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

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## 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

1 (Cevikaslan, Evans, Dedeoglu, Kalaca, & Yazgan, 2014; Evans et al., 1997; Leekam, et al.,  
2 2007) and children with diagnoses of ASD (Bishop et al., 2013; Georgiades, Papageorgiou, &  
3 Anagnostou, 2010; Honey, McConachie, Turner, & Rodgers, 2012; Lidstone et al., 2014).

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

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

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

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

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

1 earlier developmental form that predates IS behaviors, or whether RSM and IS behaviors are  
2 different classes of behaviors that develop independently. Clarifying whether one behavioral  
3 subtype has developmental significance for the other, may have clinical significance for  
4 intervention planning. It is known that some behaviors such as insistence on rigid  
5 preoccupations and extreme avoidant behaviors can be problematic for children with ASD  
6 and their families, however other types of RRBs (e.g., hand/finger movements) may have a  
7 positive function for self-soothing or attention-focusing (Leekam et al., 2011; Grahame et al.,  
8 2015). Evidence that IS and RSM behaviors develop independently of each other would  
9 support the notion that interventions should be designed to target the function of specific  
10 RRBs only, and delivered at a developmentally appropriate time point for the child.

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

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

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

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

## 21 **Method**

### 22 **Participants**

23           A community sample of 206 mothers and children (108 girls) was first recruited from  
24 health visitors' clinics and community organizations in the North East of England when  
25 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



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

## 5 **Materials and Methods**

### 6 **Procedure**

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

### 14 **Repetitive Behavior Questionnaire (RBQ-2)**

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

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

### 9 **Language**

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

### 21 **Analysis Plan**

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

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

13 In order to address the first aim—charting the frequency of RRBs in the same children  
14 at different ages—we compared frequency of mean IS and RSM behaviors across three time  
15 points using ANOVA. As noted in the Introduction, findings on the influence of gender on  
16 the expression of RRBs in typical development have been mixed, with some studies finding  
17 no gender effect (Evans et al., 1997; Zohar & Felz, 2011; Glenn Cunningham & Nananidou,  
18 2012) and other studies reporting higher levels of RRBs in males (Arnott et al., 2010;  
19 Cevikaslan et al., 2014; Leekam et al., 2007). Gender differences for each subtype were first  
20 explored using t tests. Separate hierarchical regression analyses were then used to examine  
21 whether RSM and IS behaviors tend to develop independently. This question was addressed  
22 through regression models exploring the contribution of same and different subtype behaviors  
23 at earlier ages on 77-month RRBs, with the effects of language and SES taken into account.  
24 Given previous findings showing that changes in language and changes in RRBs tend to  
25 interact during development in both TD and ASD populations, we also conducted additional

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

### 3 **Results**

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

#### 14 **Are Distinct Subtypes Found at Each Age?**

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

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

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

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

### 17 **RSM and IS levels at different ages**

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

1           There was a main effect of age for both mean RSM scores,  $F = 54.74$ ,  $p < .001$ , partial  
2   eta-squared= .39 and for mean IS scores,  $F = 12.21$ ,  $p < .001$ , partial eta-squared= .12. For  
3   RSM scores, post hoc tests using the Bonferroni correction showed that the mean RSM score  
4   at 15 months was higher than at both other ages (26 months:  $p < .001$ , Cohen's  $d = .67$ ; 77  
5   months,  $p < .001$ , Cohen's  $d = 1.13$ ), and the mean RSM score at 26 months was higher than  
6   at 77 months ( $p = .01$ , Cohen's  $d = .46$ ). For IS scores, post hoc tests showed that IS behavior  
7   scores at 26 months were higher than at 15 and 77 months (15 months:  $p < .001$ , Cohen's  $d =$   
8   .50; 77 months:  $p < .001$ , Cohen's  $d = .44$ ). See Table 2 for descriptive statistics.

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

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

14           Before conducting regression models we looked for evidence of multi-collinearity.  
15   The highest correlation was between RSM behaviors at 15 and 26 months,  $r = .58$ ,  $p < .001$ ,  
16   and between RSM at 26 and 77 months,  $r = .52$ ,  $p < .001$ ; all other correlations had  $r$ s of .38  
17   or lower, significantly lower than the .7 suggested as the threshold by Tabachnick and Fidel  
18   (2014). Furthermore, the lowest value for the collinearity diagnostics index in the regression  
19   model predicting RSM behaviors was .56 and in the one predicting IS was .50, also  
20   suggesting that multi-collinearity is not a concern. The relations among RSM and IS  
21   behaviors, and other variables of interest are presented in Table 3.

