

Running head: DEVELOPMENT OF RESTRICTED AND REPETITIVE BEHAVIORS

**Development of Restricted and Repetitive Behaviors from 15 to 77 Months: Stability of
Two Distinct Subtypes?**

Mirko Uljarević,^{1,2} Bronia Arnott³, Sarah J. Carrington^{1,4}, Elizabeth Meins⁵, Charles
Fernyhough⁶, Helen McConachie³, Ann Le Couteur³ and Susan R. Leekam¹

¹Wales Autism Research Centre, School of Psychology, Cardiff University

²Olga Tennison Autism Research Centre, School of Psychological Science, La Trobe
University, Melbourne, Australia

³Institute of Health and Society, Newcastle University

⁴School of Life and Health Sciences, Department of Psychology, Aston University

⁵Department of Psychology, University of York

⁶Department of Psychology, Durham University

Conflict of interest statement: None declared.

Abstract

A community sample of 192 parents reported on their children's restricted and repetitive behaviors (RRBs) at mean ages 15 months (N=138), 26 months (N=191), and 77 months (N=125) using the Repetitive Behavior Questionnaire-2 (RBQ-2). Consistent with previous factor analytic research, two factors were found at each age: one comprising repetitive sensory and motor behaviors (RSM), and the other comprising insistence on sameness behaviors including rigidity, routines, and restricted interests (IS). Regression analyses indicated that RSM and IS subtypes develop independently. RSM at 77 months was predicted only by RSM behaviors at 26 months and not by IS behaviors at either 15 or 26 months nor by RSM behaviors at 15 months. IS at 77 months was predicted by IS behaviors at both 15 and 26 months, but not by RSM behaviors at either 15 or 26 months. Our findings provide evidence that there is stability of two independent subtypes of RRBs, RSM and IS, across early childhood and that these subtypes develop independently of each other.

Key terms: Restricted and repetitive behaviors, longitudinal, repetitive sensory and motor behaviors, insistence on sameness behaviors, typical development

Introduction

Restricted and repetitive behaviors (RRBs) form a class of behaviors that includes repetitive motor movements, sensory reactions, rituals, routines, and restricted interests. RRBs are common during early typical development (Arnott et al., 2010; Evans et al., 1997; Leekam et al., 2007) and appear to have an adaptive function for nervous system maturation (Sprague & Newell, 1996), emotion and arousal regulation (Gesell et al., 1974; Evans et al., 2005; Werner, 1948), and acquisition of motor skills (Wolff, 1968; Thelen, 1979). RRBs are also found in developmental and psychiatric disorders, such as Autism Spectrum Disorder (ASD), Obsessive Compulsive Disorder, Prader-Willi syndrome, and intellectual disabilities (Langen, Durston, Kas, Van Engeland, & Staal, 2011). In ASD, RRBs form an essential domain for a diagnosis (American Psychiatric Association, 2013) and these behaviors are among the earliest infant predictors of a later ASD diagnosis (Ozonoff et al., 2008; Wolff et al., 2014). Furthermore, the presence of certain types of challenging RRBs in clinical disorders, particularly in ASD, can have negative impact on the child's learning and social adaptation and the family's stress management (Harrop, McBee, & Boyd, 2016; Hodgson, Grahame, Garland, Gaultier, Lecouturier, & Le Couteur, 2016; Grahame et al., 2015; South, Ozonoff, & McMahon, 2005). Therefore, charting stability and change of these behaviors in typical development is an important priority for understanding the emergence of potentially clinically significant RRBs.

Research studies using factor analysis propose two main subtypes of RRBs (see Leekam, Prior, & Uljarevic, 2011 for a review). One subtype, repetitive sensory and motor (RSM) behaviors, includes repetitive motor behaviors and unusual sensory responses such as simple motor stereotypies and excessive smelling or touching of objects. The other, insistence on sameness (IS), includes routines, rigid behaviors, and restricted interests. Support for this distinction has been found in cross-sectional studies of both typically developing children

(Cevikaslan, Evans, Dedeoglu, Kalaca, & Yazgan, 2014; Evans et al., 1997; Leekam, et al., 2007) and children with diagnoses of ASD (Bishop et al., 2013; Georgiades, Papageorgiou, & Anagnostou, 2010; Honey, McConachie, Turner, & Rodgers, 2012; Lidstone et al., 2014).

Preliminary research with typically developing individuals and with individuals who have clinical conditions suggests that the two main RRB subtypes are linked to separate brain regions or networks (Langen et al., 2011). Furthermore, research in ASD samples suggests that the RSM and IS subtypes might be genetically independent. For example, studies by Silverman et al. (2002), Szatmari et al. (2006), Lam, Bodfish, and Piven (2008) and Uljarević, Evans, Alvares, and Whitehouse (2016). found evidence for familiarity for the IS type of RRBs, but not for the RSM behavior subtype. Furthermore, Cannon et al. (2010) found that IS and RSM subtypes were linked to largely non-overlapping chromosomal regions (2q37.1-q37.3 for IS, and 15q13.1-q14 for RSM).

Despite emerging biological evidence for separate RRB subtypes, we have little behavioral evidence of how RSM and IS are related across time in typical development. Some repetitive motor behaviors are considered to be developmentally immature forms of behavior, emerging in typical development in the first year of life, then declining with the onset of neuromuscular and adaptive cognitive control (Thelen, 1979, 1981). Cross-sectional research in typical development indicates that these RSM behaviors peak in the first 12 to 15 months of life, with a lower frequency in children of 24 months and older. IS behaviors, on the other hand, start increasing from 24 months, reaching their peak around 48 months of age, and then slowly declining to typically low levels by 6 years of age (Cevikaslan et al., 2014; Evans et al., 1997).

