

The Development of Restricted and Repetitive Behaviors and Interests in Children with
Autism Spectrum Disorders

by

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In loving memory of my mother

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Abstract

The Development of Restricted and Repetitive Behaviors and Interests in Children with Autism Spectrum Disorders

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The development of restricted and repetitive behaviors and interests (RRBs) in Autism Spectrum Disorders (ASD) is examined in a series of three studies. In all studies, RRBs were assessed using the Autism Diagnostic Interview – Revised (ADI-R), a parent interview.

The first study considers RRBs in young children with ASD. Most RRBs were significantly more common in children with ASD than in children with nonspectrum developmental disorders (DD) and typical development (TD). A factor analysis supported the notion of RRB subtypes, one comprised of ‘repetitive sensorimotor’ (RSM) behaviors and the other of ‘insistence on sameness’ (IS) behaviors. Having several RSM behaviors to a severe degree distinguished children with ASD from children with DD. IS behaviors were relatively uncommon in ASD at this age.

The second paper explores how RRBs change in children with ASD over time. For children with ASD, total RSM scores tended to remain relatively high over time, indicating consistent severity, whereas total IS scores started low and increased over time, indicating worsening. For the RSM behaviors, having a higher NVIQ and milder ASD were associated with improvement. NVIQ was not associated with IS trajectories, but milder social impairment was associated with more severe trajectories of IS scores, supporting the idea that these are ‘higher order’ behaviors.

Finally, the third paper examines the stability of RRBs over time. Scores on the RSM items tended to remain high over time, particularly for children with autism and/or lower NVIQ scores. Children who did not have RSM behaviors at a young age tended to acquire them over time. Conversely, scores on IS behaviors increased over time. Compared to the RSM behaviors, children who had IS behaviors at one point in development were not as likely to maintain them, but children who did not have these behaviors were more likely to continue not to have them. Children who gained behaviors in one subtype were the most likely to gain behaviors in the other.

The findings from this series of studies are discussed in terms of their implications for our understanding of the etiology and treatment of RRBs in ASD.

Chapter I

Introduction

Autism spectrum disorders (ASD)¹ are characterized by impairments in the areas of reciprocal social interaction and communication, and the presence of restricted and repetitive behaviors and interests (RRBs). When Leo Kanner (1943) first described autistic symptoms in 11 children over 60 years ago, he noted many unusual behaviors that are remarkably similar to those described by clinicians today, and that are now thought of as RRBs. These included stereotyped motor mannerisms (e.g., shaking head from side to side, jumping up and down repeatedly); repetitive use of objects (e.g., spinning round objects, pulling the blinds up and down) preoccupation with unusual objects (e.g. cardboard boxes); all-encompassing interests (e.g. trains); unusual sensory interests and aversions (e.g., mouthing objects, reacting with distress to the sound of a streetcar); and an insistence that things be ‘just so’ (e.g. becoming upset if a toy was missing a part or if the furniture was rearranged). As this list indicates, the category of RRBs encompasses a wide range of behaviors. The common feature is the presence of an atypical behavior or interest, whereas the symptoms that fall into the social and communication domains are generally characterized by the absence of a behavior normally seen in children with typical development (e.g. eye contact, social reciprocity).

Despite the fact that RRBs are considered a core feature of ASD, they have received far less attention than the domains of social interaction and communication. This might be because lack of social reciprocity is thought of as ‘fundamental’ to the disorder, whereas RRBs, at least recently, have been less of a focus of theoretical inquiry, even being proposed as by-products of the core deficits of ASD (Baron-Cohen, Tager-

¹ Henceforth, the term ‘autism spectrum disorders’ will be used as an umbrella term, encompassing Autistic Disorder, Pervasive Developmental Disorder – Not Otherwise Specified (PDD-NOS), and Asperger’s Disorder, as defined by DSM-IV criteria. The term ‘autism’ will be used to refer to more narrowly defined Autistic Disorder.

Flusberg, & Cohen, 2000). However, we know that RRBs cannot be fully accounted for by social and communication impairments. Children with disorders that involve social and/or language difficulties, such as Social Anxiety and Specific Language Impairment, do not generally exhibit RRBs. Conversely, children with disorders that do not primarily affect social and communication skills (e.g. Obsessive Compulsive Disorder, Tourette Syndrome) display some of these behaviors. Social and communicative ability can also be teased apart from RRBs within ASD. Children with high-functioning autism or Asperger's Disorder, who often have relatively mild social impairments and fluent language, can nevertheless have RRBs that cause significant impairment (Szatmari, Bryson, Boyle, Streiner, & Duku, 2003; Walker et al., 2004). Furthermore, RRBs are considered widespread enough in ASD that they are part of the diagnostic criteria for Autistic Disorder according to the *Diagnostic and Statistical Manual for Mental Disorders, 4th Edition* (DSM-IV: American Psychiatric Association, 1994). A thorough understanding of these behaviors is therefore essential to our understanding of the disorder.

Practically speaking, RRBs are important to understand because of the degree to which they interfere with all aspects functioning in children with ASD, such as their ability to learn from and attend to the world around them. RRBs also interfere with social and communicative development. A child who focuses his or attention on spinning objects cannot receive the input necessary for normal social development. RRBs therefore have cascading effects, in that they add to the social and communicative impairment already present in ASD. These behaviors also interfere with family functioning and are cited among the most stressful behaviors for parents (Bishop et al., under review.)

Particularly little is known about how RRBs change in children with ASD over time. This represents a significant gap in our understanding of ASD, a disorder in which symptoms not only affect development but are affected *by* development. RRBs in very young children with ASD have received little attention. With an increasing emphasis on early identification and treatment of ASD, knowing which behaviors are 'red flags' for the disorder has become essential. Identifying the kinds of RRBs that children with ASD

exhibit early in life, at the same time that significant changes are taking place in brain development and other symptoms of the disorder are emerging, might also help us pinpoint the causes of the disorder. Furthermore, RRBs are one of the strongest predictors of diagnostic stability (Lord et al., 2006) and can therefore help clinicians make prognoses. We also need to understand how these behaviors evolve over time. Do children who have RRBs at young ages continue to have them when they get older? Do these behaviors tend to become worse or better over time, or do they stay the same? The answers to these questions are, needless to say, important to parents of children with ASD who want to have some sense of what to expect as their children get older.

The following studies are intended to provide a comprehensive picture of the development of RRBs in children with ASD. Chapter II, *Restricted and Repetitive Behaviors and Interests in Young Children with ASD*, describes RRBs early in development in ASD, when they are first emerging. It also explores the question of whether the category of RRBs should be broken down into subtypes. Chapter III, *Developmental Trajectories of Restricted and Repetitive Behaviors and Interests in Children with Autism Spectrum Disorders: Further Evidence of Repetitive Behavior Subdomains*, examines the variables that predict patterns of change in RRBs, and describes the different trajectories observed in children with ASD. In this paper, RRBs are considered both as a category and as part of subtypes. Finally, Chapter IV, *The Stability of Restricted and Repetitive Behaviors and Interests in Children with ASD*, examines how persistent the individual behaviors that comprise these subtypes are over time, both in terms of their presence as well as the degree of impairment they cause. Here, the focus is on identifying patterns of change in individual behaviors.

References

- American Psychiatric Association. (1994). *Diagnostic and Statistical Manual of Mental Disorders* (4th ed.). Washington DC: American Psychiatric Association.
- Baron-Cohen, S., Tager-Flusberg, H. & Cohen, D. Understanding Other Minds: Perspectives from Autism and Developmental Cognitive Neuroscience (Oxford Univ.Press, Oxford, 2000).
- Bishop, Richler, & Lord.C. Predictors of perceived negative impact in mothers of children with autism spectrum disorders. *American Journal on Mental Retardation*.
- Kanner, L. (1943). Autistic disturbances of affective contact. *Nervous Child*, 2, 217-250.
- Lord, C. Risi, S. DiLavore, P.S., Shulman, C., Thurm, A. & Pickles, A. (2006). Autism from 2 to 9. *Archives of General Psychiatry*, 63, 694-701.
- Szatmari, P., Bryson, S. E., Boyle, M. H., Streiner, D. L., & Duku, E. (2003). Predictors of outcome among high functioning children with autism and Asperger syndrome. *Journal of Child Psychology and Psychiatry*, 44, 520-528.
- Walker, D. R., Thompson, A., Zwaigenbaum, L., Goldberg, J., Bryson, S. E., Mahoney, W. J., Strawbridge, C. P., & Szatmari, P. (2004). Specifying PDD-NOS: A comparison of PDD-NOS, Asperger syndrome, and autism. *Journal of the American Academy of Child and Adolescent Psychiatry*, 43, 172-180.

Chapter II

Restricted and Repetitive Behaviors and Interests in Young Children with Autism Spectrum Disorders

Background and Significance

A hallmark of early childhood is a desire for repetition. It is common for a child to ask for the same book 4 or 5 times a day or for her parents to say the same phrase to her every night at bedtime. In a study of typically developing children, Evans et al. (1997) found that these behaviors were particularly common between the ages of 2 and 4.

Repetitive behaviors are also common for many individuals with developmental or psychiatric disorders. Such behaviors have been identified in children and adolescents with obsessive-compulsive disorder, schizophrenia, Tourette syndrome, attention deficit hyperactivity disorder, and mood-anxiety disorders (Mahone, Bridges, Prahme, & Singer, 2004). In a study of adults with mental retardation, Bodfish, Symons, Parker, and Lewis (2000) found a substantial proportion of the sample engaged in behaviors such as self-injury, compulsions, and stereotypies.

Despite the fact that the behaviors described above are seen in many different disorders and in early typical development, they are considered a core feature of autism and are seen in nearly all children with autism spectrum disorders (ASD). Restricted and repetitive behaviors and interests (RRBs) comprise one of the domains of behavior required for a diagnosis of autism according to the *Diagnostic and Statistical Manual of Mental Disorders* (American Psychiatric Association, 1994).

At first glance, some RRBs overlap with the 'Just Right' behaviors of typical toddlers and the repetitive or compulsive behaviors of children with nonspectrum disorders. Given that RRBs are considered a central feature of autism, it is important to ask which aspects of these behaviors are particular to children with autism and related disorders. Evidence from the study by Bodfish et al. (2000) suggests that RRBs may be

more common and severe in individuals with autism compared to those with nonspectrum disorders. It is not clear, however, if this is the case in very young children with ASD.

Part of the problem is that we still do not have a clear understanding of RRBs in very young children with ASD. Cox et al. (1999) found that few children exhibited clear RRBs at 20 months, according to parent report on the Autism Diagnostic Interview-Revised (Lord et al., 1994) and Stone et al. (1999) found that restricted and repetitive behaviors were less frequently endorsed by clinicians in children with autism at ages 2 and 3 than impairments in the social and communication domains. In a longitudinal study of children with ASD, Charman et al. (2005) found that at age 2, a substantial minority of children did not meet the RRB algorithm cutoff of 3 points on the ADI-R. Baranek (1999) conducted a retrospective study using home videos to identify predictors of ASD in even younger children, between 9 and 12 months of age. Findings indicated low prevalence for object and motor stereotypies in children with ASD at this age, at least as recorded on video by their families. However, in a prospective, longitudinal study of infant siblings of children with ASD using the Autism Observation Scale for Infants (Bryson, McDermott, Rombough, & Zwaigenbaum, in press), Zwaigenbaum et al. (2005) found that sensory-oriented behaviors (e.g. rubbing hands on table) at 12 months predicted a diagnosis of autism at 24 months.

The prevalence of RRBs in young children with ASD may depend on the specific behavior. Moore and Goodson (2003) found that, in their sample of 2-year-olds, children with ASD received higher ADI-R scores (indicating a greater degree of impairment) on what are thought of as 'lower order' RRB items, such as hand mannerisms, repetitive use of objects and unusual sensory interests, than on 'higher order' RRBs, such as compulsions and rituals and unusual preoccupations. Similarly, in a longitudinal study of children referred for possible autism, Lord (1995) found that 87.5% of children diagnosed with autism at age 3 were reported by their parents to have exhibited hand and finger mannerisms and unusual sensory interests at the age of 2. In a recent study, Lord et al. (2006) found that RRB domain scores at the age of 2 significantly predicted both autism (vs. PDD-NOS) and ASD at the age of 9. However, this study did not report which behaviors were most strongly associated with autism or ASD.

Taken together, these findings suggest that while, in general, social and communicative impairments might be more apparent than RRBs in very young children with ASD, some RRBs may be clearly manifested as young as age 2. Therefore, it is necessary to examine individual RRBs in very young children with ASD, in addition to considering the RRB category as a whole.

It is also important to consider the prevalence of individual RRBs. Mean scores on ADI-R RRB items do not indicate how common these behaviors are. The study by Lord (1995) was one of the only ones to look at prevalence of RRB in young children, and in this study, children were only included in the autism sample if they were deemed likely to meet criteria from the International Classification of Diseases – 10th Edition (ICD-10: see World Health Organization, 1992) for autism at age 5, a relatively stringent criterion. It is important to determine the rates of different RRBs in a sample of children with more broadly defined ASD, in order to obtain a clearer picture of RRBs in children across a range of ability. Prevalence should also be compared in subgroups of children with ASD. Walker et al. (2004) found that overall RRB scores were higher for children with autism than those with PDD-NOS, but no studies have examined whether this trend exists in very young children.

Another important issue that has not been resolved is which RRBs are particular to children with ASD at young ages. In order to address this question, it is necessary to determine how common RRBs are in children with ASD, as opposed to children who are not on the autism spectrum. Lord (1995) compared the prevalence of RRBs in children who were deemed likely to have autism at age 5 to those deemed unlikely to receive the diagnosis. It is possible that some of the children in the ‘not autism’ group *were* on the autism spectrum, but simply did not meet the strict criterion for a diagnosis of autism. Clearer differences in RRB prevalence might have observed if children with ASD had been compared to a sample of children who are delayed, but not on the autism spectrum.

Similarly, although RRBs have been examined in typically developing children, no study to date has compared prevalence of RRBs in very young children with typical development and children with ASD. As a result, we still do not have a clear sense of which behaviors can be expected in young children with typical development and which behaviors are clearly atypical.

The prevalence and severity of a given RRB, and how these vary according to diagnosis, might also be associated with the category into which the behavior falls. Several studies have found evidence for two factors: a ‘repetitive sensorimotor’ (RSM) factor, comprised of behaviors such as hand/finger and complex body mannerisms, repetitive use of objects, and unusual sensory interests; and an ‘insistence on sameness’ (IS) factor, comprised of behaviors such as compulsions and rituals, difficulties with changes in routine, and resistance to trivial changes in the environment (Cuccaro et al., 2003; Bishop, Richler, & Lord, 2006; Szatmari et al., 2006; Hus, Pickles, Cook, Risi, & Lord, 2007). Findings from the study by Bishop et al. suggest that RSM behaviors may be more common than IS behaviors in very young children with ASD. To date, however, there has not been a study combining a factor analysis of RRBs in young children with ASD with a detailed examination of RRBs in these children. This would make it possible to determine whether behaviors that cluster together show similar patterns of prevalence and severity in young children with ASD.

Although it is important to consider the prevalence of individual RRBs, it is unlikely that the presence of any one behavior will ‘rule in’ ASD. Other features of RRBs that might set children with ASD apart are their number and severity. Because most studies have focused on mean scores, we do not know whether young children with ASD have a greater number of RRBs than other children, more severe RRBs, or both. In order to tease these questions apart, it is necessary to determine both the number of RRBs present as well as the distributions of scores on individual items.

To address these unanswered questions, we compared RRBs in young children with ASD to children, nonspectrum developmental delay (DD), and typical development (TD). The following hypotheses were tested:

- (1) Factors: A factor analysis of the RRB items on the ADI-R will confirm the existence of a ‘repetitive sensorimotor’ (RSM) factor and an ‘insistence on sameness’ (IS) factor in young children with ASD.
- (2) Prevalence: The ASD sample will have a high prevalence of RSM behaviors and a low prevalence of IS behaviors. The prevalence of most RRBs will be higher in the ASD than in the DD and TD samples, and higher for children with autism than for

children with PDD-NOS. Children with ASD will have a higher average number of RSM behaviors than children with DD or TD.

(3) Severity: Children with ASD will have a higher proportion of more severe scores on the RSM items than children with DD or TD.

