

Restricted and Repetitive Behaviors in Individuals with a History of ASDs Who Have Achieved Optimal Outcomes

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Abstract Studies of autism spectrum disorders (ASDs) suggest that restricted and repetitive behaviors (RRBs) are particularly difficult to remediate. We examined present and past RRBs in 34 individuals who achieved optimal outcomes (OOs; lost their ASD diagnosis), 45 high-functioning individuals with ASD (HFA) and 34 typically developing (TD) peers. The OO group exhibited minimal residual RRBs at the time of the study. All OO participants were reported to have at least one RRB in early childhood and almost 90 % met the RRB cutoff for ASD in early childhood, but RRBs were not more present in the OO than the TD group at the time of the study. History of RRBs in the HFA and OO groups differed only in oversensitivity to noise and insistence on sameness. Reports of current behavior indicated that RRB's had almost totally disappeared in the OO group. Thus, although RRB's were present in the OO group in childhood, they resolved along with social and communication deficits.

Keywords Optimal outcome · Restricted and repetitive behaviors · Autism

Introduction

Autism spectrum disorders (ASDs) are a group of neuro-developmental disorders believed to affect as many as one in 88 individuals (Centers for Disease Control and Prevention [CDC] 2012). These disorders are characterized by deficits in communication and socialization, as well as by the presence of restricted and repetitive behaviors (RRBs). ASDs are generally considered to be lifelong disorders (American Psychiatric Association [APA] 2000). However, several studies have indicated that a small proportion of individuals who are diagnosed with an ASD early in childhood experience a reduction in autism symptomatology to such a degree that they no longer meet diagnostic criteria for any ASD as they get older (Cohen et al. 2007; Fein et al. 2013; Harris and Handleman 2000; Howlin et al. 2004; Kelley et al. 2006, 2010; Lovaas 1987; Rutter et al. 1967; Sallows and Graupner 2005; Seltzer et al. 2004; Sigman and Ruskin 1999; Szatmari et al. 1989; Venter et al. 1992; Weiss 1999; Zachor et al. 2007). However, most of these studies did not describe in detail the functioning of these individuals compared to groups of individuals who still met criteria for high-functioning ASD.

Some studies documenting such positive outcomes among individuals previously diagnosed with ASD have suggested that these individuals continue to present with subthreshold symptoms of ASDs (Piven et al. 1996). This concern is supported by multiple studies that have demonstrated that core features of ASD fluctuate with age and generally tend to lessen in severity by the time that an individual reaches adulthood (Byrd 2002; Eaves and Ho 1996; Koboyashi et al. 1992; Leekam et al. 2011; Piven et al. 1996; Rumsey et al. 1985; Seltzer et al. 2004; Shea and Mesibov 2005). As a result of this line of research, Seltzer et al. (2004) discussed the possibility that reports of

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“recovery” in ASD may reflect this general tendency for symptoms of ASD to improve with age, but argued that the core features of ASD continue to be present in these individuals.

When considering developmental changes within the three clusters of ASD symptoms in high-functioning individuals, symptoms within the RRBs cluster tend to persist into adolescence and adulthood, even while social and communication symptoms in ASD may improve (Leekam et al. 2011; Murphy et al. 2005; Piven et al. 1996; Seltzer et al. 2004). Some types of RRBs (e.g., circumscribed interests) may even increase in severity with age (Bishop et al. 2006; Rutter 1978; South et al. 2005). Because symptoms within the RRBs cluster may be particularly difficult to remediate, reports of children who lose their ASD diagnosis as they mature have raised the question of whether RRBs may persist among these individuals.

Several studies explored the current functioning of children who were once diagnosed with ASDs and who have achieved “optimal outcomes (OOs)” (Fein et al. 2013; Kelley et al. 2006, 2010). These studies defined OOs as referring to children who were diagnosed with ASDs in the preschool period and at the time of the studies no longer met diagnostic criteria for any ASD, exhibited average IQ and were mainstreamed into regular education classrooms without one-on-one assistance. Thus far, studies exploring the functioning of individuals who achieve OOs have not methodically examined the presence of all types of RRBs among these individuals. Therefore, it remains possible that individuals who achieve OOs may exhibit some RRBs that cause impairment in functioning and may require continued intervention, even while they fail to meet diagnostic criteria for ASD because of an abatement of social and communication symptoms.

The current study compares children and adolescents with OOs to high-functioning individuals with a current ASD diagnosis (HFA) and typically developing (TD) peers. We predicted that the OO group would exhibit mild RRBs that might be clinically significant, but that these behaviors would not be as frequent or as severe as in the HFA group.

The secondary aim of this study is to examine parents’ report of RRBs in the OO group in early childhood and to compare the early presentation of RRBs in the OO group to that of the HFA group. By doing this, we may be able to identify ways in which the early history of these two groups differed. Previous research suggests that children who achieved OOs exhibited somewhat milder signs of ASD in early childhood (Sutera et al. 2007; Turner and Stone 2007). Additionally, researchers have argued that the presence of RRBs prevents a child from fully attending to the environment, which could make the child unavailable to receive meaningful input from the social environment. As a result, children who engage in RRBs early in development may do

so at the cost of activities that promote cognitive, social and communicative development, leading to delays in the development of these skills and potentially to a more negative outcome (Bodfish et al. 2000; Bopp et al. 2009; Lewis 2004). In support of this connection between RRBs and prognosis, several studies have demonstrated that children who display RRBs early in their preschool years tend to have poorer school-age language outcomes than children who do not exhibit these behaviors during this age period (Charman et al. 2005; Paul et al. 2008). Based on these studies, we predicted that individuals who achieved OOs would display fewer RRBs in early childhood than would the individuals in the HFA group.

Methods

Participants

The sample included in this study was previously described by Fein et al. (2013). Thirty-four individuals with a history of ASD who have achieved OOs, 45 individuals with HFA, and 34 TD peers were tested. Participants ranged from 8 years, 1 month to 21 years, 8 months. The groups did not differ on age, gender, and nonverbal IQ (NVIQ), but differed significantly on verbal IQ (VIQ). The VIQ of the OO and TD groups was about 7 points higher than the HFA group’s VIQ (see Table 1). Six HFA participants and three OO participants were evaluated at Queens University in Kingston, Ontario, Canada. Their performance did not significantly differ from the other participants on any measure. The participants tested at the University of Connecticut were primarily from the northeast US. Participants were predominantly Caucasian, with 3 OO individuals, 2 HFA individuals, and 3 TD individuals reporting other races or ethnicities. Parents of a subset of participants in each group completed two questionnaires designed to assess their child’s RRBs and circumscribed interests (see below). No significant group differences were observed in the gender, age, VIQ, and NVIQ of participants whose parents completed these measures and those who did not. The study was approved by the Institutional Review Boards of the University of Connecticut, the Institute of Living Hartford Hospital, Children’s Hospital of Philadelphia, and Queens University.