To date, no study has charted the longitudinal progression of the two reported RRBs subtypes in a typical community sample from infancy through early childhood. However, two longitudinal studies have explored trajectories of RRBs in children with ASD and comparison

samples whilst one has focused exclusively on children with ASD. Harrop, McConachie, Emsley, Leadbitter, and Green (2013) used an observational measure of RSM in a group of 49 children with ASD (mean age 45 months) and 44 typically developing (TD) children (mean age 24 months). Observations taken of RSM behaviors at three time points across a 13-month period showed consistently higher scores in children with ASD, and no significant change across time points for either group. In a recent study, Wolff et al. (2014) compared reported repetitive behaviors measured using the Repetitive Behavior Scale-Revised (RBS-R; Bodfish, Symons, Parker, & Lewis, 2000) in 190 infants who had an older sibling with ASD and 60 infants who did not. In this study infants subsequently diagnosed with ASD were found to have elevated rates of both RSM and IS as early as the 12-month time point. Finally in a longitudinal study of RRBs in an ASD sample, Richler et al. (2010) used the ADI-R parental interview to track RRBs in children with ASD with varying ability levels at the ages of 2, 3, 5, and 9 years. RSM behaviors remained high across these four age points, decreasing by age 9 only in children who had higher non-verbal IQ at age 2. In contrast, IS behaviors started at a low level at age 2 and moderately increased in severity; these behaviors were not predicted by non-verbal IQ and the developmental relation between RSM behaviors and IS behaviors was not explored. Although the children in this study had a wide range of ability levels and general developmental delay, they all had diagnoses of ASD.

The present study investigated the reported incidence of the two subtypes of RRBs in a community sample at three age points: 15 months, 26 months, and 77 months. The first aim was to chart the frequency of RRBs in the same children at different ages, thereby adding to existing evidence on change in RRBs in typical development. The second aim was to examine for the first time whether subtype scores at younger ages predict scores on the same and different subtypes at later ages. As IS behaviors tend to increase in frequency just as RSM behaviors start to decrease, it is important to clarify whether RSM behaviors are an

earlier developmental form that predates IS behaviors, or whether RSM and IS behaviors are different classes of behaviors that develop independently. Clarifying whether one behavioral subtype has developmental significance for the other, may have clinical significance for intervention planning. It is known that some behaviors such as insistence on rigid preoccupations and extreme avoidant behaviors can be problematic for children with ASD and their families, however other types of RRBs (e.g., hand/finger movements) may have a positive function for self-soothing or attention-focusing (Leekam et al., 2011; Grahame et al., 2015). Evidence that IS and RSM behaviors develop independently of each other would support the notion that interventions should be designed to target the function of specific RRBs only, and delivered at a developmentally appropriate time point for the child.

We used factor analysis to examine whether RSM and IS are independent sub-types of behavior at the three different ages. We then used regression analysis to examine if these subtypes co-develop or develop independently. If they develop independently, RSM and IS subtypes at older ages should be predicted only by the same subtype at younger ages.

The third aim was to investigate whether presence of RRB subtypes at 77 months would be predicted by earlier RRB subtype scores even when other factors such as gender, language, and socio-economic status (SES) were taken into account. Findings on the influence of gender on the levels of repetitive behaviors in both typical development and in individuals with ASD have been mixed. Some studies of typical development in the first two years reported no difference in rates of RRBs (Evans et al., 1997; Glenn Cunningham & Nananidou, 2012; Zohar & Felz, 2011), while other studies reported that boys have higher total RRB scores (Leekam et al., 2007), sensory interests (Arnott et al., 2010), and rigid/insistence on sameness like behaviors (Cevikaslan et al., 2014). Similarly, in an ASD sample, one study using both observational assessment (Autism Diagnostic Observation Schedule (ADOS; Lord et al., 2000) and parent interview (Developmental, Dimensional, and

Diagnostic Interview; Skuse et al., 2004), found that boys (aged 3- to 18-years) had greater levels of repetitive play with objects and circumscribed interests than did girls (Mandy et al., 2012). By contrast, Joseph, Thurm, Farmer, and Shumway (2013) found no influence of gender on any of the Repetitive Behavior Scale-Revised parent questionnaire subscales in a sample of young children with ASD. However, all these studies have been cross-sectional, and included a range of age groups.

Given these mixed findings, we first examined gender differences for each RRB subtype and then took account of associations with language skills and SES. Previous research on the influence of language on RRBs has found a negative relation between language levels and RSM in typically developing children at 26 months and 4 years (Larkin, Meins, Centifanti, Fernyhough, & Leekam, 2016; Harrop et al., 2014) and in children with ASD aged 3 years (Ray-Subramanian & Weismer, 2012; Harrop et al., 2014). Furthermore, in ASD samples, evidence shows that gains in language from age 2 to 3 years, may predict reduction in total RRBs (Paul et al., 2008; Ray-Subramanian & Weismer, 2012). We were interested to establish, in a community sample, whether early RRBs would predict RRBs at a later age beyond the effect of language ability and rate of language development. We also included SES as a control variable given well-established evidence in general population samples of language differences related to SES (Fernald, Marchman & Weisleder, 2013), and greater levels of parent-reported child behavior problems in lower SES groups (Patterson, Mockford, Barlow, Pyper, & Stewart-Brown, 2002).

Method

Participants

A community sample of 206 mothers and children (108 girls) was first recruited from health visitors' clinics and community organizations in the North East of England when infants were 8 months (references omitted for blind review). Inclusion criteria were that the

1 infant was full-term with no diagnosed medical conditions or developmental delay. The
2 current study focused on data on RRBs collected at 15 months and 26 months and in a follow
3 up postal questionnaire study when children were 77 months. The Repetitive Behaviour
4 Questionnaire-2 (RBQ-2) was introduced into the data collection mid-way through the 15-
5 month phase, which resulted in fewer individuals at 15 months having data on repetitive
6 behaviors than at 26 months.

7 The sample reflected the ethnic and socioeconomic background of the local and
8 regional area. The majority of the children (97%) were White and British, and the sample
9 represented the full range of SES from deprived to affluent based on Hollingshead scores
10 (Hollingshead, 1975). Hollingshead scores ranged from 11 to 66, with 44 % of families being
11 classed as low SES (parents with no post-16 education and no/menial/manual employment).
12 Ethical approval was granted from Local Health Service Ethics committees and University
13 Ethics Committees, and parents provided written consent for their child to participate in the
14 study. All research procedures were in line with guidelines from the American Psychological
15 Association and the British Psychological Society.

