

Restricted and Repetitive Behaviors in Autism Spectrum Disorders and Typical Development: Cross-Sectional and Longitudinal Comparisons

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Abstract Restricted and repetitive behaviors (RRBs) are characteristic of autism spectrum disorders (ASD). However, compared to social and communicative impairments, less is known about their development, trajectory and etiology. This study explored RRBs in young children with ASD matched to typically developing (TD) children on non-verbal development. RRBs were coded from direct observation at three time points within 13 months of development. Children with ASD displayed higher frequency and greater diversity of RRBs at all time points,

however RRBs were not unique to ASD and evident in the TD control group albeit at a reduced frequency. RRBs did not correlate with social and communicative impairments in the ASD group, suggesting dissociation between these domains.

Keywords Autism spectrum disorder · Repetitive behaviors · Measurement longitudinal

Sections of this work have been presented at the International Meeting for Autism Research (2009; 2013).

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Introduction

Restricted and repetitive behaviors (RRBs) and interests, together with social-communication impairments, are the core diagnostic domains of autism spectrum disorder (ASD; Diagnostic and Statistical Manual of Mental Disorders; DSM-5, American Psychiatric Association 2013). RRBs are defined as the expression of repetitive mannerisms, overriding preoccupations with objects/parts of objects, sensory behaviors and strict adherence to routine and ritual (Richler et al. 2007). Kanner (1943) included RRBs in his original descriptions of the disorder; however, in comparison to the social-communication domain of ASD, RRBs are under-researched and less is known about their development, aetiology and relationship to other areas of development. The emergence and presence of RRBs have been identified early in development (and often prior to social-communication impairments) through various prospective studies exploring the emergence of ASD symptoms (Chawarska et al. 2007; McConachie et al. 2005; Ozonoff et al. 2008) supporting the need for further research into this domain.

Restricted and repetitive behaviors represent an extremely heterogeneous group of behaviors and whilst most children with ASD will show these behaviors at some point

during their development, not all children will demonstrate these pervasively (Turner 1999; Walker et al. 2004). Likewise RRBs are not unique to children with ASD and feature in both typical development (e.g. Leekam et al. 2007) and other developmental disorders (Mahone et al. 2004; Matson et al. 2009; Moss et al. 2013; Tan et al. 1997). In typical development, the conceptualisation of RRBs overlaps with the descriptions of early repetitive motor behaviors and “just right” behaviors (Evans et al. 1997). In typical development these behaviors are assumed to serve the purpose of developmental mastery and thus reduce once developmental skills are gained. However unlike in typical development, RRBs are thought to be more frequent and exert a more pervasive influence in children with ASD, show less reduction over time and influence development (Charman et al. 2005; Honey et al. 2007).

RRBs in Autism

Categories of RRBs and Change Over Time

Restricted and repetitive behaviors in ASD have been categorised into two distinct, but not mutually exclusive, categories through factor analytic studies (Szatmari et al. 2006; Turner 1999). In their earliest form, RRBs commonly manifest as ‘lower order’ behaviors. These are characterized as repetitive motor actions and movements and physical and/or sensory manipulation of objects. ‘Lower order’ behaviors are more common in children who are developmentally delayed, have severe pragmatic language impairments and/or intellectual disabilities (Barrett et al. 2004; Lam et al. 2008). ‘Higher order’ behaviors involve more advanced cognitive functions and are characterized by the presence of routines, an insistence on sameness and circumscribed interests (Szatmari et al. 2006; Turner 1999).

The developmental trajectories of RRBs in ASD are complex and findings vary according to the types of behavior studied. Honey et al. (2008) explored change in RRBs (measured using the Autism Diagnosis Interview-Revised; ADI-R, Lord et al. 1994) in preschoolers diagnosed with ASD or core autism. Within a year the severity of RRBs had reduced in terms of the impact of the behaviors on child and family life. Improvement was greater in children with ASD than in those with core autism. Moore and Goodson (2003) followed 20 children with ASD using the ADI-R at two time points (ages 2 and 4 years). RRBs generally reduced with age but became cognitively more complex. While ‘lower order’ behaviors became less frequent they did not disappear. However, Richler and colleagues (Richler et al. 2010) tracked change in RRBs (assessed by the ADI-R) over a period of 9 years

and found that repetitive sensory motor behaviors remained high. In a cross-sectional cohort study of children with ASD, Militeri et al. (2002) explored the presence of RRBs in ‘toddlers’ (2–4 years) and ‘children’ (7–11 years) using a mixture of parental report data and observations of free play. Toddlers demonstrated more frequent motor and sensory RRBs than the older group.

Although RRB behaviors are heterogeneous, clear sub-categories of behaviors have been consistently identified (Turner 1999) with these categories showing different developmental trajectories and purposes (Militeri et al. 2002; Moore and Goodson 2003).

Associations with Other Autism Domains

The relationship between RRBs and the social and communication domain of ASD is not well understood. Some researchers have proposed that phenotypically RRBs are conceptually (and possibly biologically) dissociable from the social and communicative impairments in autism (Happé et al. 2006; Hus et al. 2007; Mandy and Skuse 2008; Ronald et al. 2005, 2006). Other studies have found associations between RRBs and impairments in the communicative and social domain but the relationships have not always been clear-cut and associations weak (Lam et al. 2008; Szatmari et al. 2006; Watt et al. 2008). Likewise, some studies have failed to find an association between RRBs and the early underlying deficit of joint attention (Mundy et al. 1994).