Recruitment was done through media outlets (newspaper stories, radio interviews), private practices, and clinic referrals. Recruitment materials stated that the study sought individuals “who have lost their ASD diagnosis and have reached an excellent outcome,” as well as individuals “with high functioning autism” and “typical development.” All three groups were recruited using the same materials and the same sites were used to recruit participants for all three groups. In some cases, therapists contacted parents of

Table 1 Participant characteristics

	TD	OO	HFA	<i>F</i> or χ^2	<i>p</i>	η^2	Games-Howell Post-hoc
N	34	34	45				
Sex	31 M; 3 F	27 M; 7 F	41 M; 4 F	3.01	0.22		
Age	13.87 (2.58) (9.93–21.71)	12.77 (3.45) (8.1–21.2)	13.76 (2.72) (8.6–20.0)	1.51	0.23	0.03	
WASI: VIQ	112.00 (11.17) (93–138)	112.65 (13.72) (80–137)	105.42 (14.22) (81–142)	3.71	0.03	0.06	OO, TD > HFA
WASI: NVIQ	112.79 (11.32) (89–139)	110.29 (15.07) (81–142)	110.09 (12.63) (78–147)	0.48	0.62	0.01	
Vineland: communication	93.44 (9.12) (78–119)	98.30 (12.66) (79–122)	82.83 (13.71) (42–108)	16.02	<0.001	0.23	OO, TD > HFA
Vineland: socialization	101.74 (8.56) (86–120)	102.03 (8.44) (80–118)	75.33 (15.87) (46–109)	64.20	<0.001	0.55	OO, TD > HFA
Vineland: daily living	88.76 (9.26) (74–115)	92.30 (15.88) (65–120)	75.58 (14.14) (46–110)	16.76	<0.001	0.24	OO, TD > HFA
ADOS: communication	0.41 (0.56) (0–2)	0.47 (0.61) (0–2)	3.44 (1.45) (1–7)	116.02	<0.001	0.68	HFA > OO, TD
ADOS: socialization	0.50 (0.75) (0–2)	1.09 (1.31) (0–4)	6.76 (2.19) (4–13)	186.50	<0.001	0.77	HFA > OO, TD

Table reports means, followed by SDs and ranges. WASI and Vineland subtest mean = 100, SD = 10

children suspected to have OOs, and in some cases, parents saw media reports about the study and contacted the investigators. Participants were also referred from the principal investigators' private practices, the Psychological Services Clinic at the University of Connecticut, and from other ongoing studies at the University of Connecticut. Finally, some participants in each group were informed about the study by other participants' families. See Fig. 1 for a flow chart of inclusion and exclusion.

Inclusion Criteria

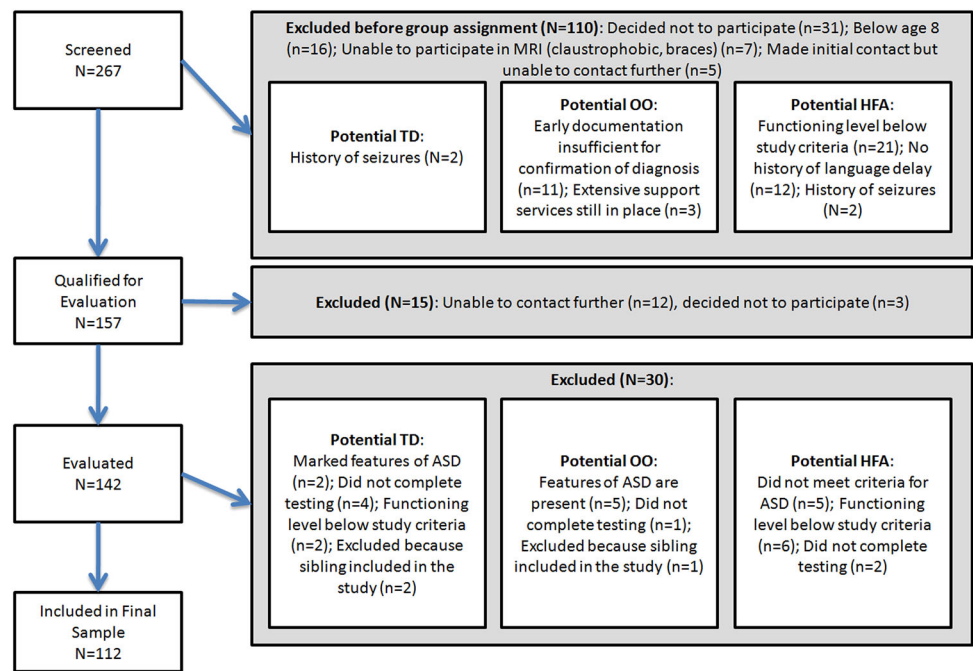
All participants had verbal, nonverbal, and full-scale IQ standard scores greater than 77 (within 1.5 standard deviations (SDs) of the average of 100) on the most current assessment. See Fein et al. (2013) for a flow chart depicting participant enrollment. Additional criteria were:

- For the OO group:
1. Participants had a documented ASD diagnosis made by a physician or psychologist specializing in autism

before the age of 5, verified in a written diagnostic report provided by parents. Early language delay (no words by 18 months or no phrases by 24 months) documented in the report was required. As a second step in confirming diagnosis, the report was edited to remove information about diagnosis, summary, and recommendations but leaving descriptions of behavior. One of the co-investigators (MB), an expert in diagnosis of ASD and Director of the University of Connecticut Psychological Services Clinic, reviewed these reports, blind to early diagnosis and current group membership. In addition to potential OO participants, she reviewed 24 "foil" reports for children with non-ASD diagnoses, such as global delay or language disorder. Four potential OO participants were rejected for insufficient early documentation, and were dropped from the study. All 24 foils were correctly rejected.

2. Participants could not currently meet criteria for any ASD according to the Autism Diagnostic Observation Schedule (ADOS; Lord et al. 2000) administered by a research-reliable interviewer. In addition, the ADOS of

Fig. 1 Flow chart of participant inclusion



all potential OO cases was reviewed by a clinician with more than 15 years of autism diagnostic experience (IME, MB, or DF) who confirmed that ADOS scores were below ASD thresholds and that in their expert clinical judgment, an ASD was not present.

