

The frequency of restricted and repetitive behaviors in a community sample of 15 month-
old infants

Running head: repetitive behaviors

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Objective: To investigate the frequency and pattern of a wide range of restricted and repetitive behaviors (RRBs) in the second year of life.

Method: Parents of 139 15-month-old typically developing infants from a community sample completed the Repetitive Behaviour Questionnaire-2 (RBQ-2), giving information on RRBs (e.g. stereotyped motor movements, sensory interests, routines and rituals and preoccupations with restricted interests) seen in their children.

Results: The RBQ-2 was found to be a reliable measure of these behaviors at this age and revealed a high frequency of particular types of repetitive motor movements in 15-month-olds.

Conclusion: These findings have implications for the early detection of disorders characterized by high levels of restricted and repetitive behaviors, such as Autism Spectrum Disorder (ASD).

Keywords: repetitive behaviors, typical development, RBQ-2, Autistic Spectrum Disorder

INTRODUCTION

Repetitive stereotyped behaviors such as rocking, flapping hands and banging objects are a normal part of infant development. Observational studies show that these behaviors are extremely frequent from 2-12 months and may have a functional significance in development as infants gain the neuromuscular control to execute complex goal driven actions^{1,2}. Although stereotypies are normal and frequent during infancy, they are also one of several types of repetitive behavior seen in clinical cases of Autism Spectrum Disorder (ASD). Four subtypes of restricted and repetitive behavior (RRB) are identified by the international classification systems DSM-IV³ and ICD-10⁴ as diagnostic criteria for autism. These are stereotyped motor mannerisms, preoccupations with part-objects or non-functional elements of objects (e.g. their sensory aspects), preoccupations with restricted, circumscribed patterns of interest and compulsive adherence to routines or rituals. Some of these repetitive behavior subtypes are also seen in other childhood clinical conditions such as Prader-Willi syndrome, obsessive compulsive disorder (OCD) and Williams syndrome.

Repetitive behaviors have received much less attention in research on the symptomatology of ASD than have social and communicative impairments, as it had been widely assumed that repetitive behaviors were not differentiating for ASD⁵. However, new observational studies of infant siblings at risk for ASD provide evidence that the earliest differences between high-risk siblings of children of autism and low-risk children may not be with social and communication impairments but with repetitive behaviors⁶. While these studies need further replication, specific differences have been found for some high risk siblings compared to control siblings as early as 12 months in

sensory reactions (unusual visual fixations on objects)⁷ and motor stereotypies, particularly arm and finger movements^{8,9}. Other observational research findings from 18- to 24-month-olds with communication delay show that those subsequently diagnosed with ASD had greater frequency and duration of repetitive motor movements, repetitive actions on objects, and sensory behaviors than comparison children with developmental delay or typical development¹⁰.

Given the clinical significance of repetitive behaviors for early diagnosis of ASD, there is a need to clarify the pattern of repetitive behaviors seen during typical development in the second year of life. There is virtually no evidence on the frequency of repetitive behaviors in a large sample of typically developing (TD) infants between 12 and 18 months, the age at which early behavioral signs of ASD may begin to emerge. The sample sizes of low risk infants in all the studies cited above were no greater than 35. Furthermore, in these studies, the type of repetitive behaviors investigated was confined to those that may be measured in an observation format, i.e., stereotypies and sensory reactions. These studies thus excluded routines and rituals or preoccupations that might only be seen at home. Given that the observation techniques used in each of these studies were also rather different from each other in terms of behaviors observed and length of observation, making comparison and consistency of measurement difficult, further validation of the presence of motor and sensory repetitive behaviors, as well as of other types of repetitive behavior, at this age is needed using other methods.

To our knowledge only two large-scale parent questionnaire studies have been carried out in early infancy with a community sample. The first used the Childhood Routines Inventory (CRI) by Evans and colleagues¹¹ and investigated one type of RRBs

(compulsive-like behaviors) in a cross sectional study of children between 8 and 72 months of age. Compulsive-like behaviors include rituals, routines, preoccupations with restricted interests, and insistence on sameness. This study found a relatively high frequency of these behaviors in 12- to 23-month-old infants, with lower frequency in an older sample of 5- to 6-year-olds. However, the CRI measure included only one item relating to repetitive actions and one item relating to sensory response, plus two other items measuring these aspects indirectly. Therefore little is known about the pattern of repetitive behaviors across the full range of expression including motor, sensory, routines and preoccupations of interest in this specific age band. Further, some of the CRI items seem inappropriate for young infants (e.g., making requests to postpone going to bed), and this may explain lower scores in infancy.