Method

Participants

Data for this study were collected as part of a larger investigation on the early diagnosis of autism, a longitudinal study of toddlers referred for possible autism at age 2. Participants consisted of 192 children under the age of 3 years referred for possible autism, 22 nonspectrum developmentally delayed children, and 65 typically developing children. Demographics for the referral sample are presented in Table 2.1.

A total of 192 children were referred because of concerns about ASD; these children comprised the ‘ASD referral group.’ Children were enrolled into the study between 1989 and 1994. The North Carolina (NC) ASD referral group consisted of 112 consecutive referrals of children younger than 3 years to four TEACCH centers (state-funded clinics providing services for children with autism and related communication disorders in NC). One child’s parents withdrew from participation, leaving a total of 111 NC ‘ASD referral’ children. The Chicago ASD referral group consisted of 81 consecutive referrals of children under age 3 (except for one child who was 37 months at the time of testing) to an autism clinic at a private university hospital. Exclusionary criteria included moderate to severe sensory impairments or cerebral palsy, known genetic abnormalities, and poorly controlled seizures.

The nonspectrum developmental disorder (DD) referral group consisted of 22 developmentally delayed children between the ages of 13 and 35 months who met the same exclusionary criteria and who had never been referred for or diagnosed with autism. They were recruited from the three largest sources of referral to the NC autism clinics. This group was comprised mainly of children with mental retardation of unknown etiology (33%), language disorder (33%), or a known genetic disorder (29%).

The third group consisted of 65 typically developing children. The majority of these participants were recruited from church groups in NC.

Procedure

Children in the ASD and DD groups were assessed when they were approximately 2, 3, 5, and 9 years old, but not all families participated at every follow-up appointment. Children in the TD group were assessed once, at age 2. At each point in the study, families underwent a two-part standardized assessment. A number of diagnostic and cognitive measures were used at the initial evaluation and at each follow-up assessment. In the present study, data from the age 2 assessment were used

Each child was assigned a clinical diagnosis of autism, Pervasive Developmental Disorder – Not Otherwise Specified (PDD-NOS) or a nonspectrum disorder when seen at ages 2, 5, and 9. Following each of the age 2 assessments, the two clinicians who had been involved in the assessment met to review the results and decide on a clinical diagnosis (i.e., a child who was referred for possible autism did not necessarily receive a diagnosis on the autism spectrum). Independent best estimate diagnoses of autism, PDD-NOS, and nonspectrum disorders were then generated by an experienced clinical researcher unfamiliar with the child’s history, using the scores and observations made during testing. When this diagnosis did not agree with the diagnosis of the research clinician who had seen the child, the study director and independent examiner reviewed all the information, watched the video of the child assessment (the Pre-Linguistic Autism Diagnostic Observation Schedule: (PL-ADOS: DiLavore, Lord, & Rutter, 1995) and reached a consensus best estimate diagnosis (see Lord et al., 2006 for a complete description of the diagnostic process).

Measures

The Mullen Scales of Early Learning

Assessments at each time point consisted of a test that would determine an overall intellectual ability score and separate verbal and nonverbal intelligence scores. For the present study, one measure of nonverbal ability and one measure of verbal ability have been selected for each child. These scores are considered to be the “best estimate” of the child’s abilities at that time.

At the age 2 assessment, all of the children with ASD and DD received the Mullen Scales of Early Learning (MSEL: Mullen, 1995), except for one child, who received the Merrill-Palmer Scale of Mental Tests (Stutsman, 1931). The MSEL is a developmental test intended for children from birth through 68 months of age. Scores on the MSEL are organized into five scales: Visual Reception, Fine Motor, Receptive Language, Expressive Language, and Gross Motor (which was not usually administered and is not used to calculate the child's IQ). Each scale results in a T-score with a mean of 50 and a standard deviation of 10, as well as an age equivalent. The sum of the 4 primary domains yields an Early Learning Composite score, which has a mean of 100 and a standard deviation of 15. The Mullen has been found to have high levels of reliability and validity (Mullen, 1995).

Because the MSEL does not yield separate verbal and nonverbal scores, these had to be derived for each child. If the child's scores fell within the standard range to calculate deviation IQs (i.e. T-scores of at least 20 on each subscale), verbal and nonverbal IQ scores were extrapolated. Nonverbal IQ (NVIQ) scores were calculated by adding the T-scores from the two nonverbal subscales, doubling this sum, and then finding the Early Learning Composite score that corresponded to this number. The same thing was done with verbal IQ (VIQ), using the verbal subscales. If it was not possible to extrapolate IQ scores, ratio IQ scores were calculated. Nonverbal ratio IQs were calculated by averaging the age equivalents of the nonverbal subtests to obtain a nonverbal mental age, and then dividing the nonverbal mental age by the chronological age and multiplying by 100. The same method was applied for calculating verbal ratio IQs (i.e. using only verbal subtest age equivalents) and full-scale ratio IQs (i.e. using both verbal and nonverbal subtest age equivalents).

Children in the TD sample received the Bayley Scales of Infant Development (Bayley, 1993). For this test, it is not possible to obtain separate verbal and nonverbal scores, and so the child's full-scale IQ (FSIQ) score was used.

The Autism Diagnostic Interview-Revised

Before each child assessment, a research associate administered a version of the Autism Diagnostic Interview-Revised (ADI-R: Lord, Rutter, & LeCouteur, 1994) to the

child's parent(s). All interviewers had previously established reliability, and reliability checks were made during at least every tenth interview. The ADI-R is a comprehensive parent interview covering most developmental and behavioral aspects of autism. There is a scoring algorithm based on DSM-IV/ICD-10 criteria for autism, which has been shown to discriminate between children with autism and non-autistic developmentally delayed children matched on chronological age and nonverbal IQ. Adequate inter-rater and test-retest reliability and validity have been established with the ADI-R for children and adults. At the initial assessment, a toddler version of the ADI-R was administered to all children in the study. The Toddler ADI-R included 32 new questions and codes specifically relevant to onset of difficulties in the early years (see Lord, Shulman, & DiLavore, 2004).

Scores for RRB items in the ADI-R range from 0 to 3, except for unusual sensory interests, which ranges from 0 to 2. A score of "0" indicates that the specified behavior is not present, a score of "1" indicates that the specified behavior is present in an abnormal form but not sufficiently severe, frequent, or marked to meet the criteria for "2", a score of "2" indicates that a definite abnormality of the type specified is present, and a score of "3" indicates that a more severe manifestation of "2" is present (Rutter, LeCouteur, & Lord, 2003).

For the RRB section of the ADI-R, the parent is asked both whether the child currently exhibits the behavior in question and whether the child has ever exhibited the behavior at any point in his or her life. Reported analyses used scores for the 'current' items only.

The ADI-R was adapted several times over the period in which participants were recruited for this study, and, as a result, some versions of the instrument included items that others did not, though the content of individual items did not change. Data for certain RRB items are therefore missing for some participants.

Results

Diagnostic Classifications

Because children in the ASD and DD samples were evaluated more than once, multiple diagnostic classifications were available for many children. ASD diagnoses

made at older ages tend to be more stable than those made at young ages (Lord et al., 2006); therefore, we chose to use the child's most recent available best estimate diagnosis, rather than their referral or initial diagnosis. Based on most recent available diagnosis, there were 165 children with ASD and 49 children with nonspectrum DD. The diagnostic breakdown was very similar to the breakdown that resulted from using the child's initial diagnosis (161 ASD, 53 DD).

In contrast, the breakdown of specific diagnoses *within* the autism spectrum was somewhat different at the initial evaluation compared to the most recent one (102 autism, 59 PDD-NOS at the initial evaluation versus 117 autism, 48 PDD-NOS at the most recent one). For this reason, analyses comparing children with autism to those with PDD-NOS were conducted twice, using initial diagnosis and most recent diagnosis.

RRB Factors

This paper focuses on RRBs at two levels: individual behaviors and groups of behaviors that have been shown to cluster together in factor analyses. Individual behaviors were examined using scores on RRB items in the ADI-R. We included **self-injury** in these analyses, even though it is not part of the RRB section of the ADI-R, because it is thought to have a repetitive component. We excluded circumscribed interests, because it was only administered to the age 9 cohort, and verbal rituals, because it was only administered to children with phrase speech. We also excluded midline hand movements, as this is a very low-frequency behavior in ASD that is included in the ADI-R to rule out Rett's Disorder.

Our examination of groups of RRBs was based on findings from previous studies that the RRB items on the ADI-R tend to load onto 2 separate factors, one comprised of **repetitive use of objects, unusual sensory interests, hand and finger mannerisms**, and **complex mannerisms** and the other comprised of **compulsions and rituals, difficulties with changes in routine**, and **resistance to trivial changes in environment**. **Unusual preoccupations, unusual attachment to objects, sensitivity to noise, self-injury**, and **abnormal/idiosyncratic response to sensory stimuli** have not been found to load consistently on either factor.

First, it was necessary to determine if we obtained the same factors in our sample as have been found in previous studies. A confirmatory factor analysis was run on the age 2 cohort, using the factors mentioned above. As in previous studies, the cutoff for including an item on a factor was ≥ 0.30 . Our findings indicated that the same factors emerged in our data at each cohort. Loadings ranged from 0.49 to 0.81 for the first factor and from 0.62 to 0.80 for the second factor (see Table 2.2). These findings provided a basis for examining characteristics of RRBs as part of clusters or factors.

For individual behaviors and factors, we examined prevalence and severity. ‘Prevalence’ refers to whether the behavior was reported to be present or not. As mentioned above, the RRB items on the ADI-R are scored from 0 to 3, where 0 indicates that the behavior is not present and scores from 1 to 3 indicate that it is present, to varying degrees of severity. To calculate the prevalence of each RRB, we included all children who received a non-zero score on that behavior. ‘Severity’ refers to the child’s specific score (i.e., from 0 to 3) on the item.

Age and IQ as Covariates

Given findings that age and IQ are strongly associated with many RRBs (Bishop et al., 2006), it was important to consider the role that these factors might play in diagnostic comparisons of RRBs at age 2. Table 2.3 provides chronological age and NVIQ scores by diagnosis. Preliminary analyses using one-way ANOVA indicated that there were group differences in age, $F(2, 276) = 75.40, p < .001$. Post-hoc analyses using Tukey’s HSD found that the mean age was significantly higher for the ASD sample than for the DD sample $t(276), = 2.36, p < .05$ and the TD sample $t(276) = 12.26, p < .001$. The age difference between the DD and TD samples was also significant, $t(276) = 7.46, p < .001$. There were also group differences in IQ. Because separate NVIQ and VIQ scores were not available for the TD sample, the three groups were compared on FSIQ score.² One-way ANOVA indicated that there were significant group differences in FSIQ score, $F(2, 273) = 263.65, p < .001$. Planned contrasts showed that the ASD group had a lower average FSIQ score than the DD sample, $t(273) = -5.86, p < .001$, and the TD sample,

² For the ASD and DD samples, FSIQ was estimated by taking the mean of the NVIQ and VIQ scores, because FSIQ scores were not available. Similar results were obtained when FSIQ score was estimated by taking the average of the child’s verbal and nonverbal mental ages and dividing by chronological age

$t(273) = -22.92, p < .001$. The difference between the DD and TD groups in average FSIQ was also significant, $t(273) = 12.88, p < .001$. The ASD and DD groups were compared on NVIQ and VIQ scores. The ASD group had a significantly lower NVIQ and VIQ scores than the DD sample ($t(212) = 2.57, p < .05$ and $t(212) = 7.75, p < .001$ for NVIQ and VIQ, respectively).

In the present study, we were interested in differences between children with ASD and children with DD that were *not* directly related to group differences in IQ and therefore decided to control for IQ when comparing these two groups. Separate NVIQ and VIQ measures were available for the ASD and DD groups, as described earlier. NVIQ was chosen, because it has been found to be more stable over time in children with ASD (Howlin, Goode, Hutton, & Rutter, 2004). The FSIQ distribution for the TD sample was virtually non-overlapping with the distributions for the ASD and DD samples, indicating a strong association between diagnosis and IQ. Consequently, IQ was excluded as a covariate from analyses that compared the TD group with the other two groups, in order to avoid multicollinearity. Chronological age was included as a covariate in all group comparisons.

Since we were also interested in comparing the prevalence of RRBs at age 2 in children with autism and children with PDD-NOS, we examined whether there were age and IQ differences between these groups. The mean age of the children with autism in months ($M = 29.2, sd = 4.8$) was very similar to the age of the children with PDD-NOS ($M = 30.0, sd = 5.0$). The mean NVIQ score at age 2 for the children with autism ($M = 62.2, sd = 16.7$) was significantly lower than for children diagnosed with PDD-NOS at age 2 ($M = 75.1, sd = 17.8$), $t(163) = -3.82, p < .001$. Because we were interested in differences in prevalence between children with autism and those with PDD-NOS that were *not* accounted for by differences in NVIQ, we decided to include NVIQ as a covariate when comparing these two groups.

Prevalence of RRBs in Young Children

Children with ASD, DD, and TD

Given claims that RRBs are relatively rare in very young children with ASD, we examined the prevalence of RRBs in the age 2 cohort. First, we calculated the prevalence

of each RRB for the ASD, DD and TD samples. Several behaviors were highly prevalent in children with ASD at age 2 (see Table 2.4). **Repetitive use of objects, unusual sensory interests, complex mannerisms and hand and finger mannerisms** were reported in over half of the ASD sample, and **unusual preoccupations** and **abnormal/idiosyncratic response to sensory stimuli** were reported in over one-third of this group.

Logistic regression analyses were conducted to determine which behaviors significantly differed in prevalence according to diagnosis. The ASD group had a higher prevalence than the TD group for the following behaviors: **unusual preoccupations** ($\beta = 1.39, p < .001$); **unusual sensory interests** ($\beta = 2.17, p < .001$); **repetitive use of objects** ($\beta = 2.48, p < .001$); **hand and finger mannerisms** ($\beta = 2.71, p = .001$); **complex mannerisms** ($\beta = 2.86, p < .001$); **abnormal/idiosyncratic response to sensory stimuli** ($\beta = 1.41, p < .05$); **difficulties with changes in routine** ($\beta = 1.21, p < .05$); and **unusual attachments** ($\beta = 1.84, p < .01$). To illustrate the significance of these differences, children with ASD were nearly 15 times more likely to have **unusual sensory interests** than children with DD. There were no significant differences in prevalence between the DD and TD samples.

When the ASD and DD samples were compared, the same behaviors were significantly more common in the ASD sample: **unusual preoccupations** ($\beta = 1.40, p < .01$); **unusual sensory interests** ($\beta = 1.42, p < .001$); **repetitive use of objects** ($\beta = 1.62, p < .001$); **hand and finger mannerisms** ($\beta = 1.54, p < .001$); **complex mannerisms** ($\beta = 1.99, p < .001$); **abnormal/idiosyncratic response to sensory stimuli** ($\beta = 1.07, p < .05$); **difficulties with changes in routine** ($\beta = .93, p < .05$); and **unusual attachments** ($\beta = .99, p < .05$). **Compulsions and rituals** were significantly more common in the ASD sample compared to the DD sample ($\beta = 1.30, p < .05$) but not the TD sample. Not surprisingly, although these differences were statistically significant, they were not as striking as the differences between the ASD and TD subgroups. For example, children with ASD were over 4 times more likely to have **hand and finger mannerisms** than children with DD. There were no significant differences between the ASD and DD subgroups in the prevalence of **sensitivity to noise, self-injury, and resistance to trivial changes in environment**.

Many RRBs that were prevalent in children with ASD were also relatively common in the nonspectrum DD group, suggesting that they are not specific to ASD. In order to test the hypothesis that the number of behaviors exhibited would distinguish the ASD group from the other groups, a univariate analysis of variance (ANOVA) was run to determine if the three groups differed in the total average number of RRB items exhibited. The mean number of items endorsed for the ASD group ($M = 4.97$, $SD = 2.26$) was significantly higher than the mean for the TD group ($M = 1.55$, $SD = 1.90$), $F(1, 275) = 56.64$, $p < .001$. The mean for the DD group ($M = 2.45$, $SD = 2.15$) was also higher than the mean for the TD group, $F(1, 271) = 35.18$, $p < .001$. The ASD group had a significantly higher number of items endorsed than the DD group, $F(1, 210) = 39.82$, $p < .001$.