16 Children's ages and sample sizes at each phase were as follows: 15 months, $N = 138$
17 ($M = 14.9$, $SD = 0.59$, range 13-17 months); 26 months, $N = 191$ ($M = 26.0$, $SD = 0.85$, range
18 24-28 months); and 77 months, $N=125$ ($M = 77.4$, $SD = 1.84$, range 73-83 months). These
19 samples were used to run factor analysis and explore frequencies of items at each time point.

20 At 77 months, six children had diagnosed medical conditions. These were Ectodermal
21 Dysplasia ($N=1$), Asthma ($N=2$), hearing problems ($N=1$), heart valve defect ($N=1$), and
22 atopic dermatitis ($N=2$). Five children were reported to have identified Special Educational
23 Needs (SEN) including one child in the sample diagnosed with ASD, with clinical description
24 of Asperger syndrome. There were no differences in RRBs at any time point in children with
25 SEN compared with the rest of the group. Furthermore, when all analyses were re-run with

these five children excluded, results of all the analyses were similar. The child who received the ASD diagnosis was excluded from subsequent analyses leaving 87 children who had RBQ-2 data across 3 time points and this sample was used to examine the association between earlier (15 and 26 months) and later (77 months) RRB subtypes.

Materials and Methods

Procedure

At all three time points, the RBQ-2 was sent to parents by post. All respondents were mothers. At 15 and 26 months, mothers were asked to bring the completed questionnaire with them when visiting the University for a testing session during which the Preschool Language Scales-Third Edition (PLS; Boucher & Lewis, 1997; Zimmerman, Steiner & Pond, 1992) was administered to their child as part of a longer battery of assessments and the CDI was completed and returned. At 77 months, mothers were asked to return the RBQ-2 questionnaire by prepaid envelope.

Repetitive Behavior Questionnaire (RBQ-2)

Mothers completed the RBQ-2 (Leekam et al., 2007), a 20-item parental questionnaire designed to assess repetitive behaviors which occur in both children with autism and in typical development, originally developed from the Diagnostic Interview for Social and Communication Disorders (DISCO: Wing, Leekam, Libby, Gould, & Larcombe, 2002) and the Repetitive Behaviours Interview (RBI: Turner, 1995). For further details on the development of the RBQ-2, see Leekam et al. (2007). Items 1-19 only have been used in previous research and are reported here. Items include behaviors such as motor stereotypies (“Make repetitive hand and/or finger movements, e.g. flap, wave, or flick, his/her hands or fingers repetitively”), sensory behaviors (“Have a special interest in the smell of people or objects”), rigid behaviors and insistence on sameness (“Insist that aspects of daily routine must remain the same”). Please see Table 1 for the full list of items.

The RBQ-2 was shown to have good psychometric properties when used in the same sample at 15 months and 26 months (Arnott et al., 2010; Leekam et al., 2007), and also in a sample of children and adolescents with ASD aged from 2 to 17 years (Lidstone, Uljarević et al., 2014). Scoring and testing of internal consistency at 77 months followed the procedures reported in these publications, which included using a three-point scale of severity/frequency and mean RBQ-2 scores based on the range of 1 (never or rarely) to 3 (marked or notable). Mean RBQ-2 subscale scores were used for all analyses. Scores were calculated based only on the factor sets resulting from the factor analyses reported below.

Language

Children's expressive and receptive language was assessed at 15 months and 26 months with the PLS (Boucher & Lewis, 1997; Zimmerman, Steiner & Pond, 1992) and the MacArthur-Bates Communication Development Inventory (CDI; Fenson et al., 2007). The PLS is a play-based assessment of language skills from birth through age 7. The CDI is a standardized parent questionnaire that records children's language and communication skills. Age-appropriate forms of this measure were completed. The CDI: Words and Gestures was completed when the child was 15 months, and the CDI: Words and Sentences was used when the child was 26 months. PLS standardized scores and CDI average percentile scores for children's total language abilities were used in the analyses (Mean PLS at 15 months = 94.0, SD = 11.01, Mean PLS at 26 months = 98.03, SD = 16.14; Mean CDI at 15 months = 43.02, SD = 20.13, Mean CDI at 26 months = 46.0, SD = 26.0).

Analysis Plan

Initial descriptive analysis of RRB item frequencies and missing data was conducted. Sample sizes for this and factor analysis were N = 138 at 15, N = 191 at 26, and N = 125 at 77 months. Factor analysis was then carried out. Categorical Principal Component Analysis with varimax rotation using Kaiser normalisation was chosen due to the nature of the data. It was

first run with varimax and then re-run with oblique rotation (direct oblimin). Both the factor solution and the individual loadings were almost indistinguishable. Results using varimax rotation are presented. An initial analysis was run for each of the data sets at each time point to obtain eigenvalues. It has been suggested, however, that simply focusing on eigen values to determine the number of factors for retention, has a tendency to retain more factors than is appropriate (Hayton et al., 2004). The other commonly used rule in guiding the decision is the Scree Plot; however, this method has been criticized for the lack of robustness and reproducibility. Therefore we used Horn's parallel analysis (Horn, 1965) which is based on the Monte Carlo simulation of random production of Eigen values to determine the number of components that should be extracted. Therefore we have run parallel analysis using the SAS-based code developed by O'Connor (O'Connor, 2000) to determine the number of factors that should be extracted.

In order to address the first aim—charting the frequency of RRBs in the same children at different ages—we compared frequency of mean IS and RSM behaviors across three time points using ANOVA. As noted in the Introduction, findings on the influence of gender on the expression of RRBs in typical development have been mixed, with some studies finding no gender effect (Evans et al., 1997; Zohar & Felz, 2011; Glenn Cunningham & Nananidou, 2012) and other studies reporting higher levels of RRBs in males (Arnott et al., 2010; Cevikaslan et al., 2014; Leekam et al., 2007). Gender differences for each subtype were first explored using t tests. Separate hierarchical regression analyses were then used to examine whether RSM and IS behaviors tend to develop independently. This question was addressed through regression models exploring the contribution of same and different subtype behaviors at earlier ages on 77-month RRBs, with the effects of language and SES taken into account. Given previous findings showing that changes in language and changes in RRBs tend to interact during development in both TD and ASD populations, we also conducted additional

hierarchical regression models specifically to examine how change in language, RSM, and IS scores predicted RRB subtypes at 77 months.