The second study investigated children's extreme intense interests. This could be considered to be a particular type of RRBs, i.e. circumscribed interests or preoccupation with a restricted interests. Using a parent questionnaire and follow up interview Deloache and colleagues¹² investigated extreme intense interests (EIIs) in children aged 11 months to 6 years ($N=177$). The authors found that 66% of children had an intense interest, with 29% of the whole sample being categorized as having an extreme intense interest (EII). There was a clear gender difference: 75% of those children who had an EII were boys, and the EIIs of boys tended to be rated as more intense. There also appeared to be clear gender differences in the content of EIIs of boys and girls, often following gender stereotypes. The age of onset of these EIIs ranged from 3 months to 42 months, with a mean onset of 18 months, and with 90% of EIIs appearing by age 2. The duration of the

EIIs was found to be long lasting (from 6 to 36 months). However, this study did not investigate RRBs beyond intense interests, such as rituals or repetitive motor movements.

Screening checklists such as the M-CHAT¹³, ESAT¹⁴ and Q-CHAT¹⁵ have been used with community samples of infants aged 14 to 54 months and include questions about repetitive behaviors. However, these questions are confined to only one or two items relating to motor stereotypies and to rigid behavior, such as questions about whether the child rocks their body, or about adaptation to a change in their routine, and do not include a comprehensive range of RRBs. To our knowledge, only two studies have specifically targeted the full range of autistic-like repetitive behaviors in infants using an informant questionnaire or interview method. One concentrated on children with medical conditions likely to result in a developmental delay¹⁶, finding elevated rates of repetitive/restricted behavior in children with ASD at 17 to 37 months of age, compared with a sample of infants with PDD-NOS and a large control group ($n= 499$) who did not qualify for a diagnosis. However their scale did not separate items into different behavioral subtypes (e.g., routines/stereotypies) and included non-repetitive items (e.g., eye-to-eye gaze). The other study found differences between repetitive behaviors in ASD and in children with developmental delay or typical development ($n=39$), but used a developmental interview that was not specific to RRBs¹⁷.

In the current study we used a parent questionnaire: the Repetitive Behaviour Questionnaire-2¹⁸ (RBQ-2) The RBQ-2 is a 20-item parental questionnaire measure that has recently been used to investigate the presence of the broader range of autistic-like RRBs (repetitive motor stereotypies, sensory interests, rituals and routines) in a general population sample of children aged 24 to 33 months. Research using this measure

indicates that RRBs of all kinds are common in TD 2-year-olds¹⁸, and factor analysis grouped these behaviors into 4 subtypes resembling the groupings reported in the DSM-IV and ICD-10 diagnostic criteria for autism. Support was also found for a 2-factor solution in which the four subtypes further combined into a motor and sensory subscale and a rigidity and insistence on sameness subscale.

The aim of the current study was to describe the frequency of a wide range of repetitive behaviors including motor, sensory, routines and rituals, and preoccupations with restricted interests in a community sample of 15-month-olds. This is an important age at which the earliest signs of ASD are beginning to emerge and consolidate. The findings will help to determine whether there is a similar frequency and pattern of behaviors at this age as was reported by parents of 2-year-olds in Leekam et al.'s study¹⁸. While observational research with typical infant populations up to 12 months suggests that repetitive motor behaviors may still be high at the end of the first year¹, large-scale cross-sectional questionnaire research indicates that compulsive-like behaviors may be lower at this age, reaching their peak between 24 and 35 months¹¹. The current questionnaire study documents the full range of RRBs in the second year of life for the first time.