ASD sample only

We also compared the rates of each behavior for children with autism and children with PDD-NOS, controlling for NVIQ score at age 2 (see Table 2.5). When the child's most recent diagnosis was used, there were no behaviors for which the prevalence significantly differed between the groups, except for **self-injury**, which was significantly *less* common in the autism sample ($\beta = .89$, $OR = .411$), $p < .05$. When the child's initial diagnosis was used, **unusual sensory interests** was significantly more prevalent in the autism subgroup, ($\beta = 1.23$, $OR = 3.42$), $p < .01$, as was **complex mannerisms** ($\beta = 1.07$, $OR = 2.90$), $p < .01$. Because the prevalence of most behaviors was similar in the autism and PDD-NOS groups, subsequent analyses examined RRBs in the ASD sample as a whole rather than dividing it into subgroups.

Number of RSM and IS Behaviors by Diagnosis

Next, we compared the three groups on the number of items endorsed within each RRB type (i.e., RSM vs. IS). As mentioned above, a factor analysis of the RRB items confirmed that behaviors loaded on 2 separate factors, one composed of RSM behaviors (**unusual sensory interests, repetitive use of objects, hand and finger mannerisms, complex mannerisms**), and the other comprised of IS behaviors (**difficulties with changes in routine, resistance to trivial changes in environment, and compulsions**

and rituals). Thus, the maximum total number of items endorsed was 4 for the RSM factor and 3 for the IS factor.

Univariate ANOVA indicated that for the RSM factor, the mean number of items endorsed for the ASD group ($M = 2.73$, $SD = 1.15$) was higher than for the TD group ($M = .63$, $SD = .93$), $F(1, 271) = 109.17$, $p < .001$. The mean for the DD group ($M = 1.19$, $SD = 1.10$) was also higher than the mean for the TD group, $F(1, 271) = 5.78$, $p < .05$. The ASD group had a higher average number of items endorsed for the RSM factor than the DD group, $F(1, 206) = 59.42$, $p < .001$. For the IS factor, the total number of items endorsed for the ASD group ($M = .60$, $SD = .84$) and the DD group ($M = .24$, $SD = .63$) did not differ from the mean for the TD group ($M = .22$, $SD = .63$). However, when the ASD and DD groups were compared, the mean number of IS items endorsed was higher for the ASD group than for the DD group $F(1, 207) = 7.25$, $p < .01$.

Severity of RRBs in Young Children

Severity of individual behaviors for children with ASD, DD, and TD

We hypothesized that, in addition to having a higher prevalence of most RRBs, children with ASD would have higher (i.e., more severe) scores than children who were not on the autism spectrum. In order to address this question, we compared the three groups on distribution of scores for each RRB item on the ADI-R. Scores of 3 were collapsed into 2's, because 3's were rare in the non-ASD groups, particularly in the TD group.

Ordinal regression analyses were run for each RRB. The ASD group had a significantly greater proportion of scores of 2 than the TD group for **unusual preoccupations** ($\beta = 1.88$, $p < .001$); **unusual sensory interests** ($\beta = 2.29$, $p < .001$); **repetitive use of objects** ($\beta = 2.59$, $p < .001$); **hand and finger mannerisms** ($\beta = 2.64$, $p < .001$); **complex mannerisms** ($\beta = 3.01$, $p < .001$); **difficulty with changes in routine** ($\beta = 1.19$, $p < .05$); **abnormal/idiosyncratic response to sensory stimuli** ($\beta = 1.38$, $p < .05$); and **unusual attachments** ($\beta = 1.89$, $p < .01$). There were no differences between the DD and TD groups in proportions of scores on any of the RRB items. The ASD group had a higher proportion of 2's than the DD group for **unusual preoccupations** ($\beta = 1.41$, $p = .001$); **unusual sensory interests** ($\beta = 1.47$, $p < .001$); **repetitive use of objects** ($\beta =$

1.69, $p < .001$); **hand and finger mannerisms** ($\beta = 1.47, p < .001$); **complex mannerisms** ($\beta = 1.98, p < .001$); **difficulty with changes in routine** ($\beta = .909, p < .05$); and **abnormal/idiosyncratic response to sensory stimuli** ($\beta = 1.01, p < .05$).

Figure 2.1 provides a visual representation for four different RRB items on the ADI-R. The score distributions for **repetitive use of objects** (Figure 2.1.1) and **unusual preoccupations** (Figure 2.1.2) were significantly different between the ASD group and the other two groups. Similar distributions were obtained for **unusual sensory interests**, **hand and finger mannerisms**, and **complex mannerisms**. For **unusual preoccupations**, the ASD group had a higher proportion of non-zero scores than the other groups, but the difference in score distributions was not quite as striking as for **repetitive use of objects**. Again, the DD and TD groups had very similar score distributions. **Difficulties with changes in routine** and **unusual attachments** had similar distributions.

The distributions for **resistance to trivial changes in environment** (Figure 2.1.3) and **sensitivity to noise** (Figure 2.1.4) did not differ by diagnosis. For the first item, the vast majority of children in all groups received scores of zero. **Compulsions and rituals** had a similar distribution. In contrast, for sensitivity to noise, all three groups had a substantial minority of non-zero scores, similar to **self-injury**.

Similar to the findings from the prevalence analyses, these results indicated a difference between the RSM items and the IS items. All four of the RSM items consistently showed differences in score distributions by diagnosis, with the ASD group having a greater proportion of more severe scores than the DD and TD samples. In contrast, only one out of the three IS items, **difficulties with changes in routine**, showed these differences, and the effects were modest. The relationship between severity of RRBs and diagnosis appears to be stronger for the RSM factor than for the IS factor.

RSM and IS Severity by Diagnosis

To test the hypothesis that diagnosis was more strongly related to RSM than IS severity, we compared the groups on mean RSM score (i.e., taking the average of all the RSM items) and mean IS score (i.e., taking the average of all the IS items). For these analyses, scores of 3 were included, so that the highest possible RSM score was 12 and

the highest possible IS score was 9. Univariate ANOVA indicated that the mean RSM score for the ASD group ($M = 4.49, SD = 2.39$) was higher than for the TD group ($M = .71, SD = 1.22$), $F(1, 271) = 98.49, p < .001$. The mean for the DD group ($M = 1.63, SD = 1.91$) was also higher than the mean for the TD group, $F(1, 271) = 4.53, p < .05$. The mean RSM score for the ASD group was higher than the mean for the DD group, $F(1, 206) = 49.76, p < .001$. In contrast, the mean IS scores for the ASD group ($M = .96, SD = 1.54$) and the DD group ($M = .43, SD = 1.26$) did not differ from the mean score for the TD group ($M = .34, SD = 1.19$). However, when the ASD and DD groups were compared, there was a significant difference between the two groups, $F(1, 207) = 4.36, p < .05$.

Discussion

Factor analysis using data from 2-year-olds supported the existence of two factors: a ‘repetitive sensorimotor’ (RSM) factor and an ‘insistence on sameness’ (IS) factor. As predicted, RSM behaviors were highly prevalent in the ASD group, whereas IS behaviors were less common. This result is not surprising, given that RSM behaviors are often associated with lower developmental levels (Gabriels, Cuccaro, Hill, Ivers, & Goldson, 2005; Bishop et al., 2006).

As predicted, children with ASD had a higher prevalence of RSM behaviors than children with DD or TD, but these behaviors were relatively common in the DD group. Consequently, the presence of any single RSM behavior could not be used to distinguish children with ASD from children with nonspectrum disorders. IS behaviors did not consistently differ in prevalence among the three groups, although there were some modest differences in the rates of IS behaviors by diagnosis, particularly for difficulties with changes in routine. Because these behaviors were relatively uncommon, however, their absence is not particularly informative in making diagnostic decisions.

These findings suggest that other aspects of RRBs must be examined to determine if there are any features that are particular to children with ASD. In our data, the ASD group had significantly more RRBs in the RSM category than the DD and TD groups. On average, children with ASD had close to three RSM behaviors, while children in the DD and TD groups had an average of approximately one or less. Although having any one

RSM behavior at age 2 is not indicative of ASD, having several of these behaviors might be. The results from the analysis of score distributions suggest that having severe behaviors at young ages, particularly RSM behaviors, is another indicator of ASD. For the RSM items, the proportion of scores of 2 was significantly higher for the ASD sample than for the DD or TD sample.

Because we examined RRBs in multiple ways, we were able to address more specific questions about these behaviors than in previous studies, which could explain why we found more evidence of RRBs in young children with ASD. Some studies have focused on whether children met the RRB domain cutoff on the ADI-R (e.g., Charman et al., 2005), which can mask important information. The ADI-R algorithm requires the clinician to code the higher of two RRB items in two pairs: repetitive use of objects and unusual sensory interests in the first pair, and hand and finger mannerisms and complex mannerisms in the second pair. It is possible, then, for a child to receive a score of 1 on each of these behaviors but still not meet the RRB cutoff of 3 points. Similarly, average RRB scores do not tell us about prevalence. In the study by Moore and Goodson (2003), average RRB scores on the ADI-R were relatively low at young ages, but it is possible that, as in the present study, many behaviors were quite common in young children with ASD. Although Cox et al. (1999) found relatively low prevalence for most RRBs, this could be because only children with ‘definite’ abnormalities (i.e. scores of 2 or 3 on the ADI-R) were counted, whereas in the present study, all children who received non-zero scores were counted. The lower estimates in their study likely excluded children who exhibited RRBs to a mild degree. Stone et al. (1999) highlighted the finding that social and communication impairments were more frequently endorsed by clinicians than RRBs, but several RRBs were endorsed for half or more of the children with ASD at age 2.

The findings from the present study have important implications for research and practice. Clinicians must understand, for example, that a child who does not meet the ADI-R RRB cutoff may still have one or two behaviors that are severe and intrusive enough to preclude him from doing other activities. It is also important that clinicians record and appropriately score all RRBs that are described or observed, even if they seem ‘typical’ for young children (e.g., hand-flapping in excitement). It might also be

appropriate for researchers to consider ways to modify both DSM-IV criteria and ADI-R cut-offs to reflect findings from the present study and growing literature on RRBs in young children.

Contrary to our prediction and to the findings of previous studies, few behaviors differed in prevalence between autism and PDD-NOS, when NVIQ score was controlled for. The fact that we obtained this result even when the most recent diagnoses were used suggests that RRBs in young children with ASD are not strongly related to later diagnoses within the autism spectrum. It was interesting to find that similar results were obtained even when initial diagnoses were used, indicating that the child's RRB profile was not a major factor in the clinicians' decisions to diagnose a 2-year-old with autism as opposed to PDD-NOS. It is possible that differences between children with autism and PDD-NOS in RRB prevalence and severity become clearer as children get older.

Some behaviors were clearly not indicative of ASD, or even of nonspectrum developmental delay, in our sample. Self-injury, sensitivity to noise, and resistance to trivial changes in the environment did not differ in prevalence or in severity across the three groups. Again, it is possible that as children get older, differences in the prevalence and severity of these behaviors begin to emerge, as some become less common in children who are not on the spectrum and more common children with ASD.

Limitations and Future Directions for Research

Our findings are based on parent report from the ADI-R. Given the potential for inaccurate reporting, it is important to corroborate these results using direct observation of young children with ASD, such as the RRB items on the ADOS (Lord, Rutter, DiLavore, & Risi, 1999). Furthermore, certain features of RRBs are not captured by the ADI-R item scores, such as the length of time that the behavior has been exhibited (e.g. one year versus one month) and the specific form of the behavior (e.g. head-banging vs. biting one's wrist or slapping one's face). This could explain why there were some behaviors for which no differences in prevalence or severity were observed.

Nevertheless, the fact that some RRBs did not differ in prevalence and severity according to diagnosis at young ages has important implications. From a practical standpoint, one certainly would not want to rely on the presence or absence of these

behaviors when making a diagnosis. From a theoretical perspective, we can learn more about which behaviors are ‘core’ features of ASD by identifying those that are not.

Another limitation is the generalizability of our findings. The sample of 2-year-olds was unique, in that it was comprised of young children who were being referred for a diagnosis of ASD at a time when such referrals were relatively rare. In order to determine if our results generalize to young children with ASD, it is important to conduct longitudinal studies with more recent samples of young children who are representative of this population.

Similarly, the DD sample is likely not representative of young children with a particular kind of developmental disorder. This group was composed of children with known developmental delays, but the specific delay or disorder varied. It is also important to note that some of the children in this sample who were originally referred for a diagnosis of ASD (i.e., members of the ‘ASD referral sample’) were ultimately diagnosed with a nonspectrum disorder. The fact that there were concerns about ASD for these children suggests they may not have been prototypical of children with nonspectrum developmental disorder. However, if these children *did* show features of ASD, then our finding of differences between the ASD and DD groups suggests that these differences would have been more striking if we had used a ‘cleaner’ DD sample. Nevertheless, to determine if the results obtained here generalize to children with nonspectrum developmental disorder, it is important to replicate these findings using a larger, more homogeneous group of children (e.g. children with language disorders, Down’s syndrome, etc.)

The primary contribution of the present study is to show that we cannot overgeneralize when speaking about RRBs in young children with ASD. Clearly, these behaviors are an important part of the symptom profile for many children with ASD. An important next step is to learn more about how RRBs change in children with ASD over time, such as which behaviors become strongly associated with ASD as children get older. RRBs that do not discriminate between young children with ASD and those not on the spectrum may do so at older ages, particularly ‘insistence on sameness’ behaviors (Silverman et al., 2002). It is important, then, to remember that the meaning of the term ‘autism-specific’ is likely to change with the child’s age.

Most importantly, we still know very little about how trajectories of RRB development vary according to characteristics of the child and of the behavior. Perhaps children with autism show a sharper increase in RRB score as they get older than children with PDD-NOS. Similarly, given that IQ has been found to be closely associated with the expression of many RRBs (Bishop et al., 2006), it likely plays an important role in moderating the relationship between age and RRB prevalence/severity. Additionally, different RRBs might show different patterns of change over time. Findings from several studies suggest that scores on some RRB items increase over time (Moore & Goodson, 2003; Charman et al., 2005; South, Ozonoff, & McMahon, 2005). By examining prevalence and severity separately, we can determine which behaviors become more common and/or impairing as children get older, as well as how stable RRBs are over time (i.e. which behaviors are most commonly acquired or lost).

The answers to these questions will add to our understanding of RRBs in children with ASD. Our findings suggest that having several RSM behaviors, particularly to a severe degree, is a strong indicator of ASD, whereas IS behaviors are not as strongly associated with the disorder, and some behaviors are actually poor diagnostic indicators in young children. This is important information for clinicians to have, as parents of young children may wonder what the presence or absence of RRBs means for their child's diagnosis. The findings from studies on RRB change will be able to tell us more about the significance of having (or not having) RRBs at young ages. If a behavior is acquired by a large proportion of children with ASD as they get older, then the fact that a child does not show that behavior at a young age does not mean that he or she will never display that behavior. Given that RRBs can be particularly stressful for parents and families to deal with (Gabriels et al., 2005), parents may ask how likely it is that these behaviors will persist, and clinicians should provide reasonable answers. Questions related to RRB development are taken up in Chapters III and IV.

Table 2.1. Demographics of Referral Sample

	ASD Referral Group (N=192)	DD Referral Group (N=22)	TD Group (N=65)
Gender			
Male	162 (84.4%)	10 (45.5%)	41 (63%)
Female	30 (15.6%)	12 (54.5%)	24 (37%)
Race			
Caucasian	127 (66.1%)	16 (72.7%)	39 (60%)
African-American	61 (31.8%)	6 (27.3%)	18 (28%)
Other/Unknown	3 (1.6%)	0 (0.0%)	8 (12%)
Site			
North Carolina	111 (57.8%)	22 (100%)	60 (92%)
Chicago	81 (42.2%)	0 (0%)	5 (8%)

Table 2.2. Confirmatory Factor Analysis of RRBs on the ADI-R

	RRB item	Factor loading
Repetitive sensorimotor factor	Repetitive use of objects	.81
	Unusual sensory interests	.67
	Hand and finger mannerisms	.55
	Complex mannerisms	.49
Insistence on sameness factor	Resistance to trivial changes in environment	.91
	Difficulties with changes in routine	.80
	Compulsions and rituals	.62

Table 2.3. Age and IQ Scores by Diagnostic Group

	ASD (N = 165)	DD (N = 49)	TD (N = 65)
Chronological age (in months) ^{abc}	29.4 (4.9)	27.3 (6.1)*	19.7 (6.3)***
Full-scale IQ ^{def}	49.4 (18.2)	67.2 (21.7)***	112.9 (17.0)***
Nonverbal IQ ^e	66.0 (20.4)	74.8 (23.4)*	--
Verbal IQ ^e	32.9 (20.1)	59.5 (24.3)***	--

Note NVIQ and VIQ scores were not available for the TD sample.