3. Participants’ scores on the Communication and Socialization domains of the Vineland (see below) had to be greater than 77 (within 1.5 SDs of the mean of 100) (see Table 4).
4. Participants had to be fully included in regular education classrooms with no one-on-one assistance and no special education services to address autism deficits (e.g., no social skills training). However, participants could be receiving limited special education services or psychological support to address impairments not specific to ASDs, such as attention or academic difficulties.

For the HFA group:

1. Following Collaborative Programs of Excellence in Autism diagnostic guidelines (Luyster et al. 2005), participants had to meet criteria for ASD on the ADOS (both Social and Communication domains and total score) and according to best estimate clinical judgment.

For the TD group:

1. Participants could not meet criteria for any ASD at any point in their development, by parent report.
2. Participants could not have a first-degree relative with an ASD diagnosis

3. Participants could not meet current diagnostic criteria for an ASD on the ADOS, or by clinical judgment (see Table 1). There was no attempt to exclude TD children for other learning or psychiatric disorders (but see general exclusion criteria).
4. Scores on the Communication and Socialization domains of the Vineland had to be greater than 77 (see Table 4).

Exclusion Criteria

Potential participants for any group were excluded from the study if (1) at the time of the telephone screening they exhibited symptoms of major psychopathology (e.g., active psychotic disorder) that would impede full participation, (2) they had severe visual or hearing impairments, or (3) they had a history of seizure disorder, Fragile X syndrome, or significant head trauma with loss of consciousness. Two potential participants in the TD group and two in the HFA group were excluded because of possible seizure disorder; none were excluded for other reasons.

Procedure

Phone screenings based on study criteria were conducted with parents of each potential participant. Those who passed screening were scheduled for an assessment. For participants under 18, parent consent and child assent was obtained prior to testing. For participants 18 and over, their informed consent was obtained. The evaluation was

administered in a quiet room over the course of two or three testing sessions at the University of Connecticut, the Institute of Living of Hartford Hospital, Queens University, or in the home. Testing lasted approximately 6 h. In most cases, parent interviews were conducted concurrently by a second examiner and lasted approximately 3 h for the OO and HFA groups and 1.5 h for the TD group. To ensure that participants met the enrollment criteria for the study, measures examining current diagnostic status (i.e., OO, HFA, TD; see Measures below) were completed at the outset of the evaluation by the same examiner who completed the remainder of the assessment. The participant's history was discussed during the collection of these measures, and, as a result, the evaluators were not blind to the participant's presumed group status. Participants received a monetary incentive for participation, even if the testing could not be completed.

Measures

Autism Diagnostic Observation Schedule (ADOS; Lord et al. 2000). Module 3 or 4 (depending on age) was used to determine current diagnostic status for the OO and HFA groups, to rule out autistic features in the TD group, to compare social interaction in the OO and TD groups, and to assess RRBs (see below). To confirm inter-rater reliability, administrations were videotaped and a rater blind to group status coded five administrations per group. Inter-rater reliability was coded based on the method of the test authors and was high for both the algorithm and total items, at 86.7 and 85.7 %.

Cognitive abilities were measured using the *Wechsler Abbreviated Scale of Intelligence* (WASI; Wechsler 1999). The *Vineland Adaptive Behavior Scales* (Sparrow et al. 1985) is a parent report measure that was used to evaluate adaptive functioning in Communication, Daily Living Skills and Socialization.

RRBs were assessed using both direct observation and parent report measures. Direct observation of repetitive motor behaviors, ritualistic behaviors, self-injurious behaviors, unusual sensory interests, restricted interests and stereotyped behavior was collected using five items from the ADOS (Lord et al. 2000).

Parent report of RRBs was collected using parent responses to nine items assessing current RRBs and a history of RRBs on the *Autism Diagnostic Interview-Revised* (ADI-R, Lord et al. 1994). The ADI-R is a semi-structured parent interview used to assess current and past behaviors necessary for the diagnosis of ASD; if a behavior was present, severity was assessed. This measure was only administered to parents of participants in the OO and HFA groups. The "Restricted, Repetitive, and Stereotyped Patterns of Behavior" domain was assessed on the diagnostic

and current behavior algorithms. This RRBs domain consisted of four subdomains: encompassing preoccupations or circumscribed interests, compulsive adherence to non-functional routines or rituals, stereotyped and repetitive motor mannerisms, and preoccupations with parts of objects or nonfunctional elements of materials.

The Repetitive Behavior Scale-Revised (RBS-R; Bodfish et al. 2000) was also used to gather parent report of RRBs. The RBS-R is a parent report measure of a child's current repetitive behaviors, which are grouped in six domains: stereotyped, self-injurious, compulsive, ritualistic, sameness and restricted behaviors. The subscale scores are totaled to arrive at an overall score. Subscale inter-rater reliability ranges from 0.55 (sameness) to 0.78 (self-injurious; Bodfish and Lewis 2002).

The *Yale Special Interests Survey* (YSIS; Klin and Volkmar 1996) is a parent-report measure of circumscribed interests, special skills, and unusual attachments to objects. The questionnaire assesses special interests separately in four age periods (i.e., preschool, elementary, adolescence and adulthood). Because of the wide age range of the participants included in the study and the variability in the age of OO participants when OOs were achieved (i.e., some participants in the OO group achieved OO during the elementary period, while others were older), this instrument was used to assess the early history of RRBs during the preschool period only (ages 2–6). This period was chosen because the inclusion criteria of the study mandate that all participants in the OO group met diagnostic criteria for ASD during this period. Using the coding scheme described by Klin et al. (2007), circumscribed interests were coded by a blind rater into eight descriptive categories (facts/verbal memory and learning, facts and activities/visual memory and learning, sensory behaviors, math, classifying/ordering information, dates and times, collecting/hoarding, letters and numbers). Published inter-rater reliability for this coding scheme ranges from 0.81 to 1.0 (Klin et al. 2007). In addition, topics of circumscribed interests were coded as being unusual or developmentally appropriate. When questions arose about how to code interests, they were to be brought to the research group for consensus coding; however, this was never necessary because the interests reported clearly fit into one of the categories included in the coding scheme.

