with ASD. For some young people there is a relationship between anxiety and motivation for engaging in RRB providing complementary data for the findings from the questionnaire data. There also appears to be a strong sense of enjoyment related to RRB. The behaviours and experiences the young people reported were varied and this must be considered when drawing conclusions. The interview data also supports the aim of gaining self-report data from young people with ASD as in

addition to the questionnaire data, the interviews demonstrate young people's insight and understanding of RRB.

*Triangulation of quantitative and qualitative data*

The qualitative data from the semi-structured interviews complements the quantitative data and allows for triangulation. A number of young people talked about anxiety in relation to their RRB, corroborating the correlation between RRB and anxiety seen in the questionnaires. The interview data provides information that can give some indication of the possible direction of this correlation. The data particularly within the themes 'Young person's insight into RRB' and 'Motivating factors' appears to suggest, in accordance with previous research, that RRB may act as a coping response to anxiety. However the interviews also demonstrate the variety in the young people's personal experiences of RRB, including motivators for RRB and influences on their behaviour, highlighting that anxiety and coping is only one part of a likely much more complicated explanation for RRB.

Interview data also provided support for the positive correlation between RRB and IU seen in the questionnaire data. Elements of uncertainty were present in a number of the situations that were discussed in the interviews. For a couple of young people there appeared to be a link between their RRB and aiming to reduce uncertainty for example maintaining a strict routine to limit surprises, or researching something entirely until there was nothing else to learn, potentially to limit uncertainty. Complementary to the correlations found between anxiety and IU, a number of the young people also described a level of uncertainty leading them to feel anxious.

## Discussion

This study aimed to shed further light on areas of the research field where questions still remain; the nature of the relationship between anxiety and RRB and whether young people's self-report would mirror earlier findings in parent report.

Levels of anxiety reported in this study are comparable to those seen in similar studies (Boulter et al., 2014; Wigham et al., 2014). The study has replicated previous findings, demonstrating a significant positive relationship between parent-reported RRB and anxiety in this sample of young people with ASD. This relationship has also been shown in young person self-report for the first time. This study replicated findings by Boulter et al. (2014), demonstrating a significant positive relationship between both parent and self-reported intolerance of uncertainty and anxiety in this sample of young people with ASD.

Findings from this study also replicated Wigham et al. (2014), demonstrating a significant positive relationship between parent reported levels of intolerance of uncertainty and RRB in young people with ASD. A positive association was also demonstrated in young person self-report, however this did not reach significance.

A significant relationship was found between parent reported levels of young person's anxiety and both types of RRB. This was also demonstrated in self-report however did not reach significance. This is different to some existing findings in the literature. Lidstone et al. (2014) and Rodgers, Glod, et al. (2012) did not find an association between anxiety and repetitive sensory motor behaviours, however the results from this study are in line with findings from Wigham et al. (2014) who found a relationship between anxiety and both types of RRB.

Findings from the interviews support the findings from the quantitative data, with an apparent relationship between anxiety and RRB in at least some of the young people's examples. There also appeared to be a relation between aspects of uncertainty and anxiety in some of the interviews. There were examples of both types of RRB being used as a coping



strategy for anxiety, which supports previous findings that anxiety may act as a motivator or trigger for RRB and that the behaviours are used as a coping strategy (Joosten et al., 2009; Rodgers, Glod, et al., 2012). Related to this, the idea that anxiety in ASD may be related to poor emotional regulation (White et al., 2014) is potentially supported by findings from the interviews where young people talked about RRB in terms of a way to feel calm and reduce anxious feelings.

Attention was also a theme identified in the interviews that may relate to function of RRB. Arousal has been identified as a potential intrinsic motivator for RRB by Joosten et al. (2009) and examples of RRB being used to both increase attention/arousal and decrease attention/arousal were described.

The findings from this study add to the growing body of work aiming to better understand the relationship between anxiety and RRB in young people with ASD. It has been able to show that previous parent-report findings are also seen with young person self-report, provide further evidence for the association between RRB and IU and has provided information about the potential relationships between anxiety and the different types of RRB, an area of the field where findings are currently mixed.