Associations with Other Areas of Development

The relationship between RRBs and other areas of development appears to vary by the category of RRB studied. ‘Lower order’ behaviors have been found associated with lower non-verbal and verbal IQ and poorer adaptive skills (Gabriels et al. 2005; Kim and Lord 2010; Lam et al. 2008; Militeri et al. 2002; Mooney et al. 2009; Szatmari et al. 2006). The association of ‘lower order’ behaviors, such as repetitive sensory motor behaviors, with younger chronological age (Lam et al. 2008; Szatmari et al. 2006) has also been found to be mediated by the presence of co-morbid intellectual disability (ID) and lower NVIQ scores (Esbensen et al. 2009).

Watt et al. (2008) assessed the association between RRBs and social behavior using the Communication and Symbolic Behavior Scales (CSBS; Wetherby and Prizant 2002). RRBs with objects correlated negatively with social skills; however the relationship was mediated by developmental level. In the same sample RRBs also correlated positively with social deficits on the Autism Diagnostic Observation Schedule (ADOS; Lord et al. 2002) at 3 years of age.

RRBs in Typical Development

Restricted and repetitive behaviors are assumed to be important in typical development, leading to mastery of actions and behaviors and the development of muscular control and action planning (Thelen 1979, 1981). Studies of the timing of onset and peak in development of RRBs in typical development have reported variable results. Through naturalistic observations of 20 typically developing infants in their first year of life, Thelen (1979, 1981) found these behaviors to be common with a range of different rhythmic and repetitive motor behaviors being observed. Repetitive behaviors were more adaptive, serving the purpose of developmental mastery, than self-stimulatory and decreased by the end of the first year. In contrast, however, Evans et al. (1997) found that parents reported these behaviors to be most common between 12 and 38 months of age and less common under 1 year. In a community sample aged 15 months Arnott et al. (2010) found that infants were reported regularly to demonstrate mostly 'lower order' RRBs.

When comparing typically developing children with children with ASD, most studies have found elevated rates of RRBs in the latter group irrespective of age (Honey et al. 2007; Kim and Lord 2010; Richler et al. 2007; Watt et al. 2008). Kim and Lord (2010) found RRBs were more common in TD toddlers than preschoolers, with the reverse pattern being found in children with ASD. However, two studies have found comparable rates of RRBs in TD and ASD children. Werner and Dawson (2005) coded RRBs from home videos at 12 and 24 months of age and found similar rates of RRBs in the two groups; whilst the TD children spent less time engaged in these behaviors, this difference was not significant. When matching children with ASD to TD children on developmental level rather than chronological age (as with Watt et al. 2008), Barber et al. (2012) found that while some behaviors remained more common in children with ASD, TD children demonstrated certain behaviors (e.g. lining up objects) more frequently. Thus the age at which the groups of children are compared influences the findings, with young TD children demonstrating significant frequency of RRBs.

Measuring RRBs

Despite attempts to clarify the development and profile of RRBs in ASD and TD, most research in this area relies on parental reports and there has been very little use of observational methods to capture RRBs in more ecologically valid environments/situations. With diagnostic measures such as the ADI-R or the ADOS, there is non-independence of measurement as they are used both to diagnose/classify children and to measure the frequency

and severity of RRBs. Free play, in particular, provides a valuable opportunity to observe the prevalence of RRBs in a naturalistic setting. To our knowledge, only two studies have explored the presence of RRBs within a free play session with the child's primary caregiver. Boyd et al. (2010) used the Direct Observation of Repetitive behaviors in autism (DORBA; Boyd et al. 2010) to measure RRB change following intervention. Militeri et al. (2002) also utilized a combination of parent report and free play sessions to explore the presence of RRBs in ASD, finding converging results across both methodological approaches. Two further studies (Barber et al. 2012; Watt et al. 2008) have used the behavioral section of the CSBS to measure RRBs in ASD, developmentally delayed (DD) and TD children.

However, observational methods have a number of disadvantages. For example, it is unclear how much diversity is captured during short observations and how much the setting influences the expression of RRBs (Gardenier et al. 2004). Observational methods also tend to limit the coding of RRBs to those which are behavioral in nature (i.e. 'lower order' RRBs). Nevertheless, this is less of a problem when assessing very young children as 'lower order' behaviors are more typical in early development.

Aims

The study aimed to build on previous research by studying the trajectory of RRBs within a short-term longitudinal study (three time points within 13 months). We used a systematic observation methodology embedded within a free play session with the child's primary caregiver to investigate the presence of 'lower order' RRBs in children in the toddler to preschool period (2–5 years at entry) in two groups of children (ASD and TD). Only one study to our knowledge (Militeri et al. 2002) has used a free play session with the caregiver to measure RRBs observationally.

Diagnosis was based on the ADOS and ADI-R, enabling us to explore how the observational measure correlated with two well-validated measures of overall symptom severity, while retaining independence of measurement in our primary measure.