* $p = .05$; ** $p < .05$; *** $p < .001$

^a ASD > TD; ^b ASD > DD; ^c DD > TD; ^d ASD < TD; ^e ASD < DD; ^f DD < TD

Table 2.4. Prevalence of RRBs by Diagnostic Group (%)

ADI-R RRB item	ASD (N = 165)	Non-spectrum DD (N = 49)	TD (N = 65)
Unusual sensory interests ^{ab}	78.2	42.9***	24.6***
Repetitive use of objects ^{ab}	79.1	39.6***	20.0***
Complex mannerisms ^{ab}	61.6	18.4***	9.2***
Hand and finger mannerisms ^{ab}	53.7	20.4***	9.2***
Unusual preoccupations ^{ab}	40.2	14.3**	9.2***
Sensitivity to noise ^c	35.0	24.5	32.3
Abnormal/idiosyncratic response to sensory stimuli ^{ab}	35.2	14.3*	6.2*
Difficulties with changes in routine ^{ab}	30.7	14.3*	6.2*
Self-injury ^c	29.7	34.7	18.5
Unusual attachments ^{ab}	26.7	12.2*	4.6**
Compulsions and rituals ^b	20.1	6.1*	9.2
Resistance to trivial changes in environment ^c	9.8	4.1	6.2

* $p < .05$; ** $p < .01$; *** $p < .001$

^aASD > TD; ^bASD > DD; ^cASD = DD = TD

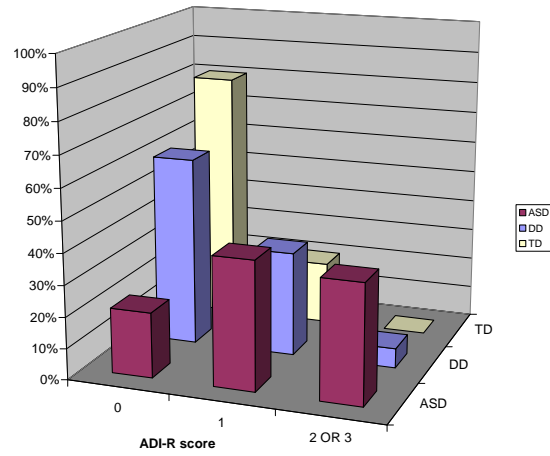
Table 2.5. Prevalence of RRBs for ASD Sample by Spectrum Diagnosis (%)

ADI-R RRB item	Autism (N = 117)	PDD-NOS (N = 48)
Unusual sensory interests	83.8	64.6
Repetitive use of objects	80.0	77.1
Complex mannerisms	64.7	54.2
Hand and finger mannerisms	57.8	43.8
Unusual preoccupations	40.5	39.6
Sensitivity to noise	37.6	28.3
Abnormal/idiosyncratic response to sensory stimuli	35.0	35.4
Difficulties with changes in routine	28.2	37.0
Self-injury	25.6	39.6*
Unusual attachments	22.2	37.5
Compulsions and rituals	21.6	16.7
Resistance to trivial changes in environment	7.7	15.2

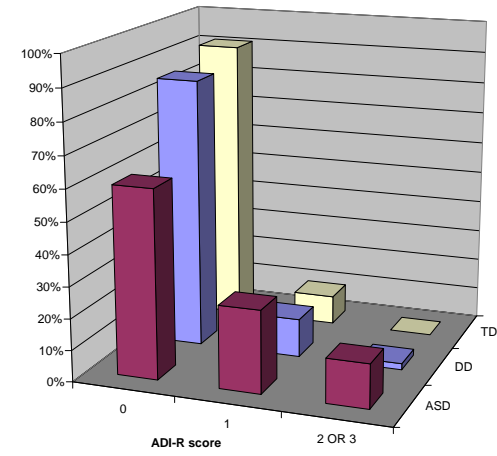
* autism < PDD-NOS, $p < .05$

Figure 2.1. Distributions of ADI-R Item Scores by Diagnostic Category

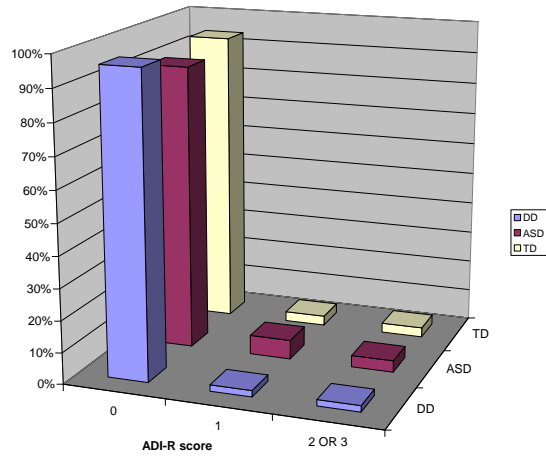
1. Repetitive use of objects



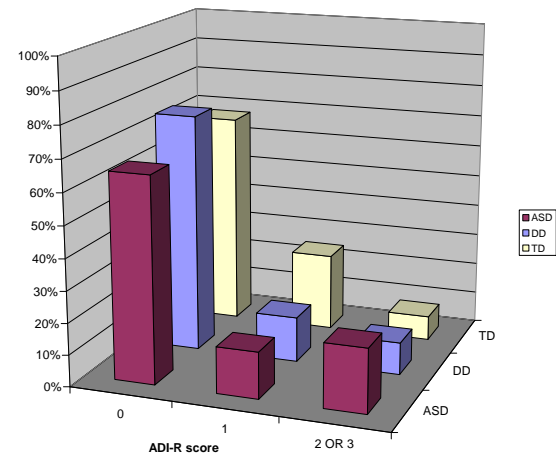
2. Unusual preoccupations



3. Resistance to trivial changes in environment



4. Sensitivity to noise



References

- American Psychiatric Association. (1994). *Diagnostic and Statistical Manual of Mental Disorders* (4th ed.). Washington DC: American Psychiatric Association.
- Baranek, G. T. (1999). Autism during infancy: A retrospective video analysis of sensory-motor and social behaviors at 9-12 months of age. *Journal of Autism & Developmental Disorders*, *29*, 213-224.
- Bayley, N. (1993). *Bayley Scales of Infant Development* (2nd ed.). San Antonio: The Psychological Corporation.
- Bishop, S. L., Richler, J., & Lord, C. (2006). Restricted and repetitive behaviors and nonverbal IQ in children with autism spectrum disorders. *Child Neuropsychology*, *12*, 247-267.
- Bodfish, J. W., Symons, F. J., Parker, D. E., & Lewis, M. H. (2000). Varieties of repetitive behavior in autism: Comparisons to mental retardation. *Journal of Autism and Developmental Disorders*, *30*, 237-243.
- Bryson, S. E., McDermott, C., Rombough, V., & Zwaigenbaum, L. (in press). The Autism Observation Scale for Infants: Scale development and Reliability. *Journal of Autism and Developmental Disorders*.
- Charman, T., Taylor, E., Drew, A., Cockerill, H., Brown, J. A., & Baird, G. (2005). Outcome at 7 years of children diagnosed with autism at age 2: predictive validity of assessments conducted at 2 and 3 years of age and pattern of symptom change over time. *Journal of Child Psychology and Psychiatry*, *46*, 500-513.
- Cox, A., Klein, K., Charman, T., Baird, G., Baron-Cohen, S., Swettenham, J., Drew, A., Wheelwright, S., & Nightingale, N. (1999). Autism spectrum disorders at 20 and 42 months of age: Stability of clinical and ADI-R diagnosis. *Journal of Child Psychology and Psychiatry*, *40*, 719-732.
- Cuccaro, M. L., Shao, Y., Grubber, J., Slifer, M., Wolpert, C. M., Donnelly, S. L., Abramson, R. K., Ravan, S. A., Wright, H. H., DeLong, R. G., & Pericak-Vance, M. A. (2003). Factor analysis of restricted and repetitive behaviors in autism using the Autism Diagnostic Interview-R. *Child Psychiatry and Human Development*, *34*, 3-17.
- DiLavore, P., Lord, C., & Rutter, M. (1995). The Pre-Linguistic Autism Diagnostic Observation Schedule. *Journal of Autism & Developmental Disorders*, *25*, 355-379.
- Gabriels, R. L., Cuccaro, M. L., Hill, D. E., Ivers, B. J., & Goldson, E. (2005). Repetitive behaviors in autism: Relationships with associated clinical features. *Research in Developmental Disabilities*, *26*, 169-181.
- Howlin, P., Goode, S., Hutton, J., & Rutter, M. (2004). Adult outcome for children with autism. *Journal of Child Psychology and Psychiatry*, *45*, 212-229.

- Hus, V., Pickles, A., Cook, E. H., Risi, S., & Lord, C. (2007). Using the autism diagnostic interview-revised to increase phenotypic homogeneity in genetic studies of autism. *Biological Psychiatry, 61*, 438-448.
- Lord, C. (1995). Follow-up of two-year-olds referred for possible autism. *Journal of Child Psychology & Psychiatry & Allied Disciplines, 36*, 1365-1382.
- Lord, C., Risi, S., DiLavore, P. S., Shulman, C., Thurm, A., & Pickles, A. (2006). Autism from 2 to 9 years of age. *Archives of General Psychiatry, 63*, 694-701.
- Lord, C., Rutter, M., DiLavore, P. C., & Risi, S. (1999). *Autism Diagnostic Observation Schedule*. Los Angeles: Western Psychological Services.
- Lord, C., Shulman, C., & DiLavore, P. (2004). Regression and word loss in autistic spectrum disorders. *Journal of Child Psychology and Psychiatry, 45*, 936-955.
- Mahone, E. M., Bridges, D., Prahme, C., & Singer, H. S. (2004). Repetitive arm and hand movements (complex motor stereotypies) in children. *Journal of Pediatrics, 145*, 391-395.
- Moore, V., & Goodson, S. (2003). How well does early diagnosis of autism stand the test of time? Follow-up study of children assessed for autism at age 2 and development of an early diagnostic service. *Autism, 7*, 47-63.
- Mullen, E. (1995). *The Mullen Scales of Early Learning*. Circle Pines, MN: American Guidance Service Inc.
- Rutter, M., LeCouteur, A., & Lord, C. (2003). *Autism Diagnostic Interview-Revised*. Los Angeles: Western Psychological Services.
- Silverman, J. M., Smith, C. J., Schmeidler, J., Hollander, E., Lawlor, B. A., Fitzgerald, M., Buxbaum, J. D., Delaney, K., & Galvin, P. (2002). Symptom domains in autism and related conditions: Evidence for familiarity. *American Journal of Medical Genetics, 114*, 64-73.
- South, M., Ozonoff, S., & McMahon, W. M. (2005). Repetitive behavior profiles in Asperger syndrome and high-functioning autism. *Journal of Autism and Developmental Disorders, 35*, 145-158.
- Stone, W. L., Lee, E. B., Ashford, L., Brissie, J., Hepburn, S. L., Coonrod, E. E., & Weiss, B. H. (1999). Can autism be diagnosed accurately in children under 3 years? *Journal of Child Psychology and Psychiatry, 40*, 219-226.
- Stutsman, R. (1931). Guide for administering the Merrill-Palmer Scale of Mental Tests. In *Mental Measurement of Preschool Children* (L.M. Terman ed., pp. 139-262). New York, NY: Harcourt, Brace & World.
- Szatmari, P., Georgiades, S., Bryson, S., Zwaigenbaum, L., Roberts, W., Mahoney, W., Goldberg, J., & Tuff, L. (2006). Investigating the structure of the restricted, repetitive behaviours and interests domain of autism. *Journal of Child Psychology and Psychiatry, 47*, 582-590.

- Walker, D. R., Thompson, A., Zwaigenbaum, L., Goldberg, J., Bryson, S. E., Mahoney, W. J., Strawbridge, C. P., & Szatmari, P. (2004). Specifying PDD-NOS: A comparison of PDD-NOS, Asperger syndrome, and autism. *Journal of the American Academy of Child and Adolescent Psychiatry, 43*, 172-180.
- World Health Organization. (1992). *The ICD 10 Classification of Mental and Behavioral Disorders: Clinical Descriptions and Diagnostic Guidelines*. Geneva, Switzerland.: World Health Organization.
- Zwaigenbaum, L., Bryson, S., Rogers, T., Roberts, W., Brian, J., & Szatmari, P. (2005). Behavioral manifestations of autism in the first year of life. *International Journal of Developmental Neuroscience, 23*, 143-152.

Chapter III. Developmental Trajectories of Restricted and Repetitive Behaviors and Interests (RRBs) in Children with Autism Spectrum Disorders: Further Evidence of RRB Subdomains

Background and Significance

In the past decade or so, autism researchers have begun to pay more attention to the role that development plays in Autism Spectrum Disorders (ASD). Longitudinal studies that have followed children from a young age have revealed different developmental trajectories in ASD, and the kinds of variables that are associated with these trajectories. For example, we now know that early verbal skills are a strong predictor of later social ability (Venter, Lord, & Schopler, 1992) and that degree of early cognitive impairment is associated with levels of independence in adults with ASD (Howlin, Goode, Hutton, & Rutter, 2004). However, relatively little is known about trajectories of development of restricted and repetitive behaviors and interests (RRBs) in ASD, such as whether these behaviors tend to become more severe or improve over time. Even less is known about which variables are predictive of different trajectories.

The current empirical findings are mixed regarding the association between age and the number and severity of RRBs in ASD. In a cross-sectional study of children aged 22-51 months, Mooney, Gray, & Tonge (2006) failed to find differences between older and younger children with ASD in RRB domain scores on the on the Autism Diagnostic Interview – Revised (ADI-R: Lord, Rutter, & Le Couteur, 1994). However, longitudinal studies have found that RRB total scores on the ADI-R increased for children with ASD between the ages of 20 and 42 months and between the ages of 24 months and 48-60 months, respectively (Cox et al., 1999; Moore & Goodson, 2003), indicating that children acquired new RRBs and/or displayed the same ones to a more severe degree as they got older.

As children move past the preschool years and into school age, most studies have found that the number and/or severity of some RRBs decreases. For example, Charman et

al. (2005) that ADI-R RRB domain scores increased from 3 to 4-5 yrs, and then decreased by age 7, indicating that the child reportedly had fewer and/or less severe RRBs by this age. Other studies have also found a tendency for RRBs to become less severe over time (Seltzer, Shattuck, Abbeduto, & Greenberg, 2004; South, Ozonoff, & McMahon, 2005). Despite this improvement, RRBs tend to persist, even into adulthood (Howlin et al., 2004).

One explanation for these mixed findings could be that the development of RRBs depends on the behavior in question. Moore & Goodson (2003) found that ADI-R scores for unusual preoccupations, compulsions and rituals, hand and finger mannerisms, and repetitive use of objects increased between 2 and 4-5 years, while complex mannerisms decreased between these ages. South et al. (2005) reported that, in a sample of participants aged 7-20, severity scores on the three domains of the Repetitive Behavior Interview (RBI: Turner, 1997) – Object Use, Motor Movements, and Rigid Routines – tended to be highest in the preschool years and then decrease over time. In contrast, scores on the circumscribed interests category of the Yale Special Interests Interview (YSII: South, Klin, & Ozonoff, 1999) tended to gradually increase over time.