Results

Initial analysis of missing RBQ-2 data showed that no more than 6.8% were missing for any questionnaire item, and there were no systematic differences in missing items within or between the three ages; all data were therefore retained for this analysis. Details of the pattern of endorsement across all 19 items are available in Appendix A (see Supplementary Material). In summary, apart from item 18 “Wearing the same clothes or refusing to wear new clothes” at 15 months, no other items had a frequency of less than 10% in the ‘occasional’ to ‘marked’ response range at any of the three time points, but the percentage of endorsement reduced with age. The number of repetitive behaviors in the occasional or marked range that was endorsed by at least 50% of parents reduced from 11/19 at 15 months to 7/19 at 26 months, and 2/19 at 77 months.

Are Distinct Subtypes Found at Each Age?

Three successive Categorical Principal Component Analyses were run (15, 26, and 77 months). Initial screening showed that, at 15, 26, and 77 months, assumptions of non-multicollinearity, sampling adequacy, and factorability were all met. The Kaiser-Meyer-Olkin (KMO) measure verified the sampling adequacy for the analysis for all three time points: KMO = .83 at 15 months, .88 at 26 months, and .80 at 77 months. Bartlett’s test of sphericity indicated that correlations were sufficiently large for PCA ($\chi^2 = 685.592, p = .000$ at 15 months, $3051.177, p = .000$ at 26 months, and $838.82, p = .000$ at 77 months).

At all three time points, five components had eigenvalues over Kaiser’s criterion of 1 and in combination explained 58.04% of the variance at 15 months, 57.08% at 26 months, and 60.03% at 77 months. At all three ages, factors 3 onwards had eigen values less than those indicated by parallel analysis (for Factor 3 at 15 months: 1.32 vs. 1.48, at 26 months:

1.26 vs. 1.39, and at 77 months: 1.41 vs. 1.48) which indicated that two-factor solutions should be retained in the final analysis. The Categorical PCA with varimax rotation was run specifying a two-factor solution at 15, 26, and 77 months. In combination, these two factors accounted for 39.65% of the total variance at 15 months, 38.99% at 26 months, and 43.45% at 77 months. Factor loadings for items were set at .40 (Stevens, 1992). Items that showed cross-loading onto more than one factor were not included. Resulting factors were interpreted as Repetitive Sensory-Motor (RSM) and Insistence on Sameness (IS). The composition of RSM and IS factors at each age, together with the detail on individual item loadings, is shown in Table 1. The RSM factor consisted of 8 items at 15 and 26 months, and 9 items at 77 months. It accounted for 29.34% of the total variance at 15, 27.83% at 26, and 28.46% at 77 months. The IS factor consisted of 6 items at 15 months, and 8 items at 26 and 77 months; it accounted for 10.31% of the total variance at 15, 11.17% at 26, and 14.99% at 77 months.

Cronbach's alpha was used to explore the internal consistency of the two factors (RSM factor: $\alpha = .79, .82, \text{ and } .85$ at 15, 26 and 77 months respectively; IS factor: $\alpha = .74, .77 \text{ and } .78$ at 15, 26 and 77 months respectively) suggesting (DeVellis, 2012) acceptable to good reliability.

RSM and IS levels at different ages

Table 2 summarizes the mean RBQ-2 subscale scores for participants who completed RBQ-2 questionnaires at all three age points ($n = 87$). For these participants, less than 1% of RBQ-2 data were missing (0.9% at 15 months, 0.4% at 26 months, and 0% at 77 months), and none of the participants had more than one data point (or RBQ-2 item) missing. A series of ANOVAs was performed in order to examine whether there were differences in (a) levels of RSM behaviors at 15, 26, and 77 months; and (b) levels of IS behaviors at 15, 26, and 77 months. Significance levels for multiple comparisons were adjusted to .01 to avoid Type 1 error.

There was a main effect of age for both mean RSM scores, $F = 54.74$, $p < .001$, partial eta-squared = .39 and for mean IS scores, $F = 12.21$, $p < .001$, partial eta-squared = .12. For RSM scores, post hoc tests using the Bonferroni correction showed that the mean RSM score at 15 months was higher than at both other ages (26 months: $p < .001$, Cohen's $d = .67$; 77 months, $p < .001$, Cohen's $d = 1.13$), and the mean RSM score at 26 months was higher than at 77 months ($p = .01$, Cohen's $d = .46$). For IS scores, post hoc tests showed that IS behavior scores at 26 months were higher than at 15 and 77 months (15 months: $p < .001$, Cohen's $d = .50$; 77 months: $p < .001$, Cohen's $d = .44$). See Table 2 for descriptive statistics.

Independent samples t tests were used to explore gender differences in IS and RSM scores at 15, 26, and 77 months. Although boys had consistently higher scores across all 3 time points, differences reached significance only for RSM at 77 months $t = 3.05$, $p = .002$, Cohen's $d = .68$.

What are the predictors of RSM and IS behaviors at age 77 months?

Before conducting regression models we looked for evidence of multi-collinearity. The highest correlation was between RSM behaviors at 15 and 26 months, $r = .58$, $p < .001$, and between RSM at 26 and 77 months, $r = .52$, $p < .001$; all other correlations had r s of .38 or lower, significantly lower than the .7 suggested as the threshold by Tabachnick and Fidel (2014). Furthermore, the lowest value for the collinearity diagnostics index in the regression model predicting RSM behaviors was .56 and in the one predicting IS was .50, also suggesting that multi-collinearity is not a concern. The relations among RSM and IS behaviors, and other variables of interest are presented in Table 3.

Using hierarchical regression, we first examined the predictors of RSM at 77 months. A summary of the full model is presented in Table 4. Analyses were first run with PLS scores and then re-run with CDI scores substituted¹. In the first step, SES and PLS scores accounted

¹The pattern of results was identical as when run with PLS.

for 4.9% of variance, $F = 1.44$, $p = .24$. In the second step, RSM and IS scores at 15 and 26 months accounted for an additional 24.7% of variance. The total model accounted for 29.5% of variance, $F = 4.79$, $p < .001$, with RSM behaviors at 26 months as the only unique predictor (see Table 4).