A further aim was to investigate the reliability of utilizing the RBQ-2 with a younger age group. The psychometric properties of the questionnaire were found to be good in a group of 2-year-olds¹⁸, with good internal consistency reflected by a high Cronbach's alpha, but it is not known if this questionnaire is also suitable for use when infants are 15 months. Gender and SES have been shown to influence reports of RRBs,

with mothers of boys and of low SES children reporting higher RRBs,¹⁸ so these variables were also included.

METHOD

Participants

Mothers of children participating in an ongoing cohort study of children's development in a community sample were asked to complete the RBQ-2 when their child was 15 months of age ($M = 14.9$, $SD = 0.59$, range 13 -17 months). Mothers of 139 children (74 boys, 65 girls) completed the questionnaire. All responded to at least 90% of the items and so were included in the final sample. The majority of the children ($n=135$) were White and British. All children were born healthy and term. Children were tested with the Preschool Language Scale¹⁹ and scores showed a normal distribution. SES using Hollingshead's scale²⁰ represented the full range of deprived to affluent. The sample reflected the ethnic and socio-economic background of the local and regional area. The majority of the mothers who completed the questionnaire ($n = 71$, 51%) were not working outside of the home, and of the 68 who did work, only 15 (10%) worked full time.

Repetitive Behaviour Questionnaire-2¹⁸ (RBQ-2)

The RBQ-2 is a 20-item questionnaire that was designed from two existing interview measures: the Repetitive Behaviours Interview (RBI)²¹ and the Diagnostic Interview for Social and Communication Disorders (DISCO)²². Its psychometric properties have been found to be good¹⁸, with high internal consistency.

Parents are asked to rate the frequency with which their child has engaged in a range of restricted and repetitive behaviors over the last month. The items and the response choices are shown in Table 1. Item 20, which asks about a child's choice of toys, follows a slightly different format. Following the procedures of previous published research, a mean Total Repetitiveness score was calculated for each child by adding the score for each item (1-3) (including item 20) completed in the questionnaire and dividing by the number of questions completed by the respondent. This produced an average score, indicating the child's level of repetitiveness ranging from 1-3, for all items combined. Subscale scores were also calculated by summing across the items within each of the 2-factor and 4-factor solutions in the Leekam et al. study. For the 2-factor solution, the motor-sensory subscale contained 9 items and the rigidity, routines, and preoccupations subscale contained 8 items. For the 4-factor solution, the motor subscale contained 5 items, the rigidity subscale 7 items, the preoccupation subscale 7 items, and the sensory subscale 4 items.

Procedure

Mothers were asked to complete the RBQ-2 questionnaire in advance of a visit to the Child Development Unit at the University when their infants were 15 months of age. Mothers who did not return their questionnaire at this visit were given a prepaid envelope and asked to return their questionnaire by post. Only two mothers did not return the questionnaire. Any mothers with reading difficulties were helped to complete the questionnaire by a researcher during the visit.

RESULTS

Frequency of RRBs at 15 months

Table 1

Table 1 shows the individual questionnaire items and response options, and the number and percentage of participants completing them. The ‘occasional/mild’ response range was endorsed by between 2-52% of the sample for each item, the ‘marked’ range was endorsed by between 2-60% for each item. Of particular note are the high proportion of parents endorsing ‘Repetitively fiddle with toys or other items’ (60%) and ‘Pace or move around repetitively’ (51%) as marked behaviors at 15 months. Items with very low endorsement (70% or more never or rarely) at 15 months are: ‘Special interest in the smell of people or objects’, ‘Collect or hoard items’, ‘Insist on things remaining the same’, ‘Get upset about minor changes to objects’, ‘Aspects of daily routine must remain the same’, ‘Insist on doing things in a certain way or re-doing things until they are “just right”’, ‘Insist on wearing the same clothes or refuse to wear new clothes’ and ‘Insist on eating the same foods, or a very small range of foods’.

Total Repetitive Behaviours and Subscale Scores at 15 months

Analysis revealed that the total and subscale scores were positively skewed and therefore parametric tests were performed on logarithmically transformed scores.