Hypotheses

Based on previously published literature we predicted that:

1. Frequencies of RRBs would be higher in children with ASD than TD children matched on non-verbal development at each time point
2. Frequencies of RRBs would remain at a consistent level across the three time points in children with ASD, but reduce over time in TD children

3. Non-verbal development and language would correlate negatively with rates of RRB in both groups
4. Higher ASD severity scores would correlate positively with higher observed frequencies of RRBs.

Descriptive exploratory analysis was conducted to explore the different categories of RRBs in the two groups and over-time.

Method

Ethical approval for the research was obtained through the University of Manchester Medical and Human Sciences Ethics Committee and Central Manchester Multi-Centre Research Ethics Committee (05/Q1407/311) and parents gave written consent for their child to participate.

Participants

Two participant groups were sampled; (1) Toddlers and preschool children with core autism ($n = 49$; ASD Group; mean age 45 months); (2) Toddlers and preschool children with typical development matched to the ASD group on non-verbal development and gender ($n = 44$; TD Group; mean age 24 months).

ASD Group

The ASD group included all children recruited in one of the Preschool Autism Communication Trial (PACT) sites (Green et al. 2010). Referrals came from a range of health professionals within the UK National Health Service based on the child meeting screening criteria: i.e. a diagnosis or suspected diagnosis of ASD; aged between 2 and 4 years 11 months; families speaking sufficient English at home to take part in the trial. Exclusion criteria included severe hearing and/or visual impairments; twins; severe parental psychiatric condition requiring treatment and a diagnosis of epilepsy requiring regular medication. Due to the absence of a treatment effect on the ADOS-G social-communication algorithm in this site (Green et al. 2010) we were able to explore the development of RRBs and their associations with other aspects of development over time in both arms of the trial.

Following referral to the study, potential participants were assessed using a battery of standardized assessments; diagnosis of core autism was confirmed through administration of the ADI-R and ADOS. The Mullen Scales of Early Learning (MSEL; Mullen 1995) was administered to establish a non-verbal developmental age of 13 months or greater. Only children who reached eligibility criteria on the MSEL, ADOS-G algorithm and above autism threshold

on two of the three ADI-R domains were included in the final sample.

TD Group

Children with no known developmental or cognitive impairment were recruited as a control group through advertisements in local nurseries and media. Inclusion criteria included the child being aged between 1 and 5 years of age, with a non-verbal developmental age of at least 13 months at study entry. Any atypical developmental profile or family history of ASD was established through initial screening and those children excluded from further study. One child was excluded following initial screening due to having a sibling with ASD.

Matching Procedure

Non-verbal development was selected as the appropriate group matching variable due to the severity of language impairments in the ASD group. As many of the children in the ASD group had a language age equivalent <12 months matching would have required TD controls to be in infancy, making a cross comparison of behavior difficult. Children in the TD group completed the MSEL upon entry to the study. Only children who could be individually matched on MSEL development quotient and gender were included in the final sample. Sixty-six children were initially screened for inclusion with 44 completing the final matched sample. Of the families not included in the final sample, we were unable to contact three following initial screening and 21 were not suitable matches based on MSEL scores.

Time Points

Children were seen at three time points (T1, T2 and T3). T1 visits were entry assessments for the children entering the study. T2 visits were conducted 7 months post entry and T3 visits were 13 months post entry. The study therefore looked at the trajectory of RRBs within a 13-month time-frame as part of a short-term longitudinal observational study. The majority ($N = 46$) of assessments in the ASD group were completed in a laboratory setting (two at home, one at school), with only three cases seen in their home or school. The TD controls were seen at home.

Measures

In addition to the ADOS (ASD Group only), ADI-R (ASD group only) and the MSEL, children in both groups completed the Preschool Language Scales (PLS; Zimmerman et al. 1992). Socio-economic status (SES) data were also

collected; indices of mass deprivation (IMD) scores were calculated based on the participants' postal codes and are an accepted measure of SES in the United Kingdom. IMD scores provide an index of deprivation within small geographically defined areas based on seven different dimensions (income; employment; health and disability; education, skills and training; barriers to housing and services; living environment; crime). IMD scores range from 0 to 100 with lower IMD scores indicating less deprivation.

Measurement of RRBs

Children were filmed playing with their caregiver for up to 20 min using a standardized set of toys selected for developmental appropriateness and variety (see section "Appendix" for full list). Through utilizing a large set of toys, children were unlikely to become accustomed to the free play session over the three time points and were thus more likely to show a range of behaviors. Caregivers were asked to play with their child as they would at home using as many or few toys as they wished.

Observational coding definitions of RRBs were based on items from previously validated measures; the Repetitive Behavior Questionnaire-2 (RBQ-2; Leekam et al. 2007), the Repetitive Behavior Scales-Revised (RBS-R; Bodfish et al. 1999) and the Diagnostic Interview for Social and Communication Disorders (DISCO; Wing et al. 2002). In addition, items from the Direct observation of repetitive behaviors in autism (DORBA; Boyd et al. 2010) and the coding scheme of Barber et al. (2012) and Watt et al. (2008) were included. Together these items were expected to capture the range of behaviors likely to be shown within a free-play session.

Operational definitions of behaviors and modifications to items are reported in Table 1. Items were selected based on the likelihood of being observed within a 10 min play session. Items representing 'higher-level' RRBs were not included as these are less common in toddlers/preschool children and not likely to be readily apparent within a limited observation period with standardized toys. Collapsing of items into broader categories was based on Honey et al.'s (2007) factor analysis which derived two factors from items from the DISCO, RBQ and original items in children with ASD and TD. As we drew from the RBQ-2 and the DISCO in the development of our coding scheme, items were only grouped together if they loaded onto the same factor previously found by Honey et al. (2007).