Results

Most scores did not meet the assumptions of normality required for parametric statistical analyses; therefore, nonparametric analyses were used. The Kruskal–Wallis test was conducted for each of the dependent variables of interest (selected measures of RRBs), with the three groups

designated as independent variables. When a statistically significant result was obtained on the Kruskal–Wallis Test, the Mann–Whitney U Test was used to determine which groups differed significantly. Mann–Whitney U Tests were conducted to determine whether the OO and HFA group differed on the domains of the ADI-R, which was not administered to the TD group. Follow-up Chi square tests were conducted for items on which the groups differed significantly. Chi square tests were also used to assess group differences on parent responses to the YSIS. One of the primary aims of this study was to examine even subtle group differences to explore how similar the OO group was as compared to the TD group with regard to RRBs. In order to detect even small group differences, we chose not to employ correction for multiple comparisons. In most studies, where there is a claim of group differences, a ‘conservative’ approach is to correct for multiple comparisons; since in the optimal outcome project, the general claim is of a lack of differences between the TD and OO groups, we interpreted the most conservative approach to be the one that preserved the greatest ability to find group differences.

To explore the effect that group differences in VIQ had on the dependent variables, the analyses reported below were repeated with VIQ included as a covariate; the significant findings reported below remained significant after controlling for VIQ. While the groups did not differ significantly on gender, more female participants were included in the OO group (21 %) than in the TD and HFA groups (9 %). To examine the effect of gender on the dependent variable, the primary analyses reported below were repeated with only the male participants; all of the significant group differences reported below remained significant. Lastly, because of the wide age range of the sample and the changes in the presentation of RRBs in individuals with ASD over time, primary analyses reported below were repeated with age included as a covariate; the significant findings remained.

Current Presentation of RRBs

Direct observation of RRBs using the ADOS revealed that the HFA group had significantly higher (more abnormal) scores than did the OO and TD groups (see Table 2). An item-by-item analysis revealed that the HFA group exhibited more unusual sensory interests, hand and finger mannerisms, and circumscribed interests than did participants in the OO and TD groups (see Table 2). No significant group differences were found on items assessing self-injurious behaviors, or compulsive or ritualistic behaviors. The TD and OO groups did not differ from each other on any RRB item of the ADOS.

Parent report of current RRBs was assessed using the ADI-R (for HFA and OO groups) and the RBS-R (for all groups). Parent report of current RRBs using the ADI-R included a group comparison of current scores on the subdomain and total domain scores of RRB’s in the current behavior algorithm. Mann–Whitney U Tests revealed that the HFA group had significantly higher mean scores than the OO group on each of the four subdomain scores and the domain total score (see Table 3). An item-by-item analysis revealed that the HFA group had significantly higher scores than the OO group on items assessing circumscribed interests, undue general sensitivity to noise, difficulties with minor changes in routine or personal environment, and complex mannerisms or stereotyped body movements (see Table 3). Chi square tests were conducted on these items to explore the distribution of responses, and indicated that significantly more HFA participants exhibited all four RRBs than did participants in the OO group (see Table 3). The item-by-item analysis also revealed that 24 % of the OO group exhibited mild circumscribed interests (i.e., exhibited special interests of an unusual degree, but that did not interfere with the participant’s other activities or family life; scored “1” on this item). In addition, 36 % of the OO group demonstrated a slight sensitivity to loud noises (i.e., score of “1”) and 15 % of the OO group displayed negative reactions to minor changes in routine that did not include significant distress or impairment in family life.

Parent report of the current presentation of RRBs using the subscales of the RBS-R, revealed that the HFA group scored significantly higher on all subscales of the RBS-R than the OO and TD groups (see Table 4). The only significant difference between the OO and TD groups was on the ritualistic behaviors subscale, with the OO group’s parents endorsing more items ($U = 249.50$, $z = -3.13$, $p = 0.002$, $r = 0.42$) and receiving higher subscale scores ($U = 247.50$, $z = -3.17$, $p = 0.002$, $r = 0.42$). These findings did not change when the groups were compared on the number of items endorsed within each subscale of the RBS-R rather than the total subscale score. An item-by-item analysis of the six items that comprise the ritualistic behaviors subscale (see Table 6 indicated that the OO group scored significantly higher than the TD group only on items assessing eating/mealtime rituals ($U = 255.00$, $z = -3.21$, $p = 0.001$, $r = 0.42$), as well as sleeping/bedtime rituals ($U = 330.00$, $z = -2.21$, $p = 0.03$, $r = 0.30$). Group means on this item indicate that, on average, these RRBs are mild and cause minimal interference on participant functioning (i.e., average score <1, and a rating of “1” is associated with mild behavior). No significant difference was found between the OO and HFA groups on these two items, but these groups differed significantly on the other four items that comprised the

Table 2 ADOS stereotyped behaviors and restricted interests domain

	TD	OO	HFA	Kruskal–Wallis χ^2	<i>p</i>	η^2	Mann–Whitney Post-hoc
N	34	34	45				
SBRI domain algorithm total	0.03 (0.17) (0–1)	0.18 (0.46) (0–2)	1.04 (1.17) (0–4)	32.44	<0.001	0.29	HFA > OO, TD
Unusual sensory interests	0 (0)	0.06 (0.24) (0–1)	0.27 (0.58) (0–2)	9.70	0.003	0.09	HFA > TD
None	100 %	94 %	80 %	10.43	0.03	0.22	HFA > TD
Mild	0 %	6 %	13 %				
Definite	0 %	0 %	7 %				
Hand and finger mannerisms	0 (0)	0.03 (0.17) (0–1)	0.22 (0.47) (0–2)	11.63	0.001	0.10	HFA > OO, TD
None	100 %	97 %	80 %	11.77	0.02	0.23	HFA > TD
Mild	0 %	3 %	18 %				
Definite	0 %	0 %	2 %				
Self-injurious behavior	0 (0)	0 (0)	0.02 (0.15) (0–1)	1.51	0.47	0.01	
Excessive interest in unusual or highly specific topic	0 (0)	0.09 (0.29) (0–1)	0.51 (0.79) (0–2)	18.40	<0.001	0.16	HFA > OO, TD
None	100 %	91 %	66 %	20.24	<0.001	0.30	HFA > OO, TD
Mild	0 %	9 %	16 %				
Definite	0 %	0 %	18 %				
Compulsions or rituals	0.03 (0.17) (0–1)	0 (0)	0.04 (0.21) (0–1)	1.48	0.48	0.01	

Table reports means, followed by SDs and ranges in parentheses of the total and item scores on the stereotyped behaviors and restricted interests (SBRI) domain of the ADOS. For significant findings, percentages of participants who exhibited mild (received a 1 on the ADOS item) or moderate to severe repetitive behavior (received a 2 or 3 on the ADOS item) are reported

ritualistic behavior subscale (see Table 5). No group differences were found between the TD and OO groups on subdomains assessing stereotyped behaviors, self-injurious behaviors, compulsive behaviors, sameness behaviors, or restricted behaviors.