Second, the predictors of IS behaviors at 77 months were examined. In the first step, SES and PLS scores accounted for 1.2% of variance, $F = 1.34$, $p = .27$. In the second step, RSM and IS scores at 15 and 26 months accounted for an additional 31.3% of variance. The total model accounted for 35.9% of variance, $F = 6.40$, $p < .001$, with IS behaviors at 26 months as the only unique predictor (see Table 4).

As previous research with an ASD sample found that gains in language from age 2 to age 3 years were associated with a reduction in repetitive behaviors (Paul et al., 2008; Ray-Subramanian & Weismer, 2012), we examined the specific predictive effect of change in language from 15 to 26 months and changes in RSM and IS RRBs over the same period as predictors of RSM scores at 77 months. Following the procedure described by Ray-Subramanian and Weismer (2012), unstandardized residual change scores for RSM, IS, and PLS scores were computed by regressing scores from 26 onto 15 months (e.g., PLS from 26 to 15 months). Full regression models for RSM and IS are shown in Table 5.

In a model predicting RSM, in the first step SES, initial PLS scores and change in PLS scores from 15 to 26 months accounted for 4.3% of the variance, $F = 1.25$, $p = .29$. In the second step, initial RSM and IS scores and the change in RSM and IS scores from 15 to 26 months accounted for an additional 24% of variance. The total model accounted for 28.3% of the variance, $F = 4.51$, $p < .001$, with initial RSM behaviors as well as the change in RSM behaviors from 15 to 25 months as unique predictors (see Table 5).

In a model predicting IS, in the first step SES, initial PLS scores and change in PLS scores from 15 to 26 months accounted for 4.8% of the variance, $F = 1.39$, $p = .25$. In the second step, initial RSM and IS scores and the change in RSM and IS scores from 15 to 26 months accounted for an additional 31.9% of variance. The total model accounted for 36.7% of the variance, $F = 6.62$, $p < .001$, with initial IS behaviors as well as the change in IS behaviors from 15 to 25 months as unique predictors (see Table 5).

Discussion

This is the first study to show that the two subtypes (RSM and IS) of RRBs in a general population community sample (a) are stable across three age-points from infancy through early childhood, and (b) develop relatively independently of each other. The results provide an important first step in understanding the emergence and developmental profile of potentially clinically significant RRBs.

RSM behavior scores were highest at 15 months and then decreased between 15 and 26, and again from 26 and 77 months. In contrast, IS were at the lowest at 15 months, significantly increased by 26 months, and then decreased towards 77 months. Although there was a trend for boys to have higher RSM and IS scores at all ages, the difference (compared to girls) reached statistical significance for RSM scores only at 77 months.

Taken together, the factor and regression analyses provide evidence that the two RRB subtypes, RSM and IS, do not co-develop but develop independently. In sum, RSM behaviors at 77 months were significantly predicted by RSM behaviors at 26 months, but not by IS behaviors at 15 and 26 months. Similarly, IS behaviors at 77 months were predicted by IS behaviors at both 15 and 26 months, but not by RSM behaviors at 15 and 26. Moreover, the change in the specific subtype of RRBs between the two earlier time points predicted only the same subtype of RRBs at 77 months, demonstrating developmental continuity. These findings were over and above the contribution of language and SES.

The findings indicate that RBQ-2 items provide a consistent measure of repetitive behaviors for very young children from a community sample and may help to inform comparisons for children with ASD. The pattern for each subtype at different ages reported in this study was different from those reported in studies of children with autism (with a range of cognitive abilities) in a study using the ADI-R by Richler et al. (2010). The children in their longitudinal study ranged from 2 to 9 years, an age range when both RSM and IS behaviors reduce according to the typical development pattern described above. Instead, Richler and colleagues found sustained high RSM behaviors and low, gradually increasing IS behaviors. These results for children with autism may be explained by developmental immaturity that maintained RSM behaviors at a higher level and possibly slowed the emergence of the IS behaviors.

Our finding that IS and RSM subtypes develop independently in a general population sample is broadly consistent with previously reviewed studies that suggest RRBs should be conceptualized as a multidimensional construct (Langen et al., 2010; Leekam et al., 2011). It will be important for future studies to incorporate neuroimaging into longitudinal designs to extend knowledge of how IS and RSM behaviors map onto neurobiological changes during regular and atypical development.

The findings reported in the current study may have clinical implications for the design of interventions for children with ASD. Not all RRBs are problematic or warrant an intervention and some may have a positive function or indicate a strength or skill. However some challenging RRBs can constitute a major barrier to learning and social adaptation as well as a source of stress for parents in atypical development (Harrop, McBee, & Boyd, 2016; South, Ozonoff, & McMahon, 2005). Interventions specifically focused on these RRBs in children with ASD and other neurodevelopmental disorders are rare (NICE 2013). One very recent RRB intervention study (Grahame et al., 2015) that identified and targeted RRBs of

most concern to parents, reported results suggesting an intervention impact on preoccupations with restricted patterns of interest and limited play. These behaviors fit the IS subtype reported here. However, changes were not reported for the repetitive sensory and motor behaviors. Our finding, along with others', that RSM and IS behaviors appear to be relatively independent may facilitate understanding of the selective nature of the types of support and intervention strategies that may be needed. Further new evidence about the relation between IS behaviors and fears and anxieties in both ASD (Rodgers, Glod, Connolly, & McConachie, 2012; Lidstone, Uljarevic, et al., 2014; Uljarevic & Evans, 2016) and typically developing samples (Evans, Gray, & Leckman, 1999) indicates that it will be important to target anxiety as well as RRBs in future intervention research. Moreover, given our finding that IS behaviors identified in children as young as 15 and 26 months selectively predicted IS behaviors at 77 months, it will be important for future research to disentangle the form and function of subtypes of RRBs across time in order to target appropriate behaviors for individually tailored interventions.