For behaviors such as "flapping", we did not count each individual "flap" but instead scored each "burst/bout" of flapping. This was based on pilot observations that indicated that children tended to engage in this behavior in

"bursts" then either briefly stop and demonstrate this behavior again, or revert to another activity (such as play).

All RRB events observed within 10 min of play were coded using Noldus Observer[®] (Noldus 1991). Ten minutes has been used frequently in observational research and is considered to represent an acceptable time frame to code a range of behaviors (Bornstein et al. 1996). A 10 min warm-up period was allowed to settle the child into the observation situation. This was employed in order to allow the children with ASD to familiarize themselves with their environment as the free play session occurred in an unfamiliar lab setting. It has been shown that RRBs are related to anxiety level in children with ASD (Rodgers et al. 2012) therefore we predicted that elevated rates would be shown in this initial period. Therefore, whilst not as ideal as having the assessment context the same between the two groups, we used this "warm-up" period to try and capture RRBs once anxiety may have been alleviated. If the total recording time was shorter than 12 min, the coding started 10 min from the absolute end of the clip. No videos were <9 min in total.

All cases were coded by the first author who was not blind to study purpose or group allocation. Inter-rater reliability (IRR) was completed on 21 % of the cases by a graduate research assistant after training by the first author. Whilst the second coder was blind to the purpose of the study, neither coder was blind to group allocation due to the age differences between the groups and different assessment contexts. IRR cases were randomly selected across the two samples and all three timepoints. IRR was calculated using intra-class correlation (ICC) coefficients. The ICC for total RRBs was 0.89. One individual category (touches part of body repeatedly) was not endorsed in any of the IRR files therefore ICCs are not available for this item. Of the remaining ten items, ICCs ranged from 0.71 to 0.99 with the exception of "unusual interest in smell/feel/sounds." The ICC for this item was 0.46 attributable to the infrequent occurrence of this behavior in the IRR sample with only 8 % of the double-coded tapes endorsing this item.

Data Analysis

Statistical analysis was completed using SPSS 20. Between group differences were explored using One-Way ANOVAs. Effect sizes were calculated for significant differences. To explore change in RRBs over time, a repeated measures ANOVA was run with the variables of group (ASD and TD) and time points (T1, T2, T3). Further within group paired-sample t tests were conducted between successive time points and over the course of the study. Associations with developmental level and ASD severity

Table 1 Operational definitions of restrictive and repetitive behaviors (RRBs)

Item description	Original source(s)	Modifications made to conceptualization	Examples
Arranges objects in rows/patterns/stacks	RBQ-2, RBS-R	Stacking of toys added. To score this item must occur across 2 + objects or for a significant amount of time	Arranges toys cars in a line
Fiddles with objects/uses objects in repetitive and non-functional manner (e.g. spins, bangs, flicks)	RBQ-2, RBS-R, DORBA	N/A	Flicks dolls eyes repeatedly Pushes one button on a pop-up toy repeatedly
Spins/rocks/paces/jumps (whole body movements)	RBQ-2, RBS-R, DORBA	Groups together three behaviors of spinning, rocking and pacing	Rocks back and forth Spins around in circles
Unusual hand/finger mannerisms	RBQ-2, RBS-R	Bouts/bursts of behaviors coded if these behaviors occur in close succession. e.g. individual "flaps" are not scored whereas "bouts" of flapping are	Flapping Flicks fingers
Unusual interest in smell/touch/sounds	RBQ-2, DISCO, RBS-R	Groups together smell, feel and sounds	Rubs toys against face for tactual sensation
Sensitive to sounds or touch	DISCO	Touch added in addition to sounds	Covers ears in response to sounds Child "reacts" to parent rolling toy car on their leg
Touches part of body repetitively	RBQ-R	N/A	Repeatedly touches back of hand to side of face
Looks at objects at certain angles/visually inspects/brings objects close to eyes	DISCO, RBS-R	Brings objects close to eyes added in addition to angles	Child brings toys to eyes, inspecting certain aspects of toys (e.g. car wheels)
Bangs/taps/shakes/throws objects	Watt et al. (2008), Barber et al. (2012)	Shaking and throwing added in addition to bangs and taps	Taps toys with finger repeatedly Bangs toys together
Mouths objects repeatedly	Watt et al. (2008), Barber et al. (2012)	Modified from 'licks objects or surfaces'	Indiscriminately mouths toys
Repetitive/scripted language and/or intonation atypicalities	DORBA		Says phrase/word repeatedly

were examined using Pearson's correlations. Correlations were conducted separately for the two groups.

Results

Sample Characteristics

Sample characteristics for both groups are reported in Table 2. The groups did not differ in their combined MSEL fine motor and visual reception scores [$t(91) = -.69$; $p = .48$] indicating the groups were sufficiently matched on non-verbal ability (Mervis and Klein-Tasman 2004). The two groups differed in their chronological age with the TD group being younger [$t(91) = 11.09$; $p = .01$]. They also differed in their expressive [$t(91) = -3.19$; $p = .01$]

and receptive language [$t(91) = -3.52$; $p = .01$] with higher scores in the TD group. These differences were anticipated based on the matching procedure implemented.