There are a number of limitations to the study. The RBQ-2A self-report measure used in this study has not been validated for this population and has only been used in undergraduate students and a small sample ( $N=29$ ) of adults with ASD (Barrett et al., 2015). Although the measure was shown to have acceptable reliability within the study, further research is needed to validate this measure in an ASD child/adolescent population. Similarly the SCAS, although regularly used in ASD research and shown to have acceptable psychometric properties in this population, has been developed with typically developing children. Given research is beginning to show that some young people with ASD may present with an atypical anxiety presentation (Kerns et al., 2014), measures developed for typically

developing children may not capture an accurate measure of anxiety in ASD. The IUS has also been developed in typically developed populations and so may not accurately capture the phenotype of IU in an ASD population.

The semi-structured interview was a new measure designed for the study. The less visible RRB such as insistence on sameness were harder to discuss within the comic strip conversation framework of the interview. Further clarity as to how to approach exploring these types of RRB should be addressed in any future revision of the semi-structured interview.

The study had a small sample size. As a result, all but one of the statistical procedures were underpowered, however low power did not appear to affect the ability to detect relationships present in the data and strong effect sizes were shown. With a larger sample and greater power, additional relationships may have been found and a greater number of the self-report relationships may have met significance. The small sample size also means that conclusions are limited in how much they can be generalised to other young people with ASD. Given a learning disability was an exclusion criteria for the study we also do not know how the findings would apply to young people with learning disabilities. Future research should aim to recruit larger sample sizes. This would allow for greater statistical power, allowing for conclusions to be made with more certainty and the potential for further more detailed analysis to be carried out, for example identifying groups of young people with high and low anxiety within samples for comparison.

Finally the small sample also consisted of young people from a wide age range. Participants will have had different levels of insight into their feelings and behaviour dependant on their developmental level. Although this makes for interesting information in a study using this methodology for the first time, future research may benefit from tighter inclusion criteria to be able to make more specific conclusions.

These limitations should be balanced against the strengths of the study. It is the first study to collect self-report information from young people with ASD about their RRB. This is important in two respects; firstly it provides evidence in support of existing findings that have previously only been evidenced through parent report and secondly it provides further information as to how young people can self-report, providing support for such practices in ASD research. The interview data has provided valuable information as to how young people can report their own behaviours with a level of insight. Insight was varied across the participants as would be expected but it is certainly clear that some of the young people were able to provide detailed information and understanding as to their behaviours which is encouraging for moving forward with self-report research.

A further strength is the successful use of mixed methods specifically the use of a SSI which has successfully engaged young people with ASD. This has allowed a novel self-report approach to be trialled and has provided data that elaborates on and corroborates quantitative data and existing theory.

With increasing evidence that IU is related to anxiety in young people with ASD, there is an increasing argument to include aspects of this construct in anxiety interventions for young people with ASD, as has been done in typically developing populations. Given that some initial findings also suggest a role for IU in understanding RRB, this could also be beneficial to include in interventions aiming to reduce RRB. Further research to understand how IU may relate to other aspects of ASD, such as social impairment would also be helpful.

The majority of young people who took part in an interview talked about some aspect of RRB being enjoyable, some explicitly described finding them helpful. This suggests that in clinical practice and learning environments, RRB should not automatically be seen as a negative aspect of the condition that we should aim to reduce but that the positive aspects of these behaviours are also explored.

*Conclusion*

This study replicated previous findings of a significant association between parent reported anxiety and RRB in young people with ASD. Importantly it provided first evidence for an association based on young person self-report of RRB and showed that young people are able to report these behaviours using a novel mixed methods design. Future investigation of anxiety in ASD from a self-report perspective would be encouraged.