The present study has a number of limitations. First, the number of children for whom there were RBQ-2 data across all three time points ($n = 87$) was too small to enable a more sophisticated analysis (e.g., growth curve modelling, bivariate latent change score models) to investigate the trajectories of the two subtypes or of particular RRBs. Further studies are required with larger general population samples to investigate how RRBs change over time. However, despite limitations, our results represent the first longitudinal evidence that RSM and IS subtypes are separate classes of RRBs, and that the two subtypes develop independently. Second, the RBQ-2 is a parent report measure. Parental characteristics and experiences may influence the reporting of children's behaviors, and children's behaviors are also likely to vary across settings. In future research, it will be important to include other

informants, as well as collecting data on the characteristics of parents. Third, there is a possibility that RSM behaviors are more easily observable and therefore reported more by parents than the IS subtype of RRBs (Leekam et al., 2011).

Furthermore, research to date using observational methods has tended to be confined to measuring behaviors in the RSM category and not in the IS category. However observational research measures of RRBs can be taken only within a brief snapshot of time, whilst parental observations are likely to more accurately reflect the child's usual behavior (Leekam et al., 2011). This might account for an incongruence of the present findings with those of Harrop et al. (2013), who reported on only one type of RRBs (fiddles with objects) in typically developing children at age 2. It will be important for future research to combine questionnaire, interview, and observational methods, and to identify individual differences in developmental trajectories of different RRBs subtypes over time, in order to explore factors that shape these potential different trajectories.

These findings will help inform future research on the genetic basis of repetitive behaviors and also their psychological functions in both typically developing and at risk populations. Future research exploring whether RSM and IS behaviors are differentially related to the emerging developmental trajectories of a range of cognitive processes might help identify potential endophenotypes associated with neurodevelopmental disorders such as autism. Potential candidates might include attention control, higher level executive skill, and social cognitive processes including emotion regulation, social imagination, and cooperation.

Conclusion

Despite limitations in terms of sample size, this paper presents the first longitudinal evidence from a community general population sample that the two subtypes of RRB (RSM and IS) develop independently of each other from infancy to early childhood. As such, the results provide an initial description against which behavior seen in children later diagnosed

1 with ASD and other neurodevelopmental disorders can be compared. If replicated, these
 2 findings also have implications for future genetic and cognitive studies of RRBs in typical
 3 and atypical populations, and potentially for the design of early interventions for problematic
 4 RRBs.

5

6

7

8

9

10

11

12

13

References

- American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders* (5th ed.). Arlington, VA: American Psychiatric Publishing.
- Arnott, B., McConachie, H., Meins, E., Fernyhough, C., Le Couteur, A., Turner, M., ... & Leekam, S. (2010). The frequency of restricted and repetitive behaviors in a community sample of 15-month-old infants. *Journal of Developmental and Behavioral Pediatrics*, 31, 223–229.
- 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. *Journal of Autism and Developmental Disorders*, 43(6), 1287-97.
- Bodfish, J. W., Symons, F. J., Parker, D. E., & Lewis, M. H. (2000). Varieties of repetitive behavior in autism: Comparisons to mental retardation. *Journal of Autism & Developmental Disorders*, 30(3), 237–243.
- Boucher, J., & Lewis, V. (1997). *Preschool Language Scale-3 (UK)*. The Psychological Corporation, Harcourt Brace & Company.
- Cannon, D. S., Miller, J. S., Robinson, R. J., Villalobos, M. E., Wahmhoff, N. K., Allen-Brady, K., ... & Coon, H. (2010). Genome-wide linkage analyses of two repetitive behaviors phenotypes in Utah pedigrees with autism spectrum disorders. *Molecular Autism*, 1(3), 1–13.
- Cevikaslan, A., Evans, D. W., Dedeoglu, C., Kalaca, S., & Yazgan, Y. (2014). A cross-sectional survey of repetitive behaviors and restricted interests in a typically developing Turkish child population. *Child Psychiatry and Human Development*, 45(4), 472-82.
- DeVellis, R.F. (2012). *Scale development: Theory and applications*. Los Angeles: Sage.

- 1 Evans, D.W., Leckman, J.F., Carter, A., Reznick, J.S., Henshaw, D., King, R.A., & Pauls,
2 D. (1997). Ritual, habit, and perfectionism: The prevalence and development of
3 compulsive-like behavior in normal young children. *Child Development*, 68, 58–68.
- 4 Evans, D. W., Gray, F. L., & Leckman, J. F. (1999). The rituals, fears and phobias of young
5 children: Insights from development, psychopathology and neurobiology. *Child*
6 *Psychiatry and Human Development*, 29, 261– 276.
- 7 Fernald, A., Marchman, V. A. and Weisleder, A. (2013), SES differences in language processing skill
8 and vocabulary are evident at 18 months. *Developmental Science*, 16, 234–248.
- 9 Fenson, L., Bates, E., Dale, P. S., Marchman, V. A., Reznick, J. S., & Thal, D. J. (2007).
10 *MacArthur-Bates communicative development inventories*. Baltimore, MD: Brookes.
- 11 Georgiades, S., Papageorgiou, V., & Anagnostou, E. (2010). Brief report: Repetitive
12 behaviors in Greek individuals with autism spectrum disorder. *Journal of Autism and*
13 *Developmental Disorders*, 40(7), 903–906.
- 14 Glenn, S., Cunningham, C., & Nanandidou, A. (2012). A cross-sectional comparison of
15 routinized and compulsive-like behaviours in typical children aged from 2 to 11 years.
16 *Eur J Dev Psychol* 9(5):614–630
- 17 Grahame, V., Brett, D., Dixon, L., McConachie, H., Lowry, J., Rodgers, J., ... & Le Couteur,
18 A. (2015). Managing Repetitive Behaviours in Young Children with Autism
19 Spectrum Disorder (ASD): Pilot Randomised Controlled Trial of a New Parent Group
20 Intervention. *Journal of Autism and Developmental Disorders*, 45(10), 3168-82.
- 21 Harrop, C., McConachie, H., Emsley, R., Leadbitter, K., Green, J., & PACT Consortium.
22 (2014). Restricted and repetitive behaviors in autism spectrum disorders and typical
23 development. *Journal of Autism and Developmental Disorders*, 44(5), 1207–1910.
- 24 Hollingshead, A. A. (1975). Four-factor index of social status. Unpublished manuscript, Yale
25 University, New Haven, CT.