Frequency of RRBs

At all three time points the ASD group demonstrated significantly higher frequencies of RRBs than the TD children (Table 3). The ASD group also displayed greater variability than the TD group as indicated by greater standard deviations. The most common behavior in both groups at all time points was 'fiddles with objects/uses objects in repetitive and non-functional manner.' The frequency of this item increased across the three time points in the ASD group (Fig. 1). This trend was not seen in the TD group (Fig. 2). In the ASD group 'bangs/taps/shakes/throws

Table 2 Sample characteristics

	ASD group (n = 49)		TD group (n = 44)	
	Male	Female	Male	Female
N	43	6	38	6
Chronological age	44.26 (8.81)	46.83 (7.41)	25.45 (9.16)	23.00 (6.26)
Non-verbal score	25.92 (6.84)	24.91 (5.15)	27.13 (7.29)	28.75 (8.02)
Language mean score	15.79 (10.05)	16.75 (9.32)	21.75 (8.82)	27.08 (11.02)
Receptive language score	16.23 (9.82)	17.33 (7.50)	21.37 (8.88)	25.67 (10.15)
Expressive language score	15.53 (10.77)	16.17 (11.34)	22.18 (9.04)	28.50 (12.03)
Indices of Mass Deprivation score	30.56 (20.03)	22.97 (21.90)	19.86 (12.54)	22.85 (18.34)
Ethnicity				
White (any background)	36	5	36	6
Other	7	1	2	0
Female caregivers	40	6	35	5
Male caregivers	3	0	3	1
ADOS module one:two	34:9	5:1	–	–
ADOS score	16.53 (2.52)	16.17 (4.16)	–	–

Language measure: PLS (Zimmerman et al. 1992)

Non-verbal development measure: MSEL (Mullen 1995)

‘Other’ ethnicities included Black African, Black Caribbean, Indian, Chines, Pakistani, Mixed race (White and Asian) and Mixed race (White and Black)

objects’ and ‘looks at objects at certain angles/brings objects close to eyes’ were also common behaviors. The greater variability in the ASD group is further shown in Figs. 1 and 2 with predominantly only one classification of behavior (fiddles with objects/uses objects in a repetitive and non-functional manner) endorsed in the TD group with many items not endorsed at all in the TD group, coupled

with more sensory behavior in the ASD group as indexed through higher frequency counts (Fig. 1).

There was a large main effect of group on frequency of RRBs [$\eta^2 = 0.37$] with higher frequencies observed in the ASD group [$F(1,43) = 25.25, p < .01$]. The difference in frequency of RRBs between the ASD and TD groups was significant at each time point [T1; $F(1, 91) = 11.87, p < .01$; T2; $F(1,91) = 16.92, p < .01$; T3; $F(1,91) = 29.79, p < .01$] with significance increasing over the course of the study. The partial eta squared effect size for the difference between the groups at T1 was classified as medium based on Cohen’s (1986) categorisation [$\eta^2 = 0.11$]. The effect sizes for both T2 and T3 were considered large [T2: $\eta^2 = 0.16$; T3: $\eta^2 = 0.25$].

Table 3 RRBs by timepoint and group

	ASD group (n = 49)	TD group (n = 44)	F	p
T1				
Mean	7.31	2.68	11.87	.001**
Min	0	0		
Max	42	14		
SD	8.23	3.56		
T2				
Mean	8.14	2.77	16.92	.000**
Min	0	0		
Max	40	18		
SD	8.02	3.45		
T3				
Mean	8.35	1.70	29.79	.000**
Min	0	0		
Max	45	7		
SD	7.86	1.91		

** Indicates significant difference $<.001$

Change in RRBs

There was no main effect of time point on the frequency of RRBs [$F(2,86) = 0.06, p = .94$]. Additionally there was no interaction between group and time point [$F(2,86) = 1.86, p = .16$]. Looking at the two groups individually, slight increases were observed between each time point and over the duration study in the ASD group. None of these changes was statistically significant. In the TD group, a slight increase was seen at T2; however a reduction below T1 values was observed by the final time point. The decrease between T2 and T3 frequencies in the TD group approached significance [$t(43) = -1.96, p = .057$].