Table 2

Table 2 shows the mean total and subscale scores for the sample ($N=139$). As can be seen from Table 2, results for the 2-factor subscales shows higher scores for the motor and sensory subscale than the rigidity/routines/preoccupations scale, $F(1, 138) = 188.74$, $p < .001$), while results for the 4-factor subscales shows that the scores are higher on the repetitive movements scale, than on the other scales, $F(3, 414) = 149.92$, $p < .001$).

To investigate the different subscale patterns and relations with gender, a series of mixed Subscale x Gender ANCOVAs were carried out with SES as a covariate. SES was included as a covariate to control for possible effects on maternal reports of RRBs. For the 2-factor model there was a significant main effect of Subscale, $F(1, 136) = 50.71$, $p < .001$, because the score on the Motor-Sensory subscale was significantly higher than the score for the Rigidity/routines/preoccupations subscale, $t(138) = 15.03$, $p < .001$. There was no significant effect of Gender, $F(1, 136) = 0.03$, n.s. and no interactions between Subscale x Gender, $F(1, 136) = 0.16$, n.s. For the 4-factor model there was a significant main effect of Subscale, $F(1, 136) = 40.55$, $p < .001$. The score on the Motor subscale was significantly higher than the score for the Rigidity subscale, $t(138) = 16.50$, $p < .001$, the Preoccupation subscale, $t(138) = 5.25$, $p < .001$, and the Sensory subscale, $t(138) = 11.84$, $p < .001$. The score on the Preoccupation subscale was significantly higher than the score for the Rigidity subscale, $t(138) = 16.82$, $p < .001$, and the Sensory subscale, $t(138) = 12.48$, $p < .001$. There was also a significant difference between the

scores on the Rigidity and Sensory subscales, $t(138) = 6.08, p < .001$, with greater frequency of Sensory scores. There was no significant effect of Gender, $F(1, 136) = 1.10, n.s.$, but there was an interaction between Subscale x Gender, $F(1, 136) = 5.61, p < .02$. Follow-up independent t-tests showed boys were showing greater frequency of Sensory RRBs compared to girls, $t(137) = 2.38, p < .02$.

RBQ-2 Reliability at 15 months

The internal consistency of responses on all items in the RBQ-2 was high (Cronbach's $\alpha = .85$). With respect to the Leekam et al. subscales: both of the 2-factor subscales showed good reliability (Cronbach's $\alpha = .81$ and $\alpha = .71$ for Motor-Sensory and Rigid/Routines/Preoccupations respectively), while for the 4-factor subscale, only the Repetitive Movements subscale and Rigidity subscale showed good internal consistency in this sample (Cronbach's $\alpha = .82$, and $\alpha = .74$) with the Preoccupation and Unusual Sensory Interest scales showing acceptable levels of reliability (Cronbach's $\alpha = .64$ and $\alpha = .51$ respectively). These results provide evidence that the RBQ-2 provides a reliable indication of total repetitive behaviors in children of this age group when utilizing either the total RRB score or the 2 factor subscales previously identified by Leekam and colleagues.

DISCUSSION

The aim of this study was to describe the frequency of RRBs and the pattern of the predominant subtypes of these behaviors at 15 months of age. We found that the

mean repetitiveness score for 15 month old infants was at a higher level than the score reported in published studies of 2-year-olds (mean total repetitiveness score 1.69, *SD* 0.33, compared with 1.54, *SD* 0.32,). In particular, the score on the motor subscale scores was high compared with the level reported in previous published studies of 2-year-olds (mean motor subscale score 2.09, *SD*, 0.58, compared with 1.52, *SD* 0.48,). Scores on other subscales were similar. Analysis of the subtypes of behavior also showed that parents of 15 month olds reported more frequent motor RRBs than any other subtype. This was the case at the subscale level analysis and also in individual item analyses. In contrast, previous research indicates that the most prevalent subtype of RRBs at 2 years was for more frequent preoccupation with restricted interests. This suggests that as children move from the second to the third year of life the main focus of their RRBs changes from stereotypies to preoccupations with restricted interests.