- 1 Honey, E., McConachie, H., Turner, M., & Rodgers, J. (2012). Validation of the repetitive
2 behaviour questionnaire for use with children with autism spectrum
3 disorder. *Research in Autism Spectrum Disorders*, 6(1), 355–364.
- 4 Horn, J. L. (1965). A rationale and test for the number of factors in factor analysis.
5 *Psychometrika*, 30, 179-185.
- 6 Larkin, F., Meins, E., Centifanti, L. C. M., Fernyhough, C., & Leekam, S. R. (2016). How
7 does repetitive behavior relate to language and social cognition in typical
8 development? *Development and Psychopathology*.
- 9 Lam, K. S. L., Bodfish, J. W., & Piven, J. (2008). Evidence for three subtypes of repetitive
10 behavior in autism that differ in familiarity and association with other symptoms.
11 *Journal of Child Psychology and Psychiatry*, 49, 1193–1200.
- 12 Langen, M., Durston, S., Kas, M. J. H., Van Engeland, H., & Staal, W. G. (2011). The
13 neurobiology of repetitive behavior: . . . and men. *Neuroscience & Biobehavioral*
14 *Reviews*, 35, 356–365.
- 15 Leekam, S.R., Tandos, J., McConachie, H., Meins, E., Parkinson, K., Wright, C., ... & Le
16 Couteur, A., (2007) Repetitive behaviours in typically developing 2 year olds *Journal*
17 *of Child Psychology and Psychiatry* 48(11), 1131-1138.
- 18 Leekam, S. R., Prior, M. R., & Uljarevic, M. (2011). Restricted and repetitive behaviors in
19 autism spectrum disorders: A review of research in the last decade. *Psychological*
20 *Bulletin*, 137(4), 562–593.
- 21 Lewis, M. H., Bodfish, J. W. (1998). Repetitive behavior disorders in autism. *Mental*
22 *Retardation and Developmental Disabilities Research Reviews*. 4, 80–9.
- 23 Lidstone, J., Uljarević, M., Sullivan, J., Rodgers, J., McConachie, H., Freeston, M., ... &
24 Leekam, S. R. (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.

National Institute for Health and Care Excellence. (2013). Autism: the management and support of children and young people on the autism spectrum. (Clinical guideline 170.).

Ozonoff, S., Macari, S., Young, G. S., Goldring, S., Thompson, M., & Rogers, S.

J. (2008). Atypical object exploration at 12 months of age is associated with autism in a prospective sample. *Autism*, 12, 457–472.

Patterson, J, Mockford, C, Barlow, J, Pyper, C & Stewart-Brown, S (2002) Need and demand for parenting programmes in general practice. *Archives of Disease in Childhood*, 87, 468-471.

Paul, R., Chawarska, K., Cicchetti, D., & Volkmar, F. (2008). Language outcomes of toddlers with autism spectrum disorders: a two year follow-up. *Autism Research*, 1, 97–107.

Richler, J., Huerta, M., Bishop, S. L., & Lord, C. (2010). Developmental trajectories of restricted and repetitive behaviors in children with autism spectrum disorders. *Development and Psychopathology*, 22, 55–69.

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, 2494-2409.

Szatmari, P., Georgiades, S., Bryson, S., Zwaigenbaum, L., Roberts, W., Mahoney, W., ... & Tuff, L. (2006). Investigating the structure of the restricted, repetitive behaviors and interests domain of autism. *Journal of Child Psychology and Psychiatry*, 47, 582–590.

Silverman, J., Smith, C., Schmeidler, J., Hollander, E., Lawlor, B., Fitzgerald, M., ... & Galvin P, (2002). Symptom domains in autism and related conditions: Evidence for familiarity. *American Journal of Medical Genetics*, 114, 64– 73.

- 1 Stevens JP (1992) Applied multivariate statistics for the social sciences (2nd edition).
2 Hillsdale, NJ:Erlbaum.
- 3 Tabachnick, B. G., & Fidell, L. S. (2014). *Using multivariate statistics (6th Edition)*. London:
4 Thelen, E. (1979). Rhythmical stereotypies in normal human infants. *Animal Behavior*, 27,
5 699–715.
- 6 Thelen, E. (1981). Rhythmical behavior in infancy: An ethological perspective.
7 *Developmental Psychology*, 17, 237–257.
- 8 Turner, M. (1995). *Repetitive behavior and cognitive functioning in autism* (Unpublished
9 doctoral thesis). University of Cambridge, England.
- 10 Uljarević, M., Evans D. W., Alvares, G. A., & Whitehouse, A. O. J. (2016). Relationship
11 Between Restricted and Repetitive Behaviours in Children with Autism Spectrum
12 Disorder and their Parents. *Molecular Autism*, 7:29.
- 13 Wing, L., Leekam, S. R., Libby, S. J., Gould, J., & Larcombe, M. (2002). The Diagnostic
14 Interview for Social and Communication Disorders: Background, inter-rater reliability
15 and clinical use. *Journal of Child Psychology and Psychiatry*, 43, 307–325.
- 16 Wolff, J. J., Botterton, K. N., Dager, S. R., Elison, J. T., Estes, A. M., Gu, H., ... & Piven, J.
17 (2014). Longitudinal patterns of repetitive behavior in toddlers with autism. *Journal*
18 *of Child Psychology and Psychiatry*, 55(8), 945-953.