Fig. 1 Individual RRBs by time point: ASD Group

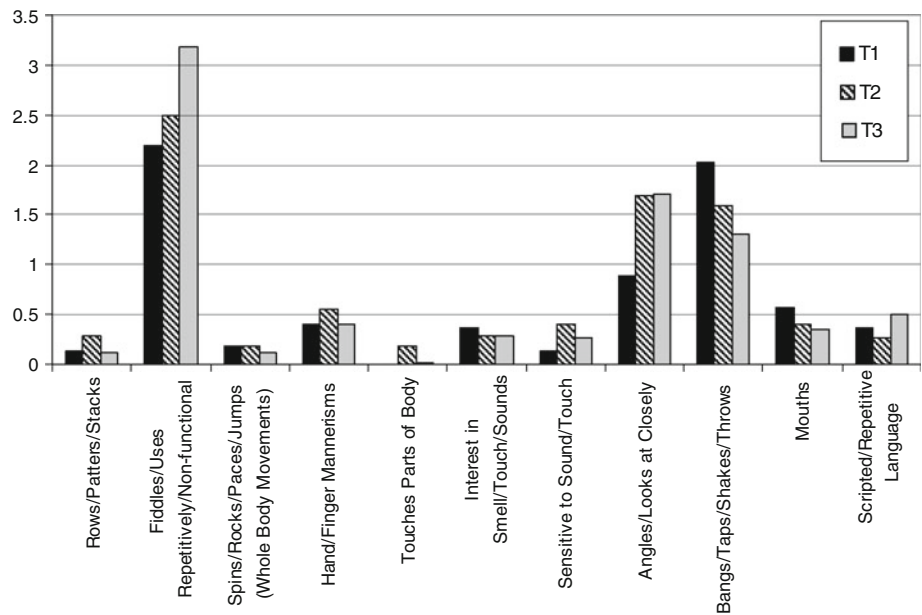
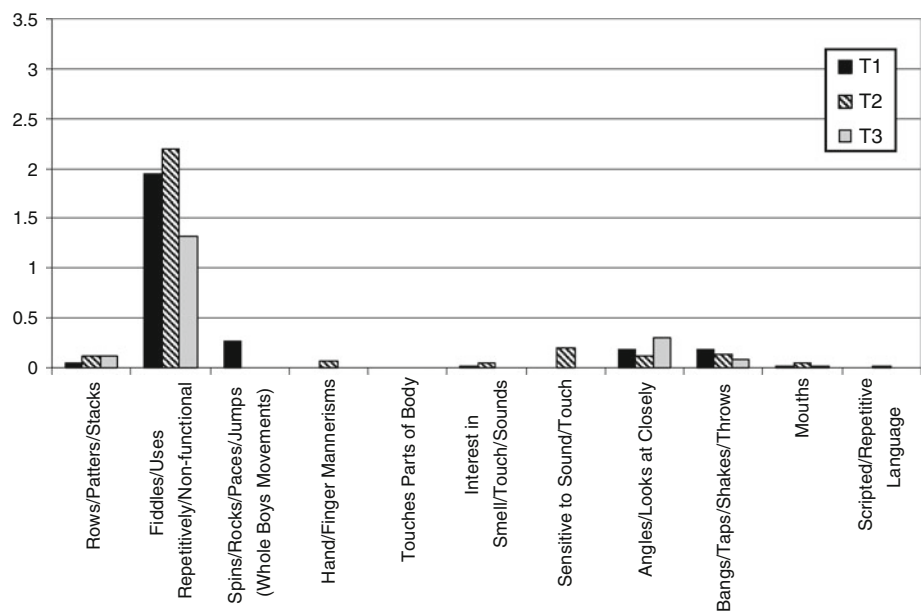


Fig. 2 Individual RRBs by time point: TD Group



Associations Between RRBs, NVIQ, Language and Chronological Age

Correlations between RRBs and developmental variables are reported in Table 4. In the ASD group, T1 NVIQ correlated negatively with the frequency of RRBs at all three time points [T1: $r(47) = -.35, p = .01$; T2: $r(47) = -.41, p < .01$; T3: $r(47) = -.43, p < .01$]. This relationship was not found in the TD group [T1: $r(42) = -.27, p = .07$; T2: $r(42) = -.12, p = .42$; T3: $r(42) = -.17, p = .25$]. Overall language (measured at T1) negatively correlated with RRB frequency at all time points in the ASD group [T1: $r(47) = -.31, p = .03$; T2: $r(47) = -.27, p = .05$; T3: $r(47) = -.44, p < .01$], however this relationship only just reached significance at T2. A similar pattern was found in

the TD group, however the relationship between RRBs and language did not reach significance at T2 [T1: $r(42) = -.34, p = .02$; T2: $r(42) = -.17, p = .26$; T3: $r(42) = -.34, p = .02$].

Chronological age did not correlate with RRBs at any time point in the ASD group [T1: $r(47) = -.02, p = .86$; T2: $r(47) = -.16, p = .26$; T3: $r(47) = -.07, p = .61$], but did correlate with T1 frequencies in the TD group [T1: $r(42) = -.38, p < .01$; T2: $r(42) = -.09, p = .54$; T3: $r(42) = -.28, p = .06$].

Associations Between RRBs and ASD Severity

T1 RRBs were as expected correlated with T1 ADOS-G ‘stereotyped behaviors and restricted interests’ algorithm

Table 4 Correlations between RRBs, NVIQ, language and chronological age by group

	T1 NVIQ		T1 language		T1 chronological age	
	ASD group	TD group	ASD group	TD group	ASD group	TD group
T1 total						
R	-.35*	-.27	-.30*	-.34*	-.02	-.39**
p	.01	.07	.03	.02	.86	.009
T2 total						
R	-.41**	-.12	-.27*	-.17	-.16	-.09
p	.003	.42	.05	.26	.26	.54
T3 total						
R	-.43**	-.17	-.44**	-.34*	-.07	-.28
p	.002	.25	.001	.02	.61	.06

Non-verbal development measure: MSEL (Mullen 1995)

Language measure: PLS (Zimmerman et al. 1992)

* Indicates significant correlation to 0.05

** Indicates significant correlation <.01

[$r(47) = .33, p = .02$]. This association was also found with T3 total RRBs [$r(47) = .31, p = .03$] but not T2 total [$r(47) = .23, p = .11$]. However there was no correlation between scores on the 'restricted, repetitive and stereotyped behaviors' subdomain of the ADI at T1 and RRB frequencies in the ASD group throughout the study [T1: $r(47) = -.19, p = .17$; T2: $r(47) = -.04, p = .79$; T3: $r(47) = -.03, p = .85$].