## References

- American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders (5th ed.)*
- Barrett, S. L., Uljarević, M., Baker, E. K., Richdale, A. L., Jones, C. R., & Leekam, S. R. (2015). The Adult Repetitive Behaviours Questionnaire-2 (RBQ-2A): A self-report measure of restricted and repetitive behaviours. *Journal of autism and developmental disorders, 45*(11), 3680-3692.
- Bishop, S. L., Hus, V., Duncan, A., Huerta, M., Gotham, K., Pickles, A., . . . Lord, C. (2013). Subcategories of restricted and repetitive behaviors in children with autism spectrum disorders. *J Autism Dev Disord, 43*(6), 1287-1297. doi: 10.1007/s10803-012-1671-0
- Boughton, K. L., & Lumley, M. N. (2011). Parent Prediction of Child Mood and Emotional Resilience: The Role of Parental Responsiveness and Psychological Control. *Depression Research and Treatment, 2011*, 9. doi: 10.1155/2011/375398
- Boulter, C., Freeston, M., South, M., & Rodgers, J. (2014). Intolerance of uncertainty as a framework for understanding anxiety in children and adolescents with autism spectrum disorders. *J Autism Dev Disord, 44*(6), 1391-1402. doi: 10.1007/s10803-013-2001-x
- Braun, V., & Clarke, V. (2006). Using thematic analysis in psychology. *Qualitative Research in Psychology, 3*(2), 77-101. doi: 10.1191/1478088706qp063oa
- Cantwell, D. P., Lewinsohn, P. M., Rohde, P., & Seeley, J. R. (1997). Correspondence between adolescent report and parent report of psychiatric diagnostic data. *J Am Acad Child Adolesc Psychiatry, 36*(5), 610-619. doi: 10.1097/00004583-199705000-00011

- Carleton, R. N., Norton, M., & Asmundson, G. J. G. (2007). Fearing the unknown: A short version of the Intolerance of Uncertainty Scale. *Journal of Anxiety Disorders, 21*(1), 105-117. doi: 10.1016/j.janxdis.2006.03.014
- Comer, J. S., & Kendall, P. C. (2004). A symptom-level examination of parent-child agreement in the diagnosis of anxious youths. *J Am Acad Child Adolesc Psychiatry, 43*(7), 878-886. doi: 10.1097/01.chi.0000125092.35109.c5
- Constantino, J. (2012). The Social Responsiveness Scale Second Edition. *Los Angeles. Western Psychological Services.*
- Constantino, J., Davis, S., Todd, R., Schindler, M., Gross, M., Brophy, S., . . . Reich, W. (2003). Validation of a Brief Quantitative Measure of Autistic Traits: Comparison of the Social Responsiveness Scale with the Autism Diagnostic Interview-Revised. *Journal of Autism and Developmental Disorders, 33*(4), 427-433. doi: 10.1023/a:1025014929212
- Dugas, M. J., Gagnon, F., Ladouceur, R., & Freeston, M. H. (1998). Generalized anxiety disorder: a preliminary test of a conceptual model. *Behaviour Research and Therapy, 36*(2), 215-226. doi: 10.1016/s0005-7967(97)00070-3
- Farrugia, S., & Hudson, J. (2006). Anxiety in Adolescents With Asperger Syndrome: Negative Thoughts, Behavioral Problems, and Life Interference. *Focus on Autism and Other Developmental Disabilities, 21*(1), 25-35. doi: 10.1177/10883576060210010401
- Freeston, M. H., Rhéaume, J., Letarte, H., Dugas, M. J., & Ladouceur, R. (1994). Why do people worry? *Personality and Individual Differences, 17*(6), 791-802. doi: [http://dx.doi.org/10.1016/0191-8869\(94\)90048-5](http://dx.doi.org/10.1016/0191-8869(94)90048-5)

Gotham, K., Bishop, S. L., Hus, V., Huerta, M., Lund, S., Buja, A., . . . Lord, C. (2013).

Exploring the relationship between anxiety and insistence on sameness in autism spectrum disorders. *Autism Res*, 6(1), 33-41. doi: 10.1002/aur.1263

Gray, C. A. (1998). *Social stories and comic strip conversations with students with Asperger syndrome and high-functioning autism*: Springer.