In summary, when the current presentation of RRBs was examined using direct observation, the OO group scores were similar to those of the TD group, and they demonstrated significantly fewer RRBs than the HFA group. Specifically, the HFA group exhibited more unusual sensory interests, hand and finger mannerisms and circumscribed interests than did participants in the other groups. According to parent report, the OO and TD groups contained fewer participants who exhibited RRBs in all domains assessed than did the HFA group. Based on parent report, the OO and TD groups only differed significantly on ritualistic behaviors, specifically rituals around eating and

sleeping. Within the OO group, these RRBs did not interfere with functioning.

History of RRBs

Parent report exploring the history of RRBs was collected using the ADI-R (for OO and HFA groups) and the YSIS (for all groups). A frequency distribution was conducted to determine how many participants in the OO group exhibited some form of RRB early in development. This revealed that every child in the OO group exhibited some form of RRB, and 87.9 % of the OO sample scored at or above the ASD cutoff on the ADI-R RRBs domain, as compared to 97.7 % of the HFA group (see Table 6). Mann–Whitney U Tests were conducted on each of the four subdomain scores for past behavior that comprise the RRBs domain of the ADI-R Diagnostic Algorithm, as well

Table 3 ADI-R restricted, repetitive, and stereotyped patterns of behaviors domain—current rating

	OO	HFA	Mann–Whitney	Z	p	r
N	33	43				
<i>Current behavior algorithm</i>						
Encompassing preoccupation or circumscribed interest	0.39 (0.61) (0–2)	1.39 (0.95) (0–4)	296.00	–4.68	<0.001	0.54
Apparently compulsive adherence to nonfunctional routines or rituals	0.18 (0.46) (0–2)	0.80 (1.13) (0–4)	505.00	–2.78	0.005	0.32
Stereotyped and repetitive motor mannerisms	0.30 (0.53) (0–2)	0.70 (0.79) (0–2)	531.50	–2.29	0.02	0.26
Preoccupation with parts of objects of nonfunctional elements of materials	0.27 (0.45) (0–1)	0.61 (0.65) (0–2)	526.50	–2.37	0.02	0.27
Total	1.15 (1.44) (0–5)	3.50 (2.25) (0–9)	262.00	–4.85	<0.001	0.56
<i>ADI RRB items—current rating</i>						
Unusual preoccupations	0.09 (0.38) (0–2)	0.23 (0.52) (0–2)	640.00	–1.52	0.13	0.17
Circumscribed interests	0.30 (0.53) (0–2)	1.34 (1.03) (0–3)	299.50	–4.69	<0.001	0.53
None	73 %	22 %	22.11		<0.001	0.54
Mild	24 %	39 %				
Moderate/severe	3 %	39 %				
Repetitive use of objects or interest in parts of objects	0.03 (0.17) (0–1)	0.20 (0.55) (0–3)	632.00	–1.83	0.07	0.21
Compulsions/Rituals	0.15 (0.36) (0–1)	0.50 (0.88) (0–3)	604.00	–1.70	0.09	0.19
Unusual sensory interests	0.27 (0.45) (0–1)	0.52 (0.63) (0–2)	580.50	–1.77	0.08	0.20
Undue general sensitivity to noise	0.42 (0.56) (0–2)	0.84 (0.81) (0–3)	517.50	–2.37	0.02	0.27
None	61 %	36 %	5.95		0.05	0.28
Mild	36 %	48 %				
Moderate/severe	3 %	16 %				
Abnormal, idiosyncratic, negative responses to specific sensory stimuli	0.27 (0.52) (0–2)	0.52 (0.73) (0–3)	597.50	–1.59	0.11	0.18

Table 3 continued

	OO	HFA	Mann–Whitney	Z	p	r
Difficulties with minor changes in own routines or personal environment	0.21 (0.48) (0–2)	0.68 (0.86) (0–3)	500.00	–2.76	0.01	0.31
None	82 %	52 %	7.67		0.02	0.32
Mild	15 %	32 %				
Moderate/severe	3 %	16 %				
Resistance to trivial changes in the environment	0.09 (0.29) (0–1)	0.27 (0.59) (0–2)	639.00	–1.42	0.16	0.16
Unusual attachment to objects	0.06 (0.24) (0–1)	0.23 (0.53) (0–2)	601.00	–1.67	0.10	0.19
Hand and finger mannerisms	0.21 (0.42) (0–1)	0.57 (0.87) (0–3)	591.50	–1.72	0.08	0.20
Other complex mannerisms or stereotyped body movements	0.09 (0.38) (0–2)	0.41 (0.76) (0–3)	558.00	–2.51	0.01	0.29
None	94 %	70 %	6.93		0.03	0.30
Mild	3 %	23 %				
Moderate/severe	3 %	7 %				

Table reports means, followed by SDs and ranges in parentheses. For significant findings on individual ADI items, percentages of participants who exhibited mildly repetitive behavior (no impairment in functioning, received “1” on the ADI item) or moderate to severe repetitive behavior (received a 2 or 3 on the ADI item) are reported

as the domain total score. No significant group differences were found between the HFA and OO groups on the RRB domain total score for past behavior or on any of the RRB subdomain scores (see Table 6).

An item-by-item analysis of RRB items included on the ADI-R was conducted using Mann–Whitney U Tests to determine whether subdomain scores may be masking significant group differences on specific types of past RRBs. This analysis revealed that the HFA group showed significantly higher (more severe) mean scores on circumscribed interests, undue general sensitivity to noise, and difficulties with minor changes in the child’s routine or personal environment, (see Table 6). The groups did not differ on unusual preoccupations, repetitive use of objects, ritualistic behavior, unusual sensory interests, abnormal responses to specific sensory stimuli, resistance to trivial changes in the environment, unusual attachment to objects, hand and finger mannerisms, stereotyped body movements and midline hand movements. Follow-up Chi square tests were conducted for the three items on which the groups differed significantly to examine how many participants in each group showed these symptoms. The results revealed that the HFA group

contained significantly more participants who exhibited undue sensitivity to noise and difficulties with minor changes in routine, but not circumscribed interests, than the OO group (see Table 6). This difference in results suggests that equal numbers of OO and HFA participants had difficulty with circumscribed interests, but that the interests of the HFA group were more severe.