22           Using hierarchical regression, we first examined the predictors of RSM at 77 months.  
23   A summary of the full model is presented in Table 4. Analyses were first run with PLS scores  
24   and then re-run with CDI scores substituted<sup>1</sup>. In the first step, SES and PLS scores accounted

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<sup>1</sup>The pattern of results was identical as when run with PLS.

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

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

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

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

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

### 7 Discussion

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

13 RSM behavior scores were highest at 15 months and then decreased between 15 and  
14 26, and again from 26 and 77 months. In contrast, IS were at the lowest at 15 months,  
15 significantly increased by 26 months, and then decreased towards 77 months. Although there  
16 was a trend for boys to have higher RSM and IS scores at all ages, the difference (compared  
17 to girls) reached statistical significance for RSM scores only at 77 months.

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



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

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

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

1 most concern to parents, reported results suggesting an intervention impact on preoccupations  
2 with restricted patterns of interest and limited play. These behaviors fit the IS subtype  
3 reported here. However, changes were not reported for the repetitive sensory and motor  
4 behaviors. Our finding, along with others', that RSM and IS behaviors appear to be relatively  
5 independent may facilitate understanding of the selective nature of the types of support and  
6 intervention strategies that may be needed. Further new evidence about the relation between  
7 IS behaviors and fears and anxieties in both ASD (Rodgers, Glod, Connolly, & McConachie,  
8 2012; Lidstone, Uljarevic, et al., 2014; Uljarevic & Evans, 2016) and typically developing  
9 samples (Evans, Gray, & Leckman, 1999) indicates that it will be important to target anxiety  
10 as well as RRBs in future intervention research. Moreover, given our finding that IS  
11 behaviors identified in children as young as 15 and 26 months selectively predicted IS  
12 behaviors at 77 months, it will be important for future research to disentangle the form and  
13 function of subtypes of RRBs across time in order to target appropriate behaviors for  
14 individually tailored interventions.

15

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

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

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

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

## 21 **Conclusion**

22 Despite limitations in terms of sample size, this paper presents the first longitudinal  
23 evidence from a community general population sample that the two subtypes of RRB (RSM  
24 and IS) develop independently of each other from infancy to early childhood. As such, the  
25 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.

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- 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 <sup>1</sup>	.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 <sup>2</sup>	-	.	.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 <sup>3</sup>	-	.	.62
Factor 2: Insistence on sameness (IS)			
11. Carries object with himself/herself <sup>4</sup>	-	.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 <sup>5</sup>	.46	.59	.-
18. Insists on wearing the same clothes/refuses new clothes <sup>6</sup>	-	.48	.56
19. Insists on eating the same foods, or small range of foods	.40	.52	.55

- 3 Note: <sup>1</sup>Item 1 had insufficient loading at 77 months; <sup>2</sup>Item 7 had insufficient loading at 15 and 26 months;  
4 <sup>3</sup>Item 10 had insufficient loading at 15 and showed cross loading onto both RSM (.47) and IS (.49) factors at 26;  
5 <sup>4</sup>Item 11 had insufficient loading at 15 months; <sup>5</sup>Item 17 had insufficient loading at 77 months; <sup>6</sup>Item 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.  
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1 Table 2

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

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

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Table 3

*Relationship between variables*

	RSM 15 months	IS 15 months	RSM 26 months	IS 26 months	RSM 77 months	IS 77 months	SES	PLS 15 months	PLS 26 months	CDI 15 months	CDI 26 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
<b>RSM 77 Months</b>						<b>IS 77 Months</b>					
<b>Step 1</b>	.05					<b>Step 1</b>	.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
<b>Step 2</b>	.29**	.25**				<b>Step 2</b>	.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
<b>RSM 77 Months</b>						<b>IS 77 Months</b>					
<b>Step 1</b>	.04	.008				<b>Step 1</b>	.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
<b>Step 2</b>	.28**	.24**				<b>Step 2</b>	.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.