- 1 Table 1.
- 2 *Factor analysis of RBQ-2 items at 15, 26 and 77 months.*

Questionnaire items within each factor	Item factor loading		
	15 Months	26 Months	77 Months
Factor 1: Motor/sensory (RSM)			
1. Arrange toys or other items in rows or patterns ¹	.48	.46	-
2. Repetitively fiddle with toys etc.	.69	.77	.81
3. Spin self around and around	.49	.65	.63
4. Rock backwards and forwards	.73	.65	.61
5. Pace/move around repetitively	.75	.77	.77
6. Repetitive hand/finger movements	.79	.76	.73
7. Have a fascination with specific objects ²	-	.	.59
8. Looks at objects from particular/unusual angles	.50	.46	.68
9. Special interest in smell of people/objects	.58	.47	.59
10. Have a special interest in the feel of different surfaces ³	-	.	.62
Factor 2: Insistence on sameness (IS)			
11. Carries object with himself/herself ⁴	-	.44	.41
13. Insists on things (e.g. in house) remaining the same	.79	.72	.77
14. Gets upset about minor changes to objects	.69	.63	.74
15. Insists on aspects of routine remaining the same	.80	.69	.76
16. Insists on doing or re-doing things in a certain way	.74	.71	.81
17. Plays same music, game, video, book repeatedly ⁵	.46	.59	.-
18. Insists on wearing the same clothes/refuses new clothes ⁶	-	.48	.56
19. Insists on eating the same foods, or small range of foods	.40	.52	.55

3 Note: ¹Item 1 had insufficient loading at 77 months; ²Item 7 had insufficient loading at 15 and 26 months;

4 ³Item 10 had insufficient loading at 15 and showed cross loading onto both RSM (.47) and IS (.49) factors at 26;

5 ⁴Item 11 had insufficient loading at 15 months; ⁵Item 17 had insufficient loading at 77 months; ⁶Item 18 had

6 very low endorsement at 15 months; Item 12 "Collect or hoard items of any sort" had insufficient loading at all

7 three time points.

8

9

Table 2

Mean RBQ-2, PLS, and CDI scores at 15, 26, and 77 months

Age	RSM	IS	PLS	CDI
	Mean (SD)	Mean (SD)	Mean (SD)	Mean (SD)
15 months	1.80 (.44)	1.34 (.37)	37.28 (22.02)	43.03 (20.13)
26 months	1.51 (.41)	1.53 (.42)	48.52 (30.99)	45.99 (25.99)
77 months	1.33 (.38)	1.36 (.39)	NA	NA

Note: CDI: Communication Development Inventory; IS: insistence on sameness; PLS: Preschool Language Scales; RSM: repetitive sensory-motor; scale for IS and RSM is 1-3.

Table 3

Relationship between variables

	RSM 15	IS 15	RSM 26	IS 26	RSM 77	IS 77	SES	PLS 15	PLS 26	CDI 15	CDI 26
	months	months	months	months	months	months		months	months	months	months
RSM 15 months	1	.35**	.57**	.26*	.31**	.39**	-.15	.02	-.03	.14	-.03
IS 15 months	.35**	1	.25*	.34**	.09	.35**	-.04	.04	.03	.26*	.05
RSM 26 months	.57**	.25*	1	.40**	.52**	.33**	-.21*	-.11	-.14	.14	-.06
IS 26 months	.26*	.34**	.40**	1	.27*	.52**	.07	-.22*	-.11	.10	-.04
RSM 77 months	.31**	.09	.52**	.27*	1	.40**	-.19	-.16	-.19	.12	-.03
IS 77 months	.39**	.35**	.33**	.52**	.40**	1	-.18	-.21*	-.15	-.01	.02

Note: * $p < .05$; ** $p < .01$; CDI: Communication Development Inventory; IS: insistence on sameness; PLS: Preschool Language Scales; RSM: repetitive sensory-motor; SES: socio-economic status.

Table 4. Regression Models predicting RSM and IS behaviours at 77 months

	R2Δ	R2Δ Change	B	SEB	β		R2Δ	R2Δ Change	B	SEB	β
RSM 77 Months						IS 77 Months					
Step 1	.05					Step 1	.05				
SES			-.004	.003	-.14	SES			-.004	.003	-.15
PLS 15 months			-.004	.004	-.06	PLS 15 months			-.004	.004	-.12
PLS 26 months			-.003	.003	-.12	PLS 26 months			-.001	.003	-.04
Step 2	.29**	.25**				Step 2	.36**	.21**			
SES			-.002	.003	-.05	SES			-.004	.003	-.16
PLS 15 months			-.001	.003	-.02	PLS 15 months			-.003	.003	-.09
PLS 26 months			-.002	.002	-.08	PLS 26 months			-.002	.002	-.07
RSM 15 months			.02	.102	.02	RSM 15 months			.17	.09	.19
IS 15 months			-.01	.11	-.01	IS 15 months			.22	.11	.21
RSM 26 months			.44	.11	.47**	RSM 26 months			-.05	.11	-.06
IS 26 months			.06	.11	.07	IS 26 months			.34	.10	.36**

Note: * $p < .05$; ** $p < .01$; IS: insistence on sameness; PLS: Preschool Language Scales; RSM: repetitive sensory-motor; SES: socio-economic status.

Table 5. Regression Models predicting RSM and IS behaviours at 77 months using the change scores

	R2Δ	R2Δ Change	B	SEB	B		R2Δ	R2Δ Change	B	SEB	β
RSM 77 Months						IS 77 Months					
Step 1	.04	.008				Step 1	.05				
SES			-.004	.003	-.15	SES			-.005	.003	-.17
PLS 15 months			.01	.02	.28	PLS 15 months			-.01	.02	-.42
PLS 15 to 26 months			-.03	.05	-.38	PLS 15 to 26 months			.02	.05	.29
Step 2	.28**	.24**				Step 2	.37**	.32*			
SES			-.002	.003	-.08	SES			-.006	.003	-.21
PLS 15 months			-.005	.02	-.15	PLS 15 months			-.02	.02	-.61
PLS 15 to 26 months			.009	.04	.10	PLS 15 to 26 months			.04	.04	.51
RSM 15 months			.25	.09	.29**	RSM 15 months			.14	.09	.16
IS 15 months			.004	.11	.004	IS 15 months			.35	.103	.34**
RSM 15 to 26 months			.44	.11	.39**	RSM 15 to 26 months			-.05	.108	-.05
IS 15 to 26 months			.06	.10	.06	IS 15 to 26 months			.34	.09	.34**

Note: * $p < .05$; ** $p < .01$; PLS: Preschool Language Scales; RSM: repetitive sensory-motor; SES: socio-economic status.