Parents were also asked to fill out the YSIS, which asked about the presence of attachment to unusual objects and circumscribed interests during the participants’ preschool years, as well as the extent of participants’ time involved in their special interests during this time. Chi square tests revealed that the OO and HFA groups did not differ from each other, and both groups showed more attachment to unusual objects, circumscribed interests and unusual interests during the preschool period than the TD group (see Table 7). Among those participants who exhibited a circumscribed interest during the preschool years, the HFA and OO groups did not differ on the frequency with which participants discussed their circumscribed interests with family members, other adults, or peers, or the amount of free time participants spent on the circumscribed interest (see Table 7).

Table 4 Repetitive behavior scale-revised (RBS)

	TD	OO	HFA	Kruskal–Wallis χ^2	<i>p</i>	η^2	Mann–Whitney Post-hoc
<i>Total subscale score</i>							
N	31	26	36				
Stereotyped behaviors	0.10 (0.30) (0–1) 10 %	0.50 (1.07) (0–4) 23 %	2.84 (2.84) (0–10) 81 %	41.79	<0.001	0.45	HFA > OO, TD
Self-injurious behaviors	0.03 (0.18) (0–1) 3 %	0.15 (0.37) (0–1) 15 %	1.05 (1.53) (0–6) 42 %	16.53	<0.001	0.18	HFA > OO, TD
Compulsive behaviors	0.26 (0.82) (0–4) 13 %	0.54 (1.24) (0–5) 27 %	2.49 (3.23) (0–12) 61 %	21.55	<0.001	0.23	HFA > OO, TD
N	30	26	36				
Ritualistic behaviors	0.17 (0.75) (0–4) 7 %	1.42 (2.00) (0–6) 42 %	4.08 (4.13) (0–14) 69 %	29.24	<0.001	0.32	HFA > OO > TD
Sameness behaviors	0.23 (0.73) (0–3) 10 %	0.92 (1.70) (0–6) 31 %	5.73 (5.94) (0–25) 81 %	40.12	<0.001	0.44	HFA > OO, TD
Restricted behaviors	0.10 (0.31) (0–1) 10 %	0.42 (0.76) (0–2) 27 %	2.41 (2.19) (0–8) 86 %	46.07	<0.001	0.51	HFA > OO, TD

Table reports means, followed by SDs and ranges in parentheses, as well as the percentage of participants who exhibited any degree of repetitive behaviors (received a score of 1 or higher on any item within the subscale)

To explore group differences in the content of circumscribed interests, the interests were coded into eight categories using the coding scheme described by Klin et al. (2007): facts/verbal memory and learning, facts and activities/visual memory and learning, sensory behaviors, math, classifying/ordering information, dates and times, collecting/hoarding, and letters and numbers. Chi square tests (see Fig. 2) revealed that facts learned through verbal and visual memory, and sensory behaviors were the most common circumscribed interests, and that only for facts learned through verbal memory did the HFA-OO difference approach significance.

To summarize the history findings, all OO participants exhibited RRBs and most (87.9 %) exhibited enough RRBs early in development to meet the RRB criteria for ASD as measured by the ADI-R. Furthermore, when general categories of RRBs were examined in early childhood, no difference was noted between the OO and HFA groups in

the severity of RRBs or the proportion of participants whose parents endorsed a history of RRBs. Differences in early RRBs were noted between the OO and HFA groups on only one of two measures used (i.e., ADI-R, not YSIS), and only when the severity of single items assessing specific RRBs were examined. The OO group received lower scores on circumscribed interests, undue general sensitivity to noise and difficulty with minor changes in routine. When compared to the TD group, significantly more participants in the OO and HFA groups exhibited attachment to unusual objects, circumscribed interests and unusual circumscribed interests during the preschool period.

Discussion

This study examined the presence of residual RRBs and the history of RRBs among individuals who were diagnosed

Table 5 Repetitive behavior scale-revised (RBS): ritualistic behavior subscale

	TD	OO	HFA	Kruskal–Wallis χ^2	<i>p</i>	η^2	Mann–Whitney post-hoc
N	30	26	36				
Eating/mealtime	0.07 (0.37) (0–2)	0.62 (0.85) (0–2)	0.92 (1.12) (0–3)	16.06	<0.001	0.17	HFA, OO > TD
None	97 %	62 %	51 %	16.84	0.002	0.35	HFA, OO > TD
Mild	0 %	15 %	19 %				
Moderate/severe	3 %	23 %	30 %				
Sleeping/bedtime	0 (0) –	0.27 (0.72) (0–3)	0.50 (0.88) (0–3)	10.93	0.004	0.12	HFA, OO > TD
None	100 %	84 %	69 %	11.25	0.02	0.25	HFA > TD
Mild	0 %	8 %	17 %				
Moderate/severe	0 %	8 %	14 %				
Self-care—bathroom and dressing	0.03 (0.18) (0–1)	0.08 (0.27) (0–1)	0.49 (0.80) (0–3)	14.22	0.001	0.15	HFA > OO, TD
None	97 %	92 %	65 %	14.63	0.006	0.28	HFA > OO, TD
Mild	3 %	8 %	27 %				
Moderate/severe	0 %	0 %	8 %				
Travel/transportation	0 (0) –	0.04 (0.20) (0–1)	0.44 (0.84) (0–3)	14.24	0.001	0.16	HFA > OO, TD
None	100 %	97 %	72 %	14.58	0.006	0.28	HFA > OO, TD
Mild	0 %	3 %	17 %				
Moderate/severe	0 %	0 %	11 %				
Play/leisure	0.03 (0.18) (0–1)	0.19 (0.49) (0–2)	0.73 (0.90) (0–3)	20.21	<0.001	0.22	HFA > OO, TD
None	97 %	84 %	51 %	20.41	<0.001	0.33	HFA > OO, TD
Mild	3 %	12 %	30 %				
Moderate/severe	0 %	4 %	19 %				
Communication/social interaction	0.03 (0.18) (0–1)	0.23 (0.51) (0–2)	1.06 (1.07) (0–3)	27.27	<0.001	0.30	HFA > OO, TD
None	97 %	21 %	42 %	20.24	<0.001	0.40	HFA > OO, TD
Mild	3 %	15 %	22 %				
Moderate/severe	0 %	4 %	36 %				

Table reports means, followed by SDs and ranges in parentheses, as well as the percentage of participants who exhibited mild (received a 1 on the RBS item) or moderate to severe repetitive behavior (received a 2 or 3 on the RBS item)

with ASDs in early childhood, but who no longer meet diagnostic criteria for any ASD. History and current presentation of RRBs in the OO group was compared to RRBs in same aged high-functioning individuals with ASD and TD peers.