Greene, J. C., Caracelli, V. J., & Graham, W. F. (1989). Toward a Conceptual Framework for Mixed-Method Evaluation Designs. *Educational Evaluation and Policy Analysis*, 11(3), 255-274. doi: 10.3102/01623737011003255

Hodgson, A. R., Freeston, M. H., Honey, E., & Rodgers, J. (2016). Facing the unknown: intolerance of uncertainty in children with autism spectrum disorder. *Journal of applied research in intellectual disabilities*.

Honey, E., Rodgers, J., & McConachie, H. (2012). Measurement of restricted and repetitive behaviour in children with autism spectrum disorder: Selecting a questionnaire or interview. *Research in Autism Spectrum Disorders*, 6(2), 757-776.

Hurtig, T., Kuusikko, S., Mattila, M.-L., Haapsamo, H., Ebeling, H., Jussila, K., . . . Moilanen, I. (2009). Multi-informant reports of psychiatric symptoms among high-functioning adolescents with Asperger syndrome or autism. *Autism*, 13(6), 583-598. doi: 10.1177/1362361309335719

Jepsen, M. I., Gray, K. M., & Taffe, J. R. (2012). Agreement in multi-informant assessment of behaviour and emotional problems and social functioning in adolescents with Autistic and Asperger's Disorder. *Research in Autism Spectrum Disorders*, 6(3), 1091-1098. doi: <http://dx.doi.org/10.1016/j.rasd.2012.02.008>

Joosten, A. V., Bundy, A. C., & Einfeld, S. L. (2009). Intrinsic and Extrinsic Motivation for Stereotypic and Repetitive Behavior. *Journal of Autism and Developmental Disorders*, 39(3), 521-531. doi: 10.1007/s10803-008-0654-7

- Kerns, C., Kendall, P., Berry, L., Souders, M., Franklin, M., Schultz, R., . . . Herrington, J. (2014). Traditional and Atypical Presentations of Anxiety in Youth with Autism Spectrum Disorder. *Journal of Autism and Developmental Disorders*, 44(11), 2851-2861. doi: 10.1007/s10803-014-2141-7
- Krain, A. L., & Kendall, P. C. (2000). The role of parental emotional distress in parent report of child anxiety. *J Clin Child Psychol*, 29(3), 328-335. doi: 10.1207/s15374424jccp2903\_4
- Leekam, S., Prior, M. R., & Uljarevic, M. (2011). Restricted and Repetitive Behaviors in Autism Spectrum Disorders: A Review of Research in the Last Decade. *Psychological Bulletin*, 137(4), 562-593. doi: 10.1037/a0023341
- Leekam, S., Tandos, J., McConachie, H., Meins, E., Parkinson, K., Wright, C., . . . Le Couteur, A. (2007). Repetitive behaviours in typically developing 2-year-olds. *J Child Psychol Psychiatry*, 48(11), 1131-1138. doi: 10.1111/j.1469-7610.2007.01778.x
- Lickel, A., MacLean, W. E., Jr., Blakeley-Smith, A., & Hepburn, S. (2012). Assessment of the prerequisite skills for cognitive behavioral therapy in children with and without autism spectrum disorders. *J Autism Dev Disord*, 42(6), 992-1000. doi: 10.1007/s10803-011-1330-x
- Lidstone, J., Uljarević, M., Sullivan, J., Rodgers, J., McConachie, H., Freeston, M., . . . Leekam, S. (2014). Relations among restricted and repetitive behaviors, anxiety and sensory features in children with autism spectrum disorders. *Research in Autism Spectrum Disorders*, 8(2), 82-92. doi: <http://dx.doi.org/10.1016/j.rasd.2013.10.001>
- Moree, B. N., & Davis, T. E., III. (2010). Cognitive-behavioral therapy for anxiety in children diagnosed with autism spectrum disorders: Modification trends. *Research in Autism Spectrum Disorders*, 4(3), 346-354.