Total ADOS-G social-communication algorithm score at T1 was not significantly associated with the total of RRBs at any time point [T1: $r(47) = -.06, p = .68$; T2: $r(47) = .23, p = .11$; T3: $r(47) = .09, p = .54$]. Looking at the individual algorithm scores for 'reciprocal social interaction', RRBs frequencies did not correlate at any time point [T1: $r(47) = -.06, p = .71$; T2: $r(47) = .20, p = .17$; T3: $r(47) = .11, p = .45$]. Likewise scores on the 'communication' algorithm did not correlate with RRBs at any time point [T1: $r(47) = -.03, p = .83$; T2: $r(47) = .07, p = .65$; T3: $r(47) = .11, p = .44$]. RRBs frequencies did however correlate positively with scores on the ADOS-G 'imagination/play' algorithm [T2: $r(47) = .40, p < .01$; T3: $r(47) = .34, p = .02$]. This association was not found at T1 [$r(47) = .24, p = .09$]. Mirroring the findings with the ADOS-G algorithm scores, scores on the ADI Communication and Reciprocal Social Interaction subdomains also failed to correlate with RRB frequency at any time point [Communication; T1: $r(47) = -.18, p = .20$; T2: $r(47) = -.04, p = .79$; T3: $r(47) = -.02, p = .89$; Reciprocal Social Interaction; T1: $r(47) = .15, p = .30$; T2: $r(47) = .23, p = .10$; T3: $r(47) = .27, p = .06$].

Discussion

Our main aims in this study were to explore the presence of RRBs in ASD and TD using an observational method of coding within a free play session. We explored trajectories of RRBs (specifically those classified as 'lower order' behaviors) within a short-term longitudinal study while also exploring concurrent associations with developmental variables and autism severity and symptomatology. On the basis of previous parental report and observational research, four principal hypotheses were proposed.

In support of our first hypothesis, children with ASD were observed to show higher frequencies of RRBs than the comparison group of children without ASD over the course of the study. These behaviors were not absent in the TD groups, however were generally limited to one form of behavior ('fiddles with objects/uses objects in repetitive and non-functional manner'). This supports previous research (e.g. Arnott et al. 2010; Deloache et al. 2007) suggesting that in the early stages of development, RRBs are observable in typically developing children. The presence of RRBs early in typical development also supports the findings of studies using parent report (Arnott et al. 2010; Leekam et al. 2007) while adding to the literature by showing at multiple time points only one form of RRB was consistently shown in our TD sample and a wider range in the ASD group. This finding supports the observational data of Watt et al. (2008) and Barber et al. (2012) but also replicates their findings at multiple time points in children with and without an ASD.

The finding that predominantly only one form of RRBs was shown in the TD group, compared to a range in the ASD group is interesting as it implies these behaviors may serve different purposes in the two groups. In TD it is presumed that these behaviors are linked to mastery of developmental skills (Thelen 1979, 1981). This theoretical view overlaps with the behavior found most commonly in our TD sample ('fiddles with objects/uses objects in repetitive and non-functional manner'). This item generally manifests itself observationally through repetitive object play (for example, continuously pressing one button on a pop up toy), therefore TD children may exhibit these behaviors in order to learn how toys work and master their use. In ASD, it is presumed this item is additionally associated with an inability to shift attention, perseverance with a non-functional play act and to serve a self-stimulatory purpose (Cuccaro et al. 2003). Additional items, such as banging of toys and looking at toys at angles, were also endorsed more frequently in the ASD suggesting that the function of these behaviors differs across the two groups.

Whilst there was a large main effect of group on the data, we did not find an effect of time point nor an

interaction between group and time. Trends suggestive of a decrease in the frequency of RRBs in the TD group, and an increase in the ASD group, were observed; however these changes were non-significant. To our knowledge, this study is the first to explore developmental change in RRBs through direct observation in typical development. Frequency of RRBs in the ASD group remained consistent throughout the 13 months of the study. This finding supports the parent report data of Honey et al. (2008) and Richler et al. (2010) who similarly found 'lower order' behaviors remained constant in young children with ASD, particularly in those with a core autism diagnosis. Similar trajectories have also been reported by studies in non-autistic clinical samples (e.g. Mahone et al. 2004; Tan et al. 1997). It is however possible that 13 months is not a sufficient time frame to observe statistically significant change in either group. We found a greater range of RRBs in the ASD group, with more sensory behaviors shown across the three time points.

Our third hypothesis predicted that, irrespective of group, RRBs would be more frequent in children with lower non-verbal ability and language. These relationships were found at all observation points in the ASD group, supporting the findings of Lam et al. (2008), Militeri et al. (2002) and Szatmari et al. (2006). These associations were not found in the TD group despite the groups being matched on this variable. This finding suggests that this relationship may be unique to ASD and higher rates of RRBs are not associated with lower development in TD children. The majority of prior research has focused on NVIQ; however we found language also associated negatively with RRBs in both groups, as did Lam et al. (2008). Despite the differences in language abilities between the two groups, language associated with RRB frequency in ASD and TD. Chronological age did not consistently associate with RRB frequency at any time point in the study with only one correlation found in the TD group at T1, suggesting that RRB expression is not simply a by-product of age. Our findings are suggestive that the associations between RRBs and development vary between TD and ASD and the sub-domain of development.