Results showed that at the time of this study, individuals who achieved OOs exhibited similarly low rates of RRBs compared to TD peers. The only difference between the OO and TD groups was in more ritualistic behaviors

around mealtime and bedtime in the OO group. The majority of participants in the OO group did not exhibit ritualistic behaviors around mealtime and bedtime, and of those that did (38 % for mealtime rituals and 16 % for bedtime rituals), none were reported to cause a severe impairment on functioning. Ritualism in daily routines is commonly observed among typically developing children and young adults, and may reduce anxiety by familiarizing the individuals with the sequences of actions during daily

Table 6 ADI-R restricted, repetitive, and stereotyped patterns of behaviors domain—ever rating

	OO	HFA	Mann–Whitney	Z	p	r
N	33	43				
<i>Diagnostic algorithm (ever rating)</i>						
Encompassing preoccupation or circumscribed interest	1.61 (1.20) (0–4)	1.91 (0.96) (0–4)	611.00	–1.25	0.21	0.14
Apparently compulsive adherence to nonfunctional routines or rituals	1.09 (1.16) (0–4)	1.36 (1.26) (0–4)	647.00	–0.86	0.39	0.10
Stereotyped and repetitive motor mannerisms	1.55 (0.71) (0–2)	1.43 (0.82) (0–2)	686.50	–0.48	0.63	0.06
Preoccupation with parts of objects of nonfunctional elements of materials	1.61 (0.70) (0–2)	1.50 (0.73) (0–2)	665.00	–0.76	0.45	0.09
Total	5.85 (2.33) (1–9)	6.20 (2.28) (1–12)	697.50	–0.30	0.77	0.03
<i>ADI RRB items—history of symptoms (ever rating)</i>						
Unusual preoccupations	0.45 (0.83) (0–3)	0.52 (0.88) (0–3)	695.00	–0.40	0.69	0.05
Circumscribed interests	1.27 (0.94) (0–3)	1.76 (0.96) (0–3)	505.00	–2.09	0.04	0.24
None	25 %	9 %	3.57		0.17	0.22
Mild	33 %	31 %				
Moderate/severe	42 %	60 %				
Repetitive use of objects or interest in parts of objects	1.85 (1.18) (0–3)	1.61 (1.13) (0–3)	635.50	–0.97	0.33	0.11
Compulsions/rituals	0.67 (0.99) (0–3)	0.73 (0.95) (0–3)	683.00	–0.50	0.62	0.06
Unusual sensory interests	0.94 (0.79) (0–2)	0.80 (0.70) (0–2)	655.00	–0.79	0.43	0.09
Undue general sensitivity to noise	1.15 (1.00) (0–3)	1.75 (1.08) (0–3)	497.00	–2.46	0.01	0.28
None	34 %	21 %	6.63		0.04	0.29
Mild	27 %	11 %				
Moderate/severe	39 %	68 %				
Abnormal, idiosyncratic, negative responses to specific sensory stimuli	0.70 (0.92) (0–3)	0.98 (1.07) (0–3)	626.00	–1.11	0.27	0.13

Table 6 continued

	OO	HFA	Mann–Whitney	Z	p	r
Difficulties with minor changes in subject's own routines or personal environment	0.61 (0.90) (0–3)	1.39 (1.17) (0–3)	449.50	−3.02	0.003	0.34
None	64 %	32 %	8.14		0.02	0.33
Mild	15 %	20 %				
Moderate/severe	21 %	48 %				
Resistance to trivial changes in the environment	0.33 (0.82) (0–3)	0.73 (1.02) (0–3)	574.50	−1.93	0.05	0.22
Unusual attachment to objects	0.79 (0.98) (0–3)	0.78 (0.95) (0–2)	751.00	−0.15	0.88	0.02
Hand and finger mannerisms	1.27 (0.88) (0–3)	1.20 (1.00) (0–3)	698.50	−0.30	0.76	0.03
Other complex mannerisms or stereotyped body movements	0.85 (1.00) (0–3)	0.93 (1.04) (0–3)	695.00	−0.35	0.73	0.04

Table reports means, followed by SDs and ranges in parentheses. Scores above 2 on the RRB Diagnostic Algorithm Total score meet the ASD cutoff on the ADI. For significant findings on individual ADI items, percentages of participants who exhibited mildly repetitive behavior (no impairment in functioning, received “1” on the ADI item) or moderate to severe repetitive behavior (received a 2 or 3 on the ADI item) are reported

Bold values indicate statistically significant group differences

activities (Glenn et al. 2012). However, we found that few individuals in our TD sample exhibited these behaviors. It remains possible that these residual ritualistic behaviors were not targeted in intervention with the OO group to the same degree as other RRBs because these behaviors did not interfere with functioning and may have served an adaptive purpose. Overall, the OO group described in this study appears to be different from previously described cases of children with ASD who achieved good remission in social and communication impairments, but who continued to exhibit symptoms of ASD within the RRBs domain that cause impairment in functioning (e.g., Piven et al. 1996; Seltzer et al. 2004). RRBs have been remediated along with symptoms within the social and communication cluster in this group of individuals who achieved OOs. These findings raise important questions about the intervention history of the OO and HFA group, which has been described by Orinstein et al. (2014) in a separate report.

As predicted, the HFA group demonstrated more frequent and more severe RRBs than the OO and TD groups across most RRB domains assessed. In accordance with previous reports, these findings suggest that of the core clusters of ASD symptomatology, the RRB cluster appears to be particularly persistent over the course of development in our sample of high-functioning individuals with ASD.

It is also important to consider what the results suggest about the history of ASD symptomatology within this OO sample. These results demonstrate that all participants in the OO group exhibited at least one type of RRB in early development, and 88 % of the OO group displayed enough RRBs to meet the ASD cutoff of the RRB domain on the ADI-R, a gold standard measure used in the diagnosis of ASD (Lord et al. 1994). Generally, the early presentation of RRBs was also similar across the HFA and OO groups. The results of this study lend support to the assertion that these individuals were accurately diagnosed with an ASD in early childhood.