These findings extend the work of Thelen^{1,2} who found that a range of rhythmic stereotypies were common in TD infants from 2 to 12 months. The current study is the first to consider RRBs in TD infants between 12 and 18 months, with the exception of the study by Iverson and colleagues⁸ which followed children from 5 to 14 months of age, with a follow up at 18 months. Iverson et al. found that TD infants showed a peak of arm and finger rhythmicities during this period. However, the Iverson et al. study had a very small sample of TD infants and was an observational study limited to considering only motor stereotypies. Therefore this current study makes an important contribution to the literature by investigating a wide range of RRBs in a large sample in the second year of life. This is likely to coincide developmentally with increasing voluntary control of motor behaviors and goal-driven behaviors.

Previous research has shown that high-risk siblings of ASD individuals show greater frequency of motor stereotypies in the second year of life compared to controls, including arm/finger movements^{8,9}. This suggests that despite the frequency of motor RRBs in TD children at this age it is possible to detect potential difficulties. This is in contrast however to the work of Werner and colleagues¹⁷ which reported a lack of stereotyped/repetitive behaviors in TD children until age 2 and a lack of difference in motor RRBs at 15 months between ASD individuals and controls. It is possible to reconcile the results since the Werner study was retrospective when children were aged 3-4 years. It may be that parents of TD children whose RRBs had since subsided may have had difficulty recalling such behaviors, whereas parents of ASD children may have been ready to report such stereotypies as they were still ongoing.

A secondary aim of this study was to investigate the reliability of the RBQ-2 at 15 months for both the overall score and the 2 and 4 factor subscales identified in the Leekam et al. study. The RBQ-2 was shown to be a reliable instrument for use with children of this age. The Cronbach's alphas for the total scores and 2 subscales identified by Leekam et al. were good. The alphas for some of the 4 subscales were acceptable and this result may have been due to low levels of endorsement for a number of items on the Sensory and Preoccupation subscales. These items included 'Insistence on wearing the same clothes' which may not be developmentally age-appropriate and may only become important in the later preschool years. Other research with 2 year old TD children has also found routines and rituals to be rare at a young age^{23,24}. It is possible that it may be appropriate to exclude such items from infant versions of the RBQ-2 and include them in a later pre-school version. These results together suggest that the total RRB score and

Motor-Sensory and Rigid/Routines/Preoccupations subscales can be reliably used as early as 15 months of age. However, the 4-factor subscales are not recommended for this age group due to the lower levels of reliability.

Analyses were also performed to investigate relations with gender, since maternal reports of RRBs have previously been shown to relate to child gender. Results showed that there was no main effect of gender but there was an interaction with subscale at 15 months, with boys showing greater frequency of Sensory RRBs. In previous research, Leekam and colleagues found a difference using the RBQ-2 in a TD sample with boys showing higher levels of total RRBs at age 2¹⁸. Similarly, Allison et al.¹⁵ found a small but significant sex difference in a control group on the Q-CHAT measure, with boys showing more rigid and repetitive behaviors than girls at 18-24 months of age. Previous research by Deloache and colleagues¹⁴ focussing on one type of RRBs, extreme intense interests (EIIs), found these were influenced by gender in early childhood. Sex of the child was related to the frequency and intensity of the EIIs, with boys being more likely to have EIIs and their EIIs were rated as more extreme. Together these results suggest that gender differences in RRBs may be related to specific subscales at different ages, with males showing more frequent behaviors.

The findings reported here may provide baseline comparison data for other studies involving at-risk and typically developing samples. It may be particularly useful to gauge the developmental significance of behaviors that are seen more frequently in children who are subsequently diagnosed with ASD. Currently we know little about the developmental mechanisms that create an atypical trajectory for children with ASD. According to Thelen² RRBs provide a transitional state between uncoordinated motor

activity and goal-directed behavior, which is perhaps why there were such high rates of behaviors in this study such as pacing and actions on objects as these are among the major developmental achievements at 15 months of age. In the first year of life when motor stereotypies are common they may easily be triggered, later when goal directed behaviors are common then more extremes of arousal (low or high) will be required to release such behaviors. Following Thelen's analysis, it is helpful to think of RRBs such as motor stereotypies at older ages as immature responses that are maintained inappropriately in the behavioral repertoire. These behaviors can be considered as an imbalance between inner state of arousal and the environment and it is necessary for future research to consider how such imbalance occurs and why RRBs rather than goal-directed actions are selected.

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