These findings raise the question of whether RRBs that are alike in some way follow similar patterns of development. Findings from various factor analyses support the notion that there are different subtypes of RRBs. Cuccaro et al. (2003) conducted a factor analysis of the ADI-R RRB items and found evidence for two factors, one called ‘repetitive sensorimotor,’ comprised of behaviors such as hand and finger and complex body mannerisms, repetitive use of objects, and unusual sensory interests, and the other, ‘resistance to change,’ comprised of behaviors such as compulsions and rituals, difficulties with changes in routine, and resistance to trivial changes in the environment. These factors have since been replicated using other datasets (Bishop, Richler, & Lord, 2006; Richler, Bishop, Kleinke, & Lord, 2007; Szatmari et al., 2006), and some differences between the factors have been noted. In the study by Richler et al. (2007), repetitive sensorimotor behaviors were very common in young children with ASD, whereas resistance to change, or what the authors referred to as ‘insistence on sameness’ behaviors, were uncommon. Adopting a factor analytic approach to the study of the

developmental course of RRBs in ASD could help us understand how these behaviors change over time, building on the recent findings that RRB development may be different in these two factors. For example, a longitudinal study that included children with severe intellectual disabilities and/or autism found that sensorimotor RRBs improved over time, whereas RRBs characterized by resistance to change did not (Murphy et al., 2005). This study did not limit its sample to children with ASD and did not use a factor analytic approach. Doing so in the future may enhance our understanding of RRB development in ASD.

In addition to examining how developmental trajectories of RRBs vary according to the characteristics of the behavior, it is also important, both clinically and theoretically, to consider how they vary according to characteristics of the child, such as gender, cognitive ability, diagnosis, and social functioning. Findings on gender and RRBs have been mixed, with some studies finding no association and (Carter, 2007) and others finding that gender was related to specific behaviors, such as unusual sensory interests (Lord, Schopler, & Revicki, 1982). Higher levels of maternal education have been associated with more rapid development in the area of verbal ability (Anderson et al., in press), but the relationship between this variable and RRB development has not been examined.

Findings from these studies suggest that individual differences in cognitive abilities in children with ASD can affect the likelihood that certain RRBs will be present at different points in development. However, the results from longitudinal work examining IQ and RRBs across development have been inconsistent. In the study by (Murphy et al., 2005), IQ at time 1 (when all children in the sample were under 15 years old) was associated with RRBs at time 1, but did not predict RRBs at time 2 (12 years later). On the other hand, the findings from a study by Butter and colleagues (2006) suggested that children with autism and PDD-NOS who underwent early intervention and had gains in IQ also demonstrated a reduction in autistic symptoms, including RRBs. A longitudinal case study found that 5 of 9 children who demonstrated a decrease in IQ from 12 to 24 months of age had increasing RRBs during the same period (Bryson et al., 2007).

Again, the kind of behavior might be important to consider. Some of the studies that have grouped RRBs into factors have found that RSM behaviors are related to lower level of adaptive and/or cognitive functioning Bishop et al., 2006; (Gabriels, Cuccaro, Hill, Ivers, & Goldson, 2005; Bishop et al., 2006; Carcani-Rathwell, Rabe-Hasketh, & Santosh, 2006), while IS behaviors are not related to level of functioning, but may be associated with communication impairments (Szatmari et al., 2006). Using cross-sectional data, Bishop et al. (2006) found that nonverbal IQ was more closely related to the likelihood of having RRBs, particularly RSM behaviors, in older children compared to younger children. These findings raise the question of whether IQ has a different relationship with the development of RSM versus IS behaviors; longitudinal data are necessary in order to address this question.

Impairments in the other core domains of ASD have also been found to be associated with RRBs. For instance, improvement in social interaction skills has been linked to decreased RRBs in children with autism (Koegel, Koegel, Hurley, & Frea, 1992). Recent longitudinal research further supports social functioning as a predictor of RRB trajectories. In one study, scores on the Reciprocal Social Interaction domain of the ADI-R at age 36 months were predictive of RRBs at age 7 (Charman et al., 2005). In addition, recent work reported having relatively mild social impairments in childhood is associated with milder RRBs in late adolescence/early adulthood (McGovern & Sigman, 2005). Although this last study did not explicitly examine the relationship between these two variables, taken together, these findings suggest that aspects social development may be important predictors of trajectories of RRBs.

The effects of language and communication skills on RRBs and their development are unclear at this point. In a study of children ages 3 to 5, language level had no significant effect on RRBs (Lord & Pickles, 1996). Szatmari and colleagues (2006) found communication impairments on the ADI-R were linked to concurrent insistence on sameness RRBs. In some children, the emergence and development of certain RRBs appear to be linked to communication impairments. For example, in the study by Bryson et al. (2007) children with ASD and language delays were more likely

than children with ASD and no language delays to exhibit repetitive motor mannerisms, restricted interests and difficulties relinquishing toys,

A child's specific diagnosis within the autism spectrum (i.e., autism vs. PDD-NOS) might also provide important information about the development of these behaviors. Walker et al. (2004) found that children with autism aged 1 to 18 years exhibited significantly more impairment than children with PDD-NOS in all of the domains of the ADI-R, including RRBs. Murphy et al. (2005) found that diagnosis at time 1 predicted RRBs and other "challenging behavior" at time 2, such that the children with autism had more RRBs than children with PDD-NOS. However, in a prospective, longitudinal study of high functioning children with ASD, Starr and colleagues (2003) did not find differences in RRB domain scores of the ADI-R when comparing children with autism and those with Asperger's syndrome from ages 6 to 8.

Findings from previous studies suggest that how RRBs change in children with ASD over time depends on a variety of factors, including the kind of behaviors being examined, and the characteristics of the children in the sample. Another important issue to consider is how RRBs, as a category of behavior, are defined and measured. Most studies of RRB development have looked at how mean RRB scores change over time on a group level, which presents some limitations. A focus on scores does not allow for a distinction to be made between prevalence (i.e., whether a behavior is present) and severity (i.e., if present, how impairing the behavior is). If RRB scores increase for a child, this could be because the child acquires more behaviors, or because the behaviors s/he already had become more impairing, or both. In order to tease these questions apart, it is necessary to look not only at how scores change, but how the number of behaviors changes over time. Furthermore, the focus on scores at a group level does not provide a clear picture of the variability in RRB trajectories among individual children with ASD. It is possible that, although children with autism or PDD-NOS tend to follow a certain trajectory on average, there is considerable within-group variability. For example, it could be that many children with autism have scores that stay the same over time, a handful have large score increases and a similar number have slight decreases. On a group level, there would be a net increase, but this would not reflect the actual

heterogeneity in trajectories. It is important to establish whether children tend to cluster into different groups of trajectories, and, if so, which variables affect the likelihood of following one trajectory over another. This understanding could help further efforts to identify relatively homogeneous ASD phenotypes, a crucial step in determining which genes are associated with the disorder.

This paper adds to the existing body of work on RRBs by using longitudinal data, collected when children were approximately 2, 3, 5, and 9 years of age, to investigate how RRBs change in children with ASD over time, and which variables predict these changes. Predictors of trajectories will be examined both in children with ASD (i.e. autism and PDD-NOS) and in a control group of children with nonspectrum developmental disorders (DD). Among children with ASD, we will also consider within-group heterogeneity and how trajectories tend to cluster together, as well as the factors that make a child more likely to follow a given trajectory. RRBs will be considered as a single category and as part of factors, and changes in both the number and severity of behaviors will be addressed. Based on previous research, we predict the following:

- 1) RRB total scores, as well as the total number of RRBs, will increase over time, with the greatest increase occurring between ages 2 and 5.
- 2) Having autism, a lower nonverbal IQ score, a greater degree of social impairment, and a lower level of language at age 2 will all be predictive of greater increases in total RRB scores over time.
- 3) For the RSM factor, we predict that scores will remain relatively stable over time and possibly even decrease by age 9, particularly for children with DD. The same predictors associated with changes RRB total scores will also be related to changes in RSM scores.
- 4) IS behaviors, which are relatively uncommon in very young children, will increase sharply over time. Children with ASD will show greater increases in IS scores over time than children with DD, but NVIQ will not be strongly related to patterns of change in IS behaviors.

By examining these hypotheses, we will obtain a more detailed picture of development in ASD. If the results indicate different trajectories of development for RSM and IS behaviors, this will provide further evidence for the idea that there are two distinct RRB ‘subtypes.’

Method

Participants

Data for this study were collected as part of a larger, longitudinal investigation on the early diagnosis of autism (see Lord et al., 2006; Anderson et al., in press). Participants consisted of children under the age of 3 years who were referred for evaluation for possible autism and children of the same age with nonspectrum developmental disorders. Children were then followed up at approximately the ages of 3, 5, and 9. A sample of typically developing children was included as a control group at the first wave of data collection, but these children were not seen in subsequent waves. (Please see Chapter II for a detailed description of the complete referral sample). Because not all families participated at every follow-up appointment, sample sizes and other characteristics vary for each period of data collection (see Table 3.1).

Procedure

Of the 214 children recruited into the study, 81 received the ADI-R at all 4 waves of data collection, 107 received the ADI-R at three different waves, 21 received the ADI-R at 2 waves, and 5 children received the ADI-R at the first wave only. Children in the DD referral group were not assessed at age 3, and, with a few exceptions, children from Chicago were not seen at age 5. Of the original participants, 42 (19.6%) were lost to follow-up by age 9 due to geographical relocation, unreachable status, or refusal to participate. Attrition was not related to original best-estimate diagnosis, gender, verbal and non-verbal IQ, adaptive functioning, and level of language (as measured by the Autism Diagnostic Interview-Revised). However, higher levels of attrition were associated with non-Caucasian race and lower levels of maternal education (Lord et al., 2006).

At ages 2, 5, and 9, each child was assigned a consensus best estimate clinical diagnosis of autism, Pervasive Developmental Disorder-Not Otherwise Specified (PDD-NOS) or a nonspectrum disorder based on clinical observations, the results of the ADI-R and ADOS, and DSM criteria. All examiners who had seen the child and/or interviewed the caregiver(s) were involved in making the diagnosis. Diagnoses were not given at the age 3 assessment. At age 5, diagnoses were made by examiners blind to the child's history. At age 9, there was always at least one examiner unfamiliar with the child, and about 70% of the time, both examiners were 'blind.' Because children were seen multiple times, diagnoses changed for some children. Even at age 2, the breakdown of ASD vs. nonspectrum diagnoses differed from that of the referral sample of 192 referred for ASD and 22 referred for nonspectrum DD. This is because some of the children referred for ASD were diagnosed as nonspectrum at age 2.

One of the aims of the present study is to understand the relationship between early child characteristics and RRB development, and thus, most analyses compare RRB scores in groups based on the child's earliest diagnosis, (i.e. at age 2). However, for children who continued participation at least through age 5 (n=196), we also conducted analyses grouping children by their most recent diagnosis, as it was thought that this diagnosis would be a more accurate reflection of their eventual outcome. The breakdown for most recent diagnosis, 151 children (77%) with ASD (autism or PDD-NOS) and 45 children (23%) with DD, was very similar to the breakdown for initial diagnosis (75% ASD, 25% DD). In contrast, the distribution of specific diagnoses within the autism spectrum was somewhat different at the initial evaluation compared to the most recent one (63% autism, 37% PDD-NOS at the initial evaluation vs. 72% autism, 28% PDD-NOS at the most recent one). This change was mostly accounted for by the fact that many of the children diagnosed with PDD-NOS at age 2 were ultimately diagnosed with autism at their most recent assessment. No one in the sample was given a diagnosis of Asperger's Disorder (AD), because we adhered to the DSM-IV criterion requiring that autism be ruled out before a diagnosis of AD can be considered (see American Psychiatric Association, 1994). In our sample, children who would have met criteria for AD also met criteria for autism.

At each point in the study, families underwent a two-part standardized assessment that included a parent interview and a child observation. Parents were administered the Autism Diagnostic Interview-Revised (ADI-R: Rutter, LeCouteur, & Lord, 2003) and the Vineland Adaptive Behavior Scales (VABS: Sparrow, Balla, & Cicchetti, 1984). Children were administered the Autism Diagnostic Observation Schedule (ADOS: Lord, Rutter, DiLavore, & Risi, 1999), as well as various cognitive and language measures.

Measures

Cognitive testing

Cognitive assessments at each point of data collection consisted of a test that would determine an overall intellectual ability score and separate verbal and nonverbal intelligence scores. For the present study, one measure of nonverbal ability and one measure of verbal ability have been selected for each child at each age. These scores are considered to be representative of the child's cognitive abilities at that time.

At the age 2 assessment, all of the children with ASD and DD received the Mullen Scales of Early Learning (MSEL: Mullen, 1995), except for one child, who received the Merrill-Palmer Scale of Mental Tests (Stutsman, 1931). (Please see Chapter II for a description of how separate VIQ and NVIQ scores were calculated on the MSEL). At follow-up assessments, the selection of psychometric followed a standard hierarchy. If a child did not have sufficient language to be administered the Wechsler Intelligence Scale for Children-3rd Edition (WISC-III: Wechsler, 1991) or the Differential Ability Scales (Elliott, 1990), then he or she was administered the MSEL.

The *Autism Diagnostic Interview-Revised (ADI-R)*

Before each child assessment, a research associate administered the ADI-R to the child's parent(s). The ADI-R is a comprehensive parent interview covering most developmental and behavioral aspects of autism. At the first assessment, a toddler version of the ADI-R was administered to all children in the study. (Please see Chapter II for a description of the ADI-R and Toddler ADI-R).

To examine RRBs as a category, we calculated an RRB ‘current total’ score for each child at each time point. The current total score was the sum of the child’s scores on the ‘current’ items in the RRB section of the ADI-R that can be administered to children of all ages and language levels. (See Table 3.2 for a list of these behaviors and examples of each). As in the study by Richler et al. (see Chapter II of this volume), we excluded midline hand movements, as this was a very low-frequency behavior in the ASD sample and is included in the ADI-R in order to rule out a diagnosis of Rett’s Disorder. We excluded unusual attachments and abnormal/idiosyncratic response to sensory stimuli, as these items were not administered to many children at age 9. We also excluded circumscribed interests, because it was only administered to the age 9 cohort, and stereotyped speech and verbal rituals, because they were only administered to children with phrase speech. Self-injury was included in the ‘current total score’, even though this item is not in the RRB section of the ADI-R, because these behaviors are thought to have a repetitive component (American Psychiatric Association, 1987).

In addition to calculating a ‘current total’ score, based on the actual item scores, we also calculated a ‘total number of items exhibited’ score. The items used to calculate this total were the same as the ones included in the ‘current total’ score described above. In order to be considered as having the behavior, the child had to receive a non-zero ‘current’ score on the item, indicating that the abnormality was reported to be present to some degree. Thus, the ‘total items’ score represents the sum of all the current items for which the child received a non-zero score.

The *Autism Diagnostic Observation Schedule (ADOS)*

Children were administered the Pre-Linguistic Autism Diagnostic Observation Schedule (PL-ADOS: DiLavore, Lord, & Rutter, 1995) at ages 2 and 3 and the ADOS at ages 5 and 9. The ADOS is a semi-structured measure consisting of tasks that allow the examiner to directly observe the child’s social and communicative behaviors. An algorithm calculates summary scores. Algorithms have recently been revised, so that there are now separate algorithm scores for the areas of Social Affect and Restricted and

Repetitive Behaviors (Gotham, Risi, Pickles, & Lord, 2007). Scores on the Social Affect (SA) subdomain were of primary interest for the present study. This subdomain includes behaviors associated with reciprocal social interaction (e.g. eye contact, quality of social overtures and responses) and communication (e.g. gestures, conversational ability). As with the ADI-R, higher scores indicate a greater degree of impairment. Children received one of three modules of the ADOS, depending on language level (Module 1 for single or no words, Module 2 for phrase speech, and Module 3 for fluent speech). Reliability of at least 80% exact agreement was obtained by raters prior to the start of the study, and was maintained throughout the study.

Analyses

One of the aims of this study was to determine which variables predicted patterns of change in RRBs in children with ASD. Growth curve analysis with SAS Proc Mixed (SAS for Windows release 9.1.3) was used to address this question. A random intercept and slope were calculated for each child to control for the high correlations between repeated measures on the same individual. The growth curve models allowed us to compare the different diagnostic groups on the average RRB score at age 2 (i.e., the intercept), the rate of change in scores from age 2 to 9 (i.e., the slope); and the pattern of change (i.e., linear vs. quadratic). Covariates were added as fixed effects to determine whether they explained any of the variance in intercepts and slopes. Age was the primary predictor of interest, since we were interested in whether scores increased over time. Diagnosis, NVIQ, and Social Affect (SA) algorithm score on the ADOS, all at the age 2 assessment, were included as well. We included a measure from the ADOS because we wanted to incorporate variables from measures that involved direct observation rather than relying exclusively on parent report. We also included gender, race (Caucasian vs. non-Caucasian³), mother's level of education (college or graduate degree vs. less than college degree) and site at which the child was recruited (North Carolina vs. Chicago) as

³ Less than 2% of the participants identified as being neither Caucasian nor African-American; therefore, these participants were categorized in the 'non-Caucasian' group, although this group was predominantly African American.

covariates. For analyses of total number of RSM and IS items, we used Proc Genmod (SAS for Windows release 9.1.3), which can be used to model ordinal data, and also controls for repeated measures.