Differences in the history of RRBs were found only in the severity of oversensitivity to noise and insistence on sameness, where individuals in the OO group exhibited milder symptoms. A number of studies have proposed that RRBs interfere with the child's ability to attend to the external environment where learning opportunities are present and, consequently, may impede learning (Bodfish et al. 2000; Bopp et al. 2009; Lewis 2004). It is possible that oversensitivity to noise and insistence on sameness may interfere with learning of social skills and communication abilities (Helt et al. 2008). For instance, oversensitivity to noise may result in avoidance of auditory input, which may limit the amount of linguistic input a child

Table 7 Yale special interest survey (YSIS): preschool period

	TD	OO	HFA	χ^2	<i>p</i>	Cramer's V	Post-hoc
Attachment to unusual objects	0 % (N = 16)	61 % (N = 18)	45 % (N = 29)	14.45	0.001	0.48	OO, HFA > TD
Circumscribed interests	4 % (N = 27)	71 % (N = 21)	88 % (N = 34)	46.36	<0.001	0.75	OO, HFA > TD
Unusual circumscribed interests	0 % (N = 27)	19 % (N = 21)	24 % (N = 34)	7.11	0.03	0.29	OO, HFA > TD
<i>Conversations about circumscribed interests</i>							
With adults	Sometimes	–	62 %	27 %	4.58	0.10	0.34
	Quite a bit	–	15 %	38 %			
	Almost always	–	23 %	35 %			
			(N = 13)	(N = 26)			
With family	Sometimes	–	23 %	4 %	3.61	0.17	0.30
	Quite a bit	–	54 %	73 %			
	Almost always	–	23 %	23 %			
			(N = 13)	(N = 26)			
With peers	Sometimes	–	46 %	13 %	4.87	0.09	0.37
	Quite a bit	–	31 %	52 %			
	Almost always	–	23 %	35 %			
			(N = 13)	(N = 23)			
Free time spent on interest	Sometimes	–	8 %	7 %	0.92	0.63	0.15
	Quite a bit	–	62 %	46 %			
	Almost always	–	31 %	46 %			
			(N = 13)	(N = 28)			

Table reports the percentage of participants who exhibited each behavior and the total number of participants in each cell

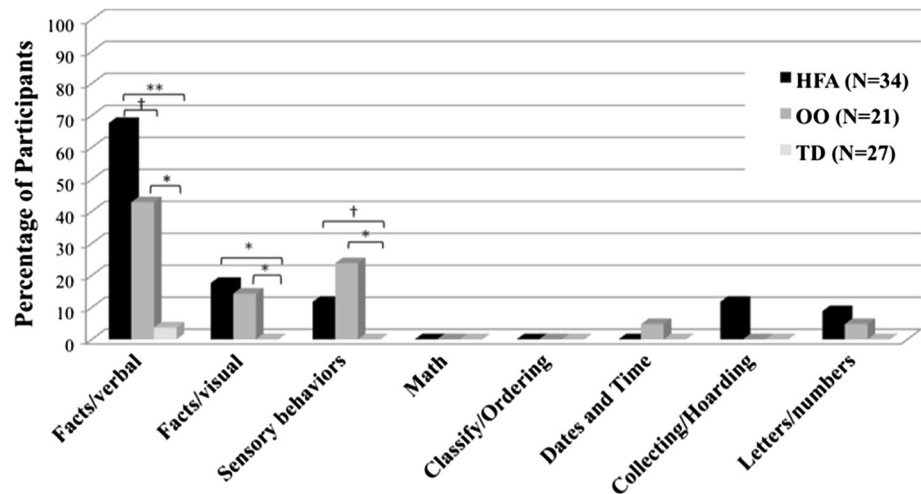
processes. A decrease in linguistic input may interfere with the child's exposure to prototypical social communication and hinder the development of this skill. Similarly, insistence on sameness may limit the types of activities in which a child may participate, which may limit the variety of linguistic input to which the child is exposed and hinder learning within the social communication domain.

Several important limitations should be considered when drawing conclusions about this study. The results of this study are limited by a homogeneous sample in terms of functioning level, ethnicity and socioeconomic status. Consequently, the generalizability of the current findings to a broader autism population may be limited. The size of the sample also limited the study's power to detect small group differences. Finally, the current study is cross-sectional and cannot shed light on whether the presentation of RRBs among individuals with OO will change over time. Future studies using a longitudinal design could confirm that RRBs subside in the OO group and explore the timing of this change in the presentation of RRBs. Finding that these behaviors subside first would lend support to the argument that the absence of RRBs may make a child more available

for learning of social and communication skills. Such a finding might also suggest that intervention of ASD should focus on RRBs early in treatment in order to allow unimpeded learning of other skills.

The current study is also limited by the fact that parent report was used to assess history of RRBs and current presentation of RRBs for this study. It is well established that parent report introduces a risk of bias, particularly when parents are asked to recall past behavior (Bradburn et al. 1987; Henry et al. 1994; Robbins 1963). In the current study, the participant's functioning at the time of participation may have biased parent report of the child's history of RRBs. Two of the measures used to assess RRBs in this study were also not designed to measure RRB severity (i.e., ADOS, ADI-R). These measures were supplemented by several parent report measures of RRBs and with a direct observation of RRBs. However, the observational measure of RRBs depended on a relatively brief observation period. Given that these behaviors were not commonly observed in the OO and HFA groups, including a time-limited assessment may have resulted in the evaluators failing to observe some RRBs that could have been

Fig. 2 Content of circumscribed interest—preschool period. *Note* Percentage of participants who exhibited each type of circumscribed interest. † $p < 0.10$. * $p < 0.05$. ** $p < 0.001$



captured during a longer observation. It remains possible that even with a longer observation period, some RRBs may only be captured by parent report as children may be less likely to engage in these behaviors in novel settings and with unfamiliar adults.

Future studies would benefit from a prospective study design and direct observation of RRBs across development that would help eliminate bias related to parent report and allow for a standardized assessment of the initial ASD diagnosis in the OO group. However, without being able to predict which children are likely to experience an OO, conducting a prospective study of this group would involve a long-term follow-up of a prohibitively large sample of children with ASD.

It is important to consider that the OO group's initial ASD diagnoses were confirmed using the diagnostic criteria published in the DSM-IV-TR (APA 2000) and it is possible that some individuals within this OO group would not have met diagnostic criteria for ASD if the recently published DSM-5 (APA 2013) criteria were used. In addition, it would have been preferable for evaluators who confirmed current diagnosis and assessed functioning of participants to be blind to the history and presumed group membership of participants. However, it was not feasible for the examiner to remain blind because the evaluation contained measures of ASD symptomatology (e.g., ADI-R, ADOS) that elicited report about diagnostic history and current functioning. To address this limitation, a blind rater reviewed a subset of the video recordings of the ADOS and high inter-reliability was established.

In summary, the results of this study suggest that individuals who were diagnosed with ASD in early childhood, but who no longer meet diagnostic criteria for any ASD, exhibit minimal residual RRBs and appear very similar to TD peers in this symptom domain. It remains for future studies to determine how central to symptom remission the

reduction of early RRBs was for these individuals, and whether these behaviors were particularly targeted in intervention, or were naturally abated in individuals who achieved OO.

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