Contrary to our fourth hypothesis, the frequency of RRBs was not associated with overall severity of social-communication impairment (ADOS algorithm scores). This is supportive evidence for the conceptual distinction between RRB and the communication and social impairments in ASD (Happé et al. 2006; Ronald et al. 2005, 2006).

On the other hand, ADOS algorithm scores for 'stereotyped behaviors and restricted interests' were associated positively with the frequency of observed RRBs, providing some validation for the methodology used in the study and indicating some consistency in children's behavior whether

interacting with a parent (in the current play-based observation) or with an examiner (in the ADOS). However this association was not found at all three time points, and the correlations were not high, suggesting that the ADOS and the play-based coding scheme also capture distinct behaviors. This suggestion of disparity between measurement approaches is further supported by the lack of association between the ADI-R subdomain score for 'restricted, repetitive and stereotyped behaviors' and the observed frequency of RRBs.

Finally, the ADOS 'imagination/play' algorithm scores were also positively correlated with the frequency of RRBs. Whilst we did not predict this association, both Bruckner and Yoder (2007) and Honey et al. (2007) found an association between RRBs and impairment in play in ASD when using parental report measures and these findings may be viewed as supporting the suggestion of Wing and Gould (1979) that RRBs may be the obverse of imagination.

Since the study involved piloting a new method for coding RRBs, certain limitations were unavoidable. The observational method of data collection meant we were unable to capture the developmental "shift" between 'lower' and 'higher' order RRBs. This problem was also encountered by Watt et al. (2008) and Barber et al. (2012) who similarly employed observational coding. Through observing RRBs within a short play session, coding focused on repetitive actions with toys, repetitive motor actions and sensory/self stimulatory behaviors. Observational coding across a range of contexts, both in free play and more structured assessments, is likely to increase both the frequency and variability of behaviors seen. The two groups were also filmed in different settings (clinic vs. home). Whilst research has shown that play behaviors are comparable across different settings (Bornstein et al. 1997), particularly when initially assessed children with ASD may have exhibited more RRBs than usual because they are faced with a new and unfamiliar setting. Additionally, children in both groups may become accustomed to the toys within the standardized set, reducing the novelty of such items on subsequent time-points.

We also recognize that the coders were not blind to study purpose and/or group allocation. As the primary coder (CH) developed the measure implemented in this research, this was an unavoidable side effect. Our second coder (for the purpose of IRR) was blind to the purpose of the study. However it was not possible for either to be blind to group allocation due to the different contexts in which the play session was filmed. Future research should aim to standardize settings further.

Whilst we studied the developmental course of RRBs over several observation periods, we did not find the predicted decrease over time in the TD group. It is possible

that 13 months is not a sufficient length of time to reflect meaningful change and that additional time points are required to observe this. It may also be that a play situation with a range of new toys constrains the types of RRBs that might be observed. A higher proportion of ASD children in the 2–3 years age group would also have been preferable as it is within this developmental window that atypical behaviors are assumed to emerge and deviate from the typical course of development.

As we were interested in the association between RRBs and language, we did not control for this variable in the group analysis despite difference in the two samples. Language differences were expected due to the matching approach taken and in line with previous research. However we found greater rates of RRBs in children with lower language abilities in both ASD and TD suggesting that this association is not a by-product of the matching differences. While we acknowledge this as a limitation, recruiting language matched typically developing controls would have led to the comparison group being poorly matched on chronological age as some of the children in the ASD group had a language age as low as 5 months of age (using the PLS). Recruiting young infants as a comparison group would have limited the behaviors observable as many of these children would not have the motor abilities to play independently making the measurement context incomparable. Ideally future research would benefit from the inclusion of both TD and DD children to compare to children with ASD.

Conclusions

Preschool children with ASD show more RRBs within a free play session than typically developing controls. This difference was consistently replicated over three observations in 13 months. RRBs did not change significantly in either group over the course of the study. Irrespective of group, higher frequencies of RRBs were associated with lower language; however RRBs only related to lower non-verbal developmental ability in the ASD group. In the ASD group, observed RRBs did not relate to severity of social interaction and communication impairments, suggestive of a dissociation between these domains in ASD.

Acknowledgments We are grateful to the families who participated in the research. Thank you to Faye Plummer for her work on the project and the Institute of Brain, Behavior and Mental Health Research Group at the University of Manchester for discussion of these findings. C.H. was supported by a University of Manchester Strategic Studentship as part of the Preschool Autism Communication Trial. H.Mc., R.E., K.L. and J.G. were also supported by the Medical Research Council

Appendix

Toys used in free play session:

- Garage
- Cars
- Tea-set
- Doll
- Toy food boxes
- Plastic food and utensils
- Shopping basket and cash register
- 2 × Telephones with pull along cord
- Marble maze and marbles
- Pop-up toy
- Stacking cups
- Bubbles
- Crayons and Paper
- Jigsaw

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