- Neil, L., Olsson, N. C., & Pellicano, E. (2016). The Relationship Between Intolerance of Uncertainty, Sensory Sensitivities, and Anxiety in Autistic and Typically Developing Children. *Journal of autism and developmental disorders*, 1-12.
- Rodgers, J., Glod, M., Connolly, B., & McConachie, H. (2012). The Relationship Between Anxiety and Repetitive Behaviours in Autism Spectrum Disorder. *Journal of Autism and Developmental Disorders*, 42(11), 2404-2409. doi: 10.1007/s10803-012-1531-y
- Rodgers, J., Riby, D. M., Janes, E., Connolly, B., & McConachie, H. (2012). Anxiety and Repetitive Behaviours in Autism Spectrum Disorders and Williams Syndrome: A Cross-Syndrome Comparison. *Journal of Autism and Developmental Disorders*, 42(2), 175-180. doi: 10.1007/s10803-011-1225-x
- Russell, E., & Sofronoff, K. (2005). Anxiety and social worries in children with Asperger syndrome. *Aust N Z J Psychiatry*, 39(7), 633-638. doi: 10.1111/j.1440-1614.2005.01637.x
- Silverman, W. K., & Ollendick, T. H. (2005). Evidence-based assessment of anxiety and its disorders in children and adolescents. *J Clin Child Adolesc Psychol*, 34(3), 380-411. doi: 10.1207/s15374424jccp3403\_2
- Sofronoff, K., Attwood, T., & Hinton, S. (2005). A randomised controlled trial of a CBT intervention for anxiety in children with Asperger syndrome. *Journal of Child Psychology and Psychiatry*, 46(11), 1152-1160. doi: 10.1111/j.1469-7610.2005.00411.x
- Spence, S. H. (1998). A measure of anxiety symptoms among children. *Behaviour Research and Therapy*, 36(5), 545-566. doi: 10.1016/s0005-7967(98)00034-5
- Spiker, M. A., Lin, C. E., Van Dyke, M., & Wood, J. J. (2012). Restricted interests and anxiety in children with autism. *Autism*, 16(3), 306-320. doi: 10.1177/1362361311401763

- Stratis, E. A., & Lecavalier, L. (2015). Informant Agreement for Youth with Autism Spectrum Disorder or Intellectual Disability: A Meta-analysis. *J Autism Dev Disord*, 45(4), 1026-1041. doi: 10.1007/s10803-014-2258-8
- Uljarević, M., Carrington, S., & Leekam, S. (2016). Brief Report: Effects of sensory sensitivity and intolerance of uncertainty on anxiety in mothers of children with Autism Spectrum Disorder. *Journal of autism and developmental disorders*, 46(1), 315-319.
- Walker, S. (2009). What do we know about the relationship between intolerance of uncertainty and worry in young children? *Clinical Psychology Doctorate Unpublished Thesis. Newcastle University, Newcastle upon Tyne.*
- White, S. W., Mazefsky, C. A., Dichter, G. S., Chiu, P. H., Richey, J. A., & Ollendick, T. H. (2014). Social-cognitive, physiological, and neural mechanisms underlying emotion regulation impairments: understanding anxiety in autism spectrum disorder. *International Journal of Developmental Neuroscience*, 39(0), 22-36. doi: <http://dx.doi.org/10.1016/j.ijdevneu.2014.05.012>
- Wigham, S., Rodgers, J., South, M., McConachie, H., & Freeston, M. (2014). The Interplay Between Sensory Processing Abnormalities, Intolerance of Uncertainty, Anxiety and Restricted and Repetitive Behaviours in Autism Spectrum Disorder. *J Autism Dev Disord*. doi: 10.1007/s10803-014-2248-x
- Wood, J. J., & Gadow, K. D. (2010). Exploring the Nature and Function of Anxiety in Youth with Autism Spectrum Disorders. *Clinical Psychology-Science and Practice*, 17(4), 281-292. doi: 10.1111/j.1468-2850.2010.01220.x