The sequence of analysis was as follows: First, we ran a model using Proc Mixed in SAS, with child's age at assessment as the only predictor, in order to have a baseline from which to assess the contribution of other factors. Age-squared was also included in order to determine if there were quadratic effects for age, i.e. if increases in RRB scores over time slowed at any point. Age was centered at 29 months, the approximate mean age of children at the initial assessment, to allow for interpretation of the intercepts. Next, we added fixed effects to see if they accounted for any variance in RRB scores at age 2. NVIQ and SA scores at age 2 were centered at the overall mean. Finally, we added interactions with age and age-squared, in order to see if these variables affected the rate of change in scores over time.

Our second objective was to learn more about the different patterns of RRB change among children with ASD. To explore this question, we used a modeling procedure called Traj (Jones, Nagin, & Roeder, 2001), an exploratory procedure written for use in SAS that identifies linear and nonlinear patterns in longitudinal data and classifies the sample into groups based on each individual's trajectory. We ran a series of models using the censored, normal distribution, to see if distinct groups would emerge within ASD. (For total number of RSM and IS items, we used the zero-inflated, Poisson distribution, which can be used for count data in which there are more zeros than would be expected under the Poisson assumption. See Jones et al., 2001 for further explanation). In order to decide which model provided the best fit, we compared the absolute value of the Bayesian Information Criterion (BIC) between different models, where smaller values indicate a better fit (see Jones et al., 2001 for the use of the BIC for model selection).

Next, we assessed whether different variables significantly affected the likelihood of assignment to one group over the others, using t-tests for the individual parameter estimates. We considered a variable to be a risk factor if it significantly affected the

relative likelihood of being in a particular trajectory group, controlling for the other variables in the model.

Results

RRB Total Scores

Predictors of change in children with ASD and DD

A set of models was run to examine predictors of RRB total scores over time. The same set of models was run with total number of RRB items endorsed as the outcome variable instead of total score, and similar results were obtained. Therefore, only results for RRB total score are reported here.

(i) Reduced model

First, we ran a model using Proc Mixed in SAS, with child's age at assessment as the only predictor, in order to have a baseline from which to assess the contribution of other factors. Age-squared was included in order to determine if there were quadratic effects for age, i.e., if increases in RRB scores over time slowed at any point.

Results are reported in model 1 of Table 3.3. The overall intercept (i.e., the average RRB total score for the whole sample at age 2) was approximately 6. The significant positive effect of age indicates that as age increased, so did RRB total scores. There was not a significant effect for age-squared. The random effects at the bottom of the table indicate that there was still significant variability in children's RRB scores at the age 2 assessment, as well as in their rates of change, after age at testing was taken into account. It was necessary to add more variables to the model to explain some of this variability.

(ii) Differences by diagnosis

Model 2 of Table 3.3, depicted graphically in Figure 3.1, tested for differences in RRB total scores according to age 2 diagnosis. Age continued to have a significant,

positive relationship with scores. There was also a significant main effect for age-squared, indicating that, collapsing across diagnostic groups, increases in scores slowed as children got older. The intercept for children with nonspectrum DD was 3.28, significantly lower than the intercepts for children with autism and PDD-NOS, which were 7.13 and 6.00, respectively. The difference between the intercepts for children with autism and PDD-NOS approached but did not reach significance. As predicted, the difference in intercepts by age 2 diagnosis remained significant even after controlling for demographic variables, including the child's race and gender, the mother's level of education, and the site at which the child participated. The interaction between age and diagnosis was not significant. However, only the autism group had a significant linear slope, $t(459) = 2.73, p < .01$, and a significant quadratic effect, $t(460) = -2.08, p < .05$. Thus, the main effect of age and age-squared was driven by the autism group; as can be seen in Figure 3.1, slopes for the PDD-NOS and nonspectrum groups were not as steep and did not change over time.

(iii) Covariates affecting the intercept

Model 3 of Table 3.3 added several covariates that were hypothesized to explain additional variance in the intercepts. We also wanted to see if age 2 diagnosis would remain significant once these variables were controlled for. NVIQ and SA score at age 2 were added.

Age continued to have a significant positive relationship with RRB total scores. NVIQ was highly significant; children with higher NVIQ scores at age 2 tended to have lower RRB total scores at the initial assessment. SA score at age 2 did not have a significant effect on RRB scores. It is important to note, however, that when the same analysis was run without age 2 diagnosis, SA score *was* significant; children with higher SA scores, indicating a greater degree of social impairment, also had higher RRB total scores. Children with DD continued to have lower RRB scores at age 2 than either children with autism or PDD-NOS, even after controlling for NVIQ and SA score.

(iv) Covariates affecting the slopes

Next, we wanted to see whether the variables introduced in Model 3 influenced the slope of RRB scores over time. Linear and quadratic effects were added for NVIQ score and SA score (see Model 4 of Table 3.3). There were no significant linear or quadratic effects for SA score at age 2. As predicted, there was a significant linear effect for NVIQ at age 2; children with lower NVIQs at age 2 tended to show a greater increase in RRB total scores over time compared to children with higher NVIQs at age 2.

In order to understand the nature of this interaction, we divided the sample into 4 NVIQ sub-groups based on the overall group mean NVIQ score: children more than one standard deviation below the group mean, children within one standard deviation below the group mean, children within one standard deviation above the group mean, and children more than one standard deviation above the group mean NVIQ score. Only children in the two lower NVIQ groups showed a significant increase in scores (see Figure 3.2). This interaction explains why, in this model, there was no longer a significant overall effect of age or of age-squared; age was only strongly related to RRB scores for children on the lower end of the NVIQ distribution.

Although diagnosis at age 2 was consistently found to be strongly associated with children's RRB total scores at age 2, it was not a strong predictor of trajectories of change in RRB scores. Knowing that more than half of the children initially diagnosed with PDD-NOS eventually received diagnoses of autism (Lord et al., 2006), we wondered if the weak predictive value of early diagnosis was partly due to the lack of stability of early diagnoses of PDD-NOS. Children whose diagnosis changed from PDD-NOS at age 2 to a more severe diagnosis of autism at a subsequent time point may have followed a different (and presumably more severe) trajectory of RRB scores than children who maintained a PDD-NOS diagnosis through the age 9 assessment.

In order to test this hypothesis, we ran a model of RRB total score with diagnostic change from initial to most recent assessment and its interaction with age and age-squared as predictors. Very few children changed from a nonspectrum to an ASD diagnosis or vice versa (see Lord et al., 2006). Thus, for the diagnostic change variable, only four subgroups of interest were included (see Figure 3.3): children who maintained

an autism diagnosis ($n = 86$), children who switched from autism to PDD-NOS ($n = 15$), children who maintained a PDD-NOS diagnosis ($n = 23$), and children who switched from PDD-NOS to autism ($n = 29$). There were no main effects of diagnostic change. However, as expected, there was a significant interaction between age and diagnostic change. Not surprisingly, children who maintained an autism diagnosis showed more of an increase in RRB scores over time than children who maintained a PDD-NOS diagnosis, $t(351) = 2.51, p < .05$. More interesting, and consistent with our prediction, was the finding that children who switched from PDD-NOS to autism showed more of an increase in scores over time than children who maintained a PDD-NOS diagnosis, $t(350) = 2.05, p < .05$. In contrast, there was no difference in trajectories between the group who maintained a PDD-NOS diagnosis and those who switched from autism to PDD-NOS. As seen in Figure 3, both groups who had PDD-NOS as their most recent diagnosis followed negative trajectories, indicating improving scores over time.

When we ran Model 4 (see Table 3.3) replacing initial diagnosis with most recent diagnosis, there was a significant interaction between diagnosis and age. Interestingly, the difference was between autism and PDD-NOS, with children with autism showing more of an increase in scores than children with PDD-NOS, $t(467) = 2.14, p < .05$, but not children with DD.

Summary

In sum, RRB total scores at age 2 were associated with diagnostic status (ASD vs. NS) and NVIQ at age 2. When diagnosis was controlled for, SA score at age 2 was not significantly associated with RRB scores at age 2. Change in RRB scores over time was also associated with NVIQ at age 2; children with lower NVIQ scores at the first assessment tended to show greater increases in RRB total scores over time than children with higher NVIQ scores. In contrast, change in RRB scores was not strongly associated with diagnosis at 2. This was partly due to fact that many children with PDD-NOS changed to a diagnosis of autism in subsequent assessments.

The random effects at the bottom of Table 3.3 show a clear reduction in the unexplained variance of the intercepts, by about 30%. The overall reduction in the

variance of the slopes was more modest (about 10%) and appeared to be due largely to the addition of the interaction between NVIQ and age in Model 4. Therefore, NVIQ at age 2 explained some of the variability in individual slopes. However, there was relatively little variance in the slopes to begin with.

Patterns of Change among Children with ASD

The analyses thus far highlight the variables that predicted initial scores and patterns of change over time in children with ASD and DD. However, they do not provide a detailed picture of the different patterns of change. In order to address this issue, we used Proc Traj (see Jones et al., 2001). We included only children ASD (i.e., PDD-NOS or autism) at age 2, since the previous analyses indicated that RRB scores were generally very low for children who were not on the autism spectrum at age 2.

First, we compared different models for RRB total score. The five-group model fit the data best, based on the BIC. The average probability of being in a given group ranged from 79% to 91%. Evidence of good fit can be seen in Figure 3.4.1, where discrepancies between observed scores (solid lines) and expected scores (dashed lines) are minimal. This solution yielded five groups that will henceforth be referred to as ‘consistently mild’ ($n = 25$); ‘worsening’ ($n = 22$); ‘consistently moderate’ ($n = 84$); ‘improving’ ($n = 13$); and ‘consistently severe’ ($n = 17$).

When the same analyses were run using number of items instead of RRB total score, the three-group solution yielded the best fit (see Figure 3.4.2). The groups could best be described as having consistently few RRBs ($n = 25$), slightly increasing RRBs ($n = 90$), and consistently many RRBs ($n = 46$).

Table 3.4 shows how the five groups from the analysis of RRB scores mapped onto the three-group solution for total number of items. Of the 28 children with few RRBs, 24 (86%) had consistently mild RRB total scores and the remaining children had moderate scores. The group with moderate/increasing numbers of RRBs included 86 children and was comprised primarily children with moderate RRB total scores (83%) with the remaining children falling into the worsening score group. Of the 43 children with many RRBs, the largest proportion (20%) had consistently severe scores, as would

be expected. However, the next largest subgroup (26%) was comprised of children who had improving scores over time. The remaining children were either in the consistently moderate score group (16%) or the worsening score group (19%). Thus, just over one-quarter of children who maintained many RRBs over time nevertheless showed improvement in scores, indicating that behaviors became less severe over time.

Risk factors affecting likelihood of group assignment

Next, we considered ‘risk factors’ that might affect a child’s likelihood of following a particular trajectory for RRB total score. Diagnosis, ADOS SA score, and NVIQ score at age 2 were entered into the model. We also included the child’s gender, race, and recruitment site, and the mother’s level of education as covariates. Having a higher NVIQ score significantly decreased the likelihood of being in the ‘worsening’ group ($\beta = -.06, se .03, p < .05$), the ‘moderate’ group ($\beta = -.04, se .02, p < .05$), and the ‘consistently severe’ group ($\beta = -.08, se .03, p < .01$) relative to the ‘consistently mild’ group. Converting the estimate to an odds ratio, each one-point increase in NVIQ decreased the likelihood of being in the highest-scoring group by 8%. The average NVIQ score at age 2 was 54.8 in the highest-scoring group and 78.1 in the lowest-scoring group. None of the other covariates was a significant risk factor.

We wondered if diagnosis and SA score had not emerged as significant risk factors because they were closely related to each other, as well as to NVIQ score, which was also in the model. A t-test confirmed that the mean SA score at age 2 in the autism subgroup ($M = 16.1, sd = 2.1$) was significantly higher than the mean SA score in the PDD-NOS subgroup ($M = 11.4, sd = 4.0$), $t(75.8) = 8.38, p < .001$. The mean NVIQ score for the autism subgroup ($M = 62.3, sd = 17.0$) was significantly lower than for the PDD-NOS group ($M = 72.5, sd = 22.4$), $t(97.1) = -3.03, p < .01$. SA and NVIQ scores at age 2 were also significantly related, ($r = .32, p < .001$). The negative correlation indicates that children with lower NVIQ scores were more socially impaired.

We therefore re-ran the model above twice, first with diagnosis at age 2 as a risk factor, dropping SA and NVIQ, and the second time with SA score at age 2 as a risk factor, dropping diagnosis and NVIQ. Diagnosis was a significant risk factor in this

reduced model; children with autism were significantly more likely than children with PDD-NOS to be in the worsening group, the moderate group, and the consistently severe group, relative to the consistently mild group. To illustrate, having autism made a child over 5 times more likely to be in the highest-scoring group than the lowest-scoring group. However, SA score was not a significant risk factor, even after dropping NVIQ and diagnosis.

RSM and IS Behaviors

In addition to examining RRBs as a category, one of the aims of this study was to examine different subtypes of RRBs and determine if they followed different patterns of change over time. Our examination of subtypes was based on previous findings that RRB items on the ADI-R tend to cluster into a repetitive sensorimotor (RSM) factor and an insistence on sameness (IS) factor. The specific behaviors that have loaded on each factor have varied somewhat among different studies, but most behaviors have consistently loaded on one of the two factors: for the RSM factor, these include **repetitive use of objects, unusual sensory interests, hand and finger mannerisms, and complex mannerisms**; and for the IS factor, they are **compulsions and rituals, difficulties with changes in routine, and resistance to trivial changes in environment**.

Before doing any analyses of RSM and IS behaviors, it was necessary to determine if we obtained these factors for each cohort in our sample. A previous paper using the age 2 cohort (see Richler et al., in press) found that these factors emerged in the youngest group. A confirmatory factor analysis was run to see if these factors also emerged in the other cohorts. As in previous studies, the cutoff for including an item on a factor was ≥ 0.30 . Factor loadings are reported in Table 3.5. Loadings were consistently high for the RSM factor, ranging from 0.49 to 0.87. For the IS factor, there was somewhat more variability, with loadings ranging from 0.47 to 1.00, with one loading of .30, for compulsions and rituals at age 3.

As with the RRB category, we examined factors by looking both at total score and total number of items within a factor. Thus, the child's RSM score was the sum of their

scores on the items comprising the RSM factor, and the child's IS score was the sum of their scores on the IS factor.

Predictors of change in children with ASD and DD

The same series of models described above for RRB total scores was run on total RSM score and total IS score. Results were similar when the number of RSM/IS items was used as the outcome instead of scores; therefore, only results for total scores are reported here. Analyses were run on the entire sample (i.e., including the DD children).

In the 'full' model (i.e., Model 4 above, with main effects and interactions), age was significantly related to RSM scores, but in a negative direction, indicating that scores decreased with age. There was a strong main effect of diagnosis at age 2. Children with autism had an average RSM score of 4.87 at intercept, compared to 3.56 for PDD-NOS and 2.20 for nonspectrum DD. The difference between the autism and PDD-NOS groups was significant $t(259) = 3.12, p < .01$, as was the difference between PDD-NOS and DD, $t(261) = 2.88, p < .01$. NVIQ at 2 had a significant negative main effect, $t(266) = -2.84, p < .01$, indicating that as NVIQ scores increased, RSM scores decreased. There was also a significant interaction between NVIQ and age; children with higher NVIQ scores at age 2 showed more of a decrease in scores over time compared to children with lower NVIQs, $t(466) = -3.11, p < .01$. As in the analysis of total scores, there were no significant effects for SA score at age 2. However, as in the previous analyses, when diagnosis at 2 was omitted, there was a significant effect of SA, such that RRB scores increased as SA scores increased (i.e., became more abnormal), $t(268) = 4.17, p < .001$. Also as in the previous analysis, there were no linear effects for diagnosis at age 2 (see Figure 3.5.1), but when a model was run with the diagnostic change variable described above, there was a significant interaction. Children who maintained an autism diagnosis or switched from PDD-NOS to autism exhibited a greater increase in scores over time than children who maintained a PDD-NOS diagnosis or switched from autism to PDD-NOS, $F(3,357) = 4.49, p < .01$. There was no difference in slopes for the two groups whose most recent diagnosis was autism, and the same was true for the two groups whose most recent diagnosis was PDD-NOS.

Because IS scores were negatively skewed, it was necessary first to perform a log transformation. In the ‘full’ model, age was significantly and positively related to IS score, $t(489) = 3.80, p < .001$, indicating that scores tended to increase with age. There was also a significant negative quadratic effect, $t(494) = -3.35, p < .001$, indicating that the rate of increase slowed as children got older. Converting from the logarithmic scores, actual intercepts were .59, .54, and .39 for the autism, PDD-NOS, and DD groups, respectively. The difference between the autism and DD groups was significant, $t(255) = -2.88, p < .01$. Unlike for total scores and RSM scores, there was not a significant effect of NVIQ at age 2 on intercepts or rates of change. There was a significant main effect of ADOS SA score at age 2. Interestingly, children with higher SA scores, indicating a greater degree of social impairment, had lower IS scores at age 2 than children with lower SA scores, $t(257) = -2.57, p < .05$. As in previous analyses, diagnosis at age 2 was not a predictor of trajectories of change in scores (see Figure 3.5.2.) However, unlike in previous analyses, diagnostic change did not have a significant effect on patterns of change in RRB scores over time; slopes were similar across groups. Also interesting was the finding that when diagnosis at age 2 was removed from the analysis, the main effect of SA score disappeared, but there was a significant interaction between SA score and age, indicating that IS scores increased more for children with lower SA scores (i.e. milder social impairment) than those with higher SA scores, $t(480) = 2.64, p < .01$.

Patterns of change among children with ASD

As for total scores, patterns of change in RSM and IS scores were examined using Proc Traj to assess the degree of variability among children with ASD. For RSM score, a three-group linear solution provided the best fit (see Figure 3.6.1). This solution yielded a consistently mild group ($n = 41$), a slightly increasing group ($n = 75$), and a consistently severe group ($n = 45$).

When we added covariates, as above, NVIQ score at age 2 emerged as a significant risk factor. With every one-point increase in NVIQ score, the likelihood of assignment to the mild group relative to the severe group increased by approximately 8% and the likelihood of assignment to the increasing group relative to the severe group

increased by approximately 5%. Having a higher NVIQ also increased the likelihood of assignment to the slightly increasing group relative to the severe group, by approximately 5% per one-point increase. The average NVIQ score was 75.0 for children in the mild group, 69.2 for children in the increasing group and 47.9 for children in the severe group. Diagnosis was a significant risk factor, even when NVIQ at age 2 was included in the model. Children with PDD-NOS were over 90% less likely than children with autism to be in the severe group. SA score was not significant in this model. However, when the model was run dropping NVIQ and diagnosis at age 2, SA was a significant risk factor. Having a higher SA score, indicating greater social impairment, increased the likelihood of being in the severe group relative the mild group. Each one-point increase in SA score increased the risk of being in the severe group by 24%.

When Proc Traj was run with the number of RSM items as the predictor, the two-group solution provided the best fit. One group had consistently few RSM behaviors, starting at a mean of about 1 in younger children and decreasing slightly as children got older. The other group had several RSM behaviors that remained constant over time, at a mean of approximately 3. When these groups were cross-tabulated with those from the analysis of RSM scores, children in the consistently mild scoring group were split approximately evenly among the 2 RSM item groups, with 22 of 38 falling in the ‘few RRBs’ group and the remaining 16 falling in the ‘many RSM behaviors’ group. All of the children in the slightly decreasing RSM score group were classified in the ‘many RSM behaviors’ group. Thus, even though scores improved slightly for these children, they continued to have a relatively high number of RSM behaviors. Likewise, all of the children in the increasing group fell into the group with many RSM behaviors.

For IS score, the three-group model provided the best fit of the linear models (see Figure 3.6.2), yielding a mild group ($n = 20$), an increasing group ($n = 116$), and a moderate group ($n = 25$). On average, IS scores in the middle group increased between ages 2 and 5, from approximately 0 to close to 2.

We ran Proc Traj using number of IS behaviors, and the two-group quadratic solution fit best. The groups had similar patterns of change, with the sharpest increases in the number of IS items occurring up until approximately 60 months of age. In

‘mild/increasing’ group, the average number of items at the initial assessment was close to 0, and increased to 1 by the final assessment. In the ‘moderate/increasing’ group, the number of IS items started at approximately 1 and increased to about 2.

As would be expected, all of the children in the mild IS score group fell into the mild/increasing IS item group. Of the 51 children in the increasing score group, 49 (96%) were categorized as having mild/increasing number of IS items. Finally, 57 of 70 children (81%) in the moderate IS score group fell into the group that had moderate/increasing numbers of IS items.

Diagnosis was significant risk factor for IS scores, controlling for NVIQ and SA scores at 2. Having a diagnosis of PDD-NOS significantly decreased the likelihood of being in the highest-scoring group. Approximately 45% of the children in this group had PDD-NOS, whereas almost 70% of the children in the mildest group had PDD-NOS. SA score was not a significant risk factor in this model. However, when NVIQ and diagnosis at age 2 were dropped, having a higher SA score decreased the likelihood of being in the highest-scoring group. Children in this group had an average SA score of 8.4, compared to 14.6 and 11.9 for the mild and increasing groups, respectively. Thus, having milder social impairment was associated with following a more severe trajectory for IS behaviors.

Discussion

The findings from the present study add to the existing evidence that there are distinct subtypes of RRBs in ASD. First, the variables that predict patterns of change over time in children with ASD are different for the two subtypes. Second, the developmental patterns themselves are different. These findings, building on those of other investigators, call into question a simple conceptualization of RRBs as a single entity. Given the striking differences in findings for the two types of behaviors, careful consideration of the implications of these differences is warranted.

Differences in Predictors of Change

Our findings indicate that a relatively high NVIQ and mild social impairment are protective against developing RSM behaviors. The same is not for IS behaviors, which are relatively independent of NVIQ, and are actually more common in children with mild social impairment. Several studies have found that IS behaviors are not related to IQ (e.g., Cuccaro et al., 2003). Given that IS behaviors are present in children with ASD across the entire range of cognitive functioning, it is not surprising that children's RRBs, but not their NVIQ scores, have been found to be associated with perceived negative impact in parents of children with ASD. These findings have important clinical implications. Low cognitive functioning at an early age is a poor prognostic indicator for RSM behaviors. In contrast, having relatively mild social impairment at a young age might actually add to the risk of having IS behaviors later on. Clinicians making prognoses about outcome should bear these findings in mind; a relatively high-level of functioning seems to be a protective factor only for certain RRBs. Thus, a young child with relatively mild social impairments can be expected to acquire some RRBs in later years, although these behaviors may be different from those of a lower-functioning child.

The predictive value of early RRBs also differs for RSM and IS behaviors. Children who have many and/or severe RSM behaviors are likely to continue to show this profile, and children who have relative few and/or mild RSM behaviors are likely to maintain this milder profile. In contrast, early IS behaviors are not as informative for making prognoses about future IS behaviors, as many children have very few or none of these behaviors at young ages, but acquire them as they get older. This information may be useful to parents who want to know whether their children will continue to show 'unusual' behaviors as they get older.

Differences in Patterns of Change

The findings about patterns of change in RRB scores highlight the high degree of variability among children with ASD. This is consistent with the considerable phenotypic heterogeneity seen in other core domains of the disorder. For those trying to identify genes associated with ASD, this degree of variability can be problematic. Studies have stratified

samples according to characteristics such as IQ and language acquisition in order to identify subgroups that are relatively homogeneous phenotypically (see Hus, Pickles, Cook, Risi, & Lord, 2007). More recently, RRBs have been used to identify phenotypes (Brune et al., 2006). Looking at specific subtypes of RRBs might further this endeavor. As in the present study, Hus et al. (2007) found that IS behaviors were relatively independent of IQ and diagnosis, and suggested that this group of behaviors might be useful for identifying ASD phenotypes, precisely because they represent a feature of the disorder that is relatively independent from other features.

The findings about change in IS scores over time in the present study suggest that development must also be taken into account when considering phenotypes. Most children with ASD had relatively low IS scores at young ages, but as children got older, trajectories began to diverge, with some children continuing to have low scores, and others increasing. It might be useful to define phenotypic groups based on patterns of change, for example in IS behaviors, rather than on the presence or severity of these behaviors at a single point in development.

Taken together, the strikingly different findings for RSM and IS behaviors provide further support for the idea that there are two distinct kinds of RRBs: one subtype is comprised of repetitive, sensory and/or motor-related behaviors, and the other of adherence to particular routines or rituals, or an insistence that things be ‘just so.’ To some extent, these subtypes map onto what have been termed ‘higher order’ and ‘lower order’ RRBs (Turner, 1999). RSM behaviors are clearly associated with lower levels of functioning, in terms of NVIQ, language level, and autism severity. It is not as clear whether IS behaviors are ‘higher order,’ as they do not appear to be strongly related to IQ. However, the fact that, in the present study, there was an inverse relationship between social impairment and IS behaviors supports the idea that these behaviors are associated with higher functioning.

However, the fact that we found a relationship between milder social impairment and more and/or more severe IS behaviors does not necessarily mean that these behaviors are uncommon in lower functioning children. Perhaps it is simply that IS behaviors are easier to identify in children who are more socially aware and have functional language, since these children are better able to convey when they are distressed by some change in a routine or

ritual. In contrast, a child with no functional language and marked difficulty relating to others might be upset by similar changes, but might not know how to convey this to someone else (Geschwind & Levitt, 2007).

Methodological Considerations

The findings on differences in RSM and IS behaviors help explain why results in previous studies of RRB development have been mixed. How RRBs change over time clearly depends, in part, on the kind of behavior. In the present study, we started by looking at total scores in order to see if we obtained results similar to those of other studies. Like other studies, we found some evidence for increases in RRB scores over time. However, given that RSM behaviors developed so differently from IS behaviors, looking only at how total scores changed over time would not have provided an accurate picture of RRB development. Researchers should consider this issue when deciding how to analyze RRB data. There might be instances in which analyzing total scores is useful, such as when trying to make distinctions between children with ASD and children with nonspectrum disorders using a relatively general measure of RRBs. Decisions about how RRBs are measured and analyzed should be based on the types of questions being asked.

In a similar vein, looking at changes over time in the number of RRBs provided information that could not have been obtained by looking only at total scores. Although the number of behaviors children had tended to follow patterns similar to their scores, there was a notable exception: many children who maintained a relatively high number of RSM behaviors still showed improvements, as indicated by decreasing RSM scores, nevertheless. This is encouraging, as it suggests that behaviors can improve, even if they do not completely disappear.

Although decisions about how to measure and analyze RRBs strengthened our findings, they necessarily presented some constraints. For example, scores were higher for RSM behaviors than for IS behaviors. For children in the highest RSM group, scores reached a peak of approximately 7 out of a maximum of 11, while in the highest IS group, scores only reached a peak of less than 2 out of a maximum of 9. The difference in numbers of behaviors relative to the maximum number possible was not as striking. One might conclude from this

finding that RSM behaviors are more severe in children with ASD than IS behaviors. It is difficult to directly compare severity across items, however, because codes are written differently depending on the behavior. For example, to receive a score of 2 on the ADI-R IS items, the child must clearly experience distress (e.g. crying if the furniture in the living room is rearranged). In contrast, to receive a score of 2 on the RSM items, the child has to spend a substantial amount of time engaged in the behavior, but does not have to display distress if interrupted. The reason why fewer children received scores of 2 on the IS items could therefore be that these items have a higher ‘threshold.’ Another reason why scores were higher for RSM items could be that different aspects of a single behavior can be coded in different RSM items. For example, a child who likes to spin the wheels repeatedly on a toy car and watches the wheels very closely as they spin would be coded under both repetitive use of objects, for the spinning, and under unusual sensory interests, for the close visual inspection. In contrast, there might be less overlap between the IS items; if a child insists on carrying out a particular ritual, such as touching things in a certain order, this should only be coded under compulsions and rituals and not under difficulties with changes in routines in routine. As a result, it might be easier for a child to obtain a high combined score on RSM items than on the IS items on the ADI-R.

Limitations and Future Directions for Research

Children were first recruited into this study at a time when early diagnosis of ASD was relatively uncommon. For this reason, our sample might not be representative of young children currently referred for a diagnosis of ASD. Presumably, given the increased awareness of ASD and ability to recognize milder variants, the children in the present study may have had more severe symptoms, on average, than children referred today. It is important that longitudinal studies continue, so that we have more up-to-date information on development in ASD.

Another limitation of the present study was that, although the rate of attrition was relatively low, it was higher in families with lower socioeconomic status. It is possible that we might have found more significant effects for demographic variables if we had been able to continue following these families.

Findings in the present study are based on a parent report measure. Using parent report has some advantages, in that it allows the collection of data about behaviors that might not be observed in a short assessment. On the other hand, parent report is necessarily subjective, which can be problematic. Having clinicians who are experienced in administration and coding helps elicit accurate descriptions of behaviors. Nevertheless, it is important to corroborate the findings from the present study with data obtained from other measures, particular those that involve direct observation.

A final caveat about the findings presented here is that they do not account for the effects of treatment on the development of RRBs. It is crucial that we determine whether treatment can reduce RRBs, and if so, which methods are most effective. Because these behaviors can significantly interfere with both the child's and the family's functioning, parents want to know what can be done to address them. To date, little research has directly addressed this question. There is some evidence that RRBs can be reduced by increasing social demands in the child's environment (Clark & Rutter, 1981). Behavioral techniques (e.g. use of reinforcers) have also been effective in decreasing RRBs such as stereotypies (see Horner, Carr, Strain, Todd, & Reed, 2002), and there is some evidence that pharmacological treatments can decrease compulsive behaviors (Hollander, 2005). Given the findings from the present study, it would be useful to investigate whether the effectiveness of treatments for RRBs depends on characteristics of the behavior and the child.

Although the results from this study provided new information on RRB development, the way in which these behaviors change over time in children with ASD can be examined even more closely. In addition to looking at subtypes of RRBs, it is also crucial to consider how individual behaviors change over time. Is it the case that, once a child acquires a given behavior, s/he is likely to continue to have that behavior in older years, or are some behaviors commonly lost? Conversely, if a child does not exhibit a given behavior at a young age, does that mean s/he won't acquire it later on? Are 'lower order' behaviors in young children replaced by 'higher order' behaviors as they get older? These questions are addressed in Chapter IV.

The findings from the present study suggest that RRBs in ASD are complex and

deserve the same careful attention that the other core domains of ASD have received. Attention to issues of development and behavior subtypes will enhance our overall understanding of RRBs, thereby filling in some of the pieces of the ASD puzzle.

Table 3.1. Sample Demographics by Wave

	age 2	age 3	age 5	age 9
Total n (children who received ADI)	214	184	136	172
Gender				
Male	172 (80%)	155 (84%)	106 (78%)	138 (80%)
Female	42 (20%)	29 (16%)	30 (22%)	34 (20%)
Ethnicity				
Caucasian	143 (67%)	122 (66%)	81 (60%)	122 (71%)
African American	67 (31%)	58 (32%)	54 (40%)	46 (27%)
Other	4 (2%)	4 (2%)	1 (<1%)	4 (2%)
Maternal Education				
Graduate/Professional	33 (15%)	28 (15%)	15 (11%)	30 (17%)
College Degree	40 (19%)	36 (20%)	22 (16%)	38 (22%)
Some College	63 (29%)	55 (30%)	45 (33%)	46 (27%)
High School	55 (26%)	44 (24%)	38 (28%)	42 (24%)
Less than High	14 (7%)	13 (7%)	14 (10%)	11 (6%)
Diagnosis*				
Autism	102 (48%)	n/a	68 (50%)	100 (58%)
PDD-NOS	59 (26%)	n/a	29 (21%)	35 (20%)
Nonspectrum	53 (25%)	n/a	39 (29%)	37 (22%)
Site				
North Carolina	133 (62%)	106 (58%)	125 (92%)	104 (60%)
Chicago	81 (38%)	78 (42%)	11 (8%)	68 (40%)
Mean Age at ADI	28.9 (5.2)	42.7 (6.0)	57.0 (8.8)	112.3 (15.4)

Note Best estimate diagnoses were made only at the age 2, age 5, and age 9 assessments.

Table 3.2. Examples of RRBs on the ADI-R

RSM Behaviors	Unusual sensory interests	Peering at objects from the side
	Repetitive use of objects	Lining up toys
	Hand/finger mannerisms	Flicking/twisting fingers
	Complex body mannerisms	Spinning in circles
IS Behaviors	Difficulties with minor changes in routine	Insisting on always sitting in the same seat in the car
	Resistance to trivial changes in the environment	Becoming upset when a family member gets a haircut
	Compulsions and rituals	Insisting on always turning right out of the driveway
'Other' Behaviors	Self-injury	Banging head
	Sensitivity to noise	Becoming distressed by the sound of a vacuum cleaner
	Unusual preoccupations	Having an intense interest in rocks or strings

Table 3.3. Growth Models for Changes in RRB Total Scores from Age 2 to Age 9

<u>Predictors</u>	<u>Model 1</u>	<u>Model 2</u>	<u>Model 3</u>	<u>Model 4</u>
	Coefficient (S.E.)	Coefficient (S.E.)	Coefficient (S.E.)	Coefficient (S.E.)
Fixed Effects				
Intercept	6.08*** (.29)	7.73*** (.62)	7.18*** (.74)	7.59*** (.87)
Age at Testing	.03* (.01)	.05** (.02)	.06 ** (.02)	.04 (.03)
Age-squared	-0.002 (-.0001)	-.0004 * (.0002)	-.00* (.00)	-0.00 (.00)
Age 2 Diagnosis				
Autism		--	--	--
PDD-NOS		-1.12 (.65)	-.40 (.79)	-1.03 (.84)
Nonspectrum		-3.85*** (.71)	-3.20** (1.03)	-3.92** (1.17)
Gender				
Female		-.46 (.62)	-.71 (.61)	-.69 (.61)
Site				
North Carolina		.06 (.55)	-.36 (.55)	-.44 (.55)
Race				
Non-White		-.42 (.55)	-.63 (.53)	-.61 (.53)
Maternal Education				
Graduate or college degree		-.37 (.54)	-.11 (.51)	-.15 (.51)
NVIQ at 2			-.05 *** (.01)	-.03** (.02)
ADOS Social Affect			-0.06 (.07)	-0.09 (.10)
Algorithm Score				
Linear Slopes:				
Age*NVIQ				-.00* (.00)
Random Effects				
Intercept variance	12.52*** (1.61)	9.68*** (1.37)	9.36***(1.44)	8.93*** (1.38)
Slope variance	.001*** (.0003)	.001***(.0003)	.00***(.00)	.00**(.00)
Intercept/slope covariance	-0.02 (.02)	-0.02 (.01)	-0.04 *(.01)	-0.03 (.02)

***p < .001 ** p < .01 *p < .05

Note Due to space constraints, only significant linear effects are shown. None of the quadratic effects was significant and are therefore not shown.

Figure 3.1. Predicted RRB Total Scores by Diagnosis at Age 2

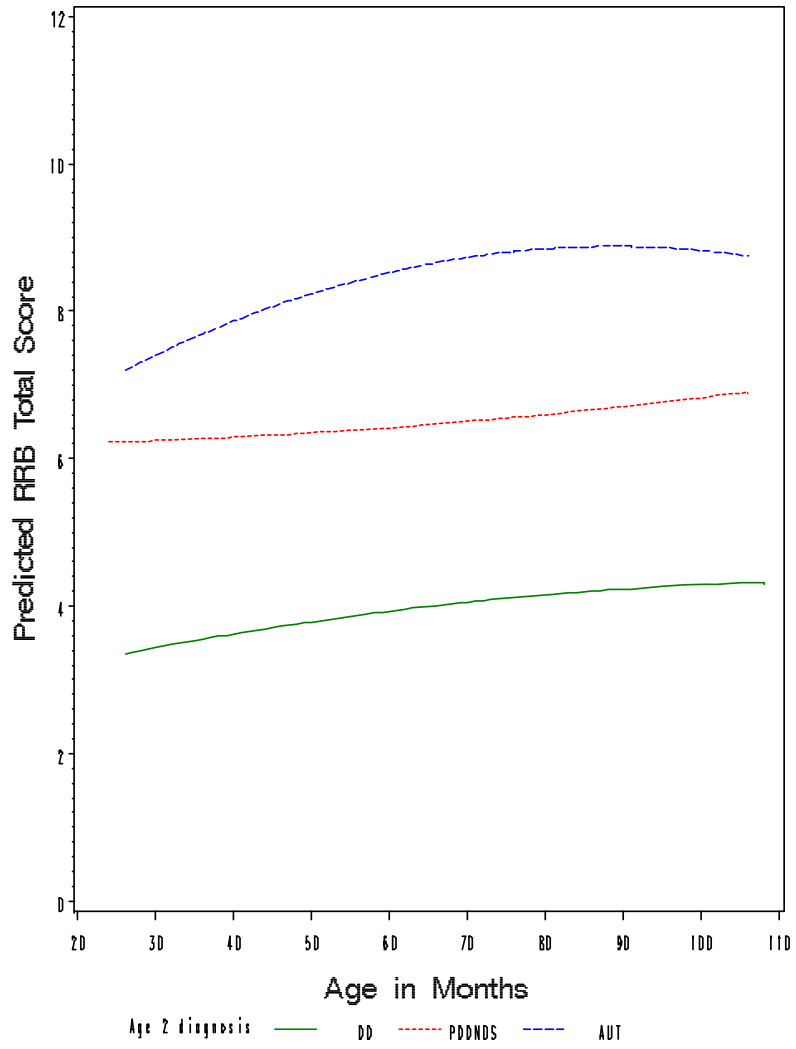


Figure 3.2. Predicted RRB Total Scores by NVIQ at Age 2

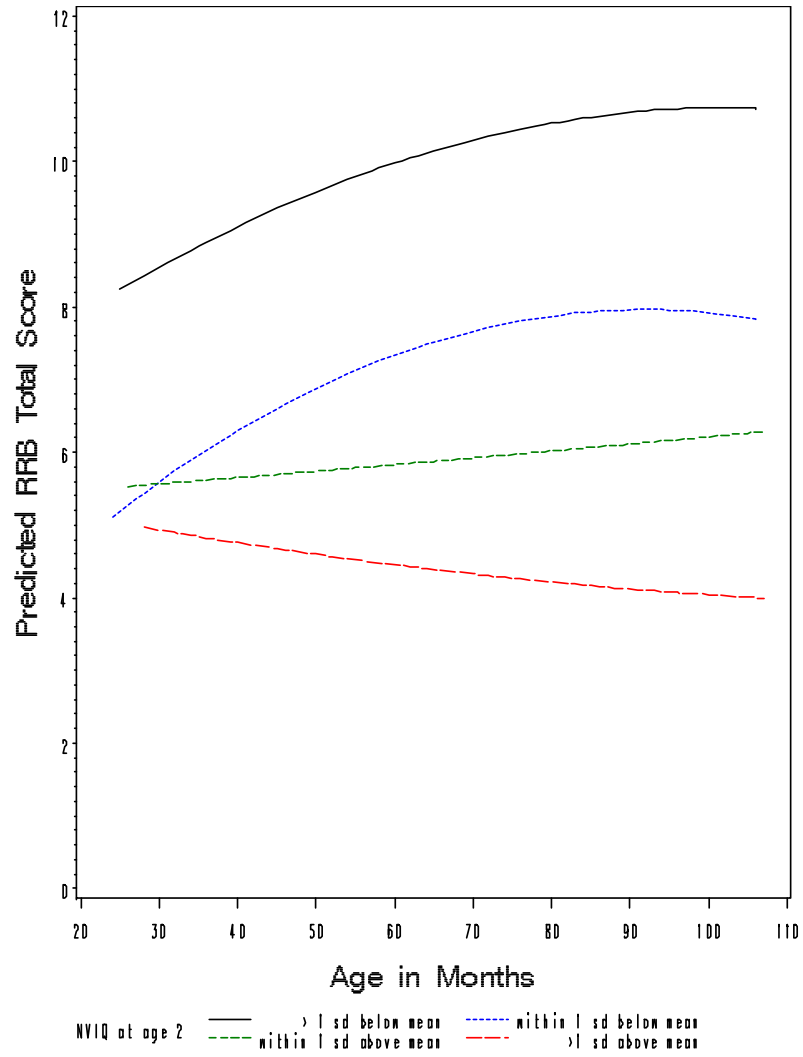


Figure 3.3 Predicted RRB Total Scores by Diagnostic Change from Initial to Most Recent Assessment

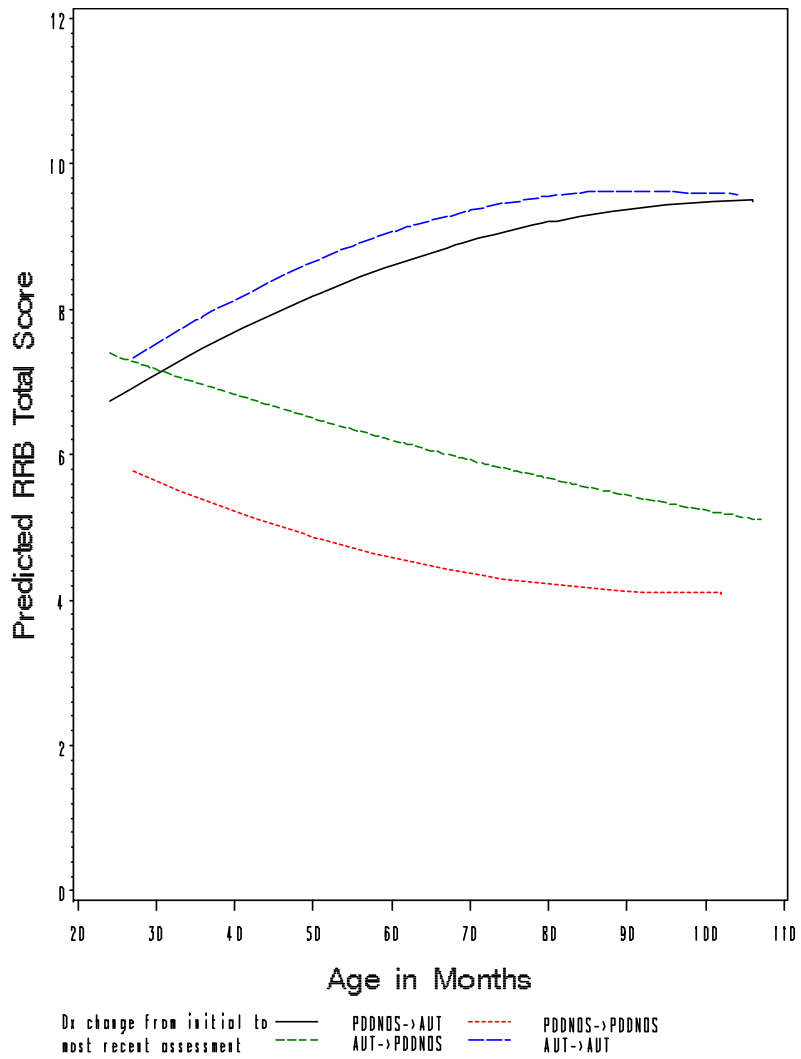
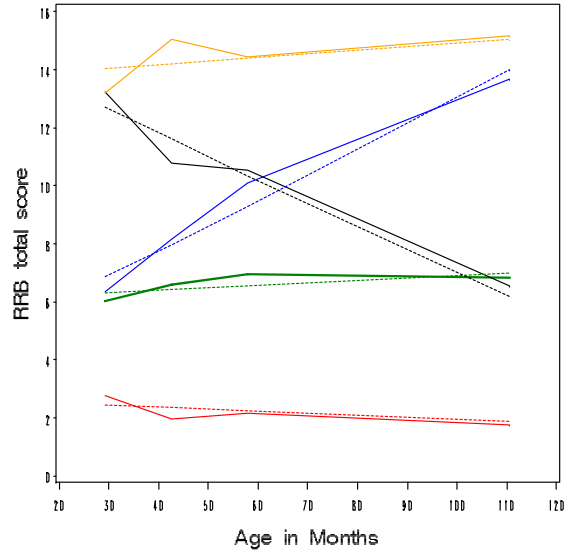


Figure 3.4. Patterns of Change in RRB Totals for ASD only

1. Five-group solution for RRB total scores



2. Three-group solution for total number of RRB items

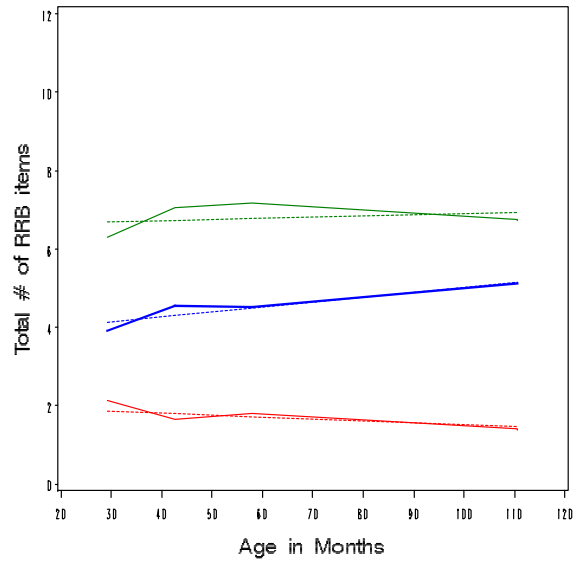


Table 3.4. Cross-Tabulation of Five-Group Solution for RRB Total Scores and Three-Group Solution for Total Number of RRBs

		groups for total RRB items		
		few RRBs	increasing RRBs	many RRBs
Groups for total RRB score	mild	24 (86%)	0 (0%)	0 (0%)
	worsening	0 (0%)	13 (17.1%)	8 (18.6%)
	moderate	4 (14%)	63 (82.8%)	7 (16.3%)
	improving	0 (0%)	0 (0%)	11 (25.6%)
	severe	0 (0%)	0 (0%)	17 (39.5%)

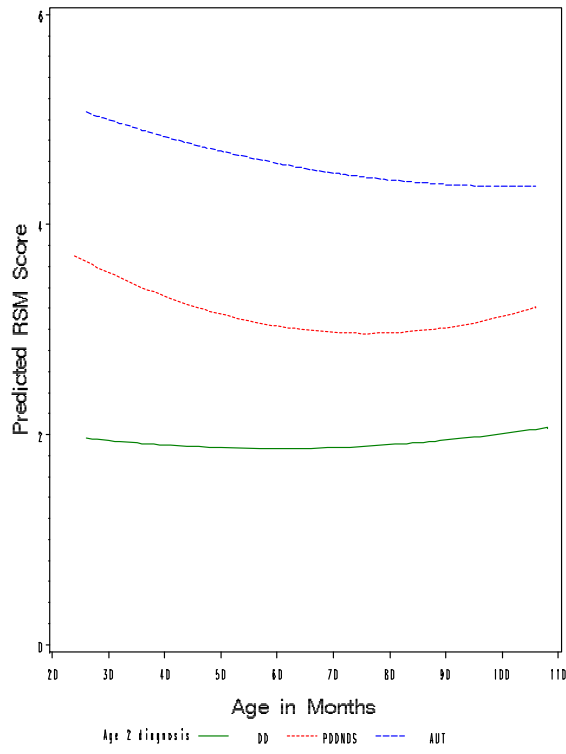
Note Percentages are expressed as a proportion of each of the three item groups.

Table 3.5. Factor Loadings of ADI-R RRB Items by Wave

	RRB item	Age 2	Age 3	Age 5	Age 9
RSM factor	Repetitive use of objects	.81	.74	.72	.73
	Unusual sensory interests	.67	.81	.75	.70
	Hand/finger mannerisms	.55	.60	.87	.73
	Other complex mannerisms	.49	.70	.75	.72
IS factor	Resistance to trivial changes in environment	.91	.77	1.00	.78
	Difficulties with changes in routine	.80	.56	.67	.75
	Compulsions and rituals	.62	.30	.62	.47

Figure 3.5. Predicted RSM and IS scores by Diagnosis at Age 2

1. RSM score



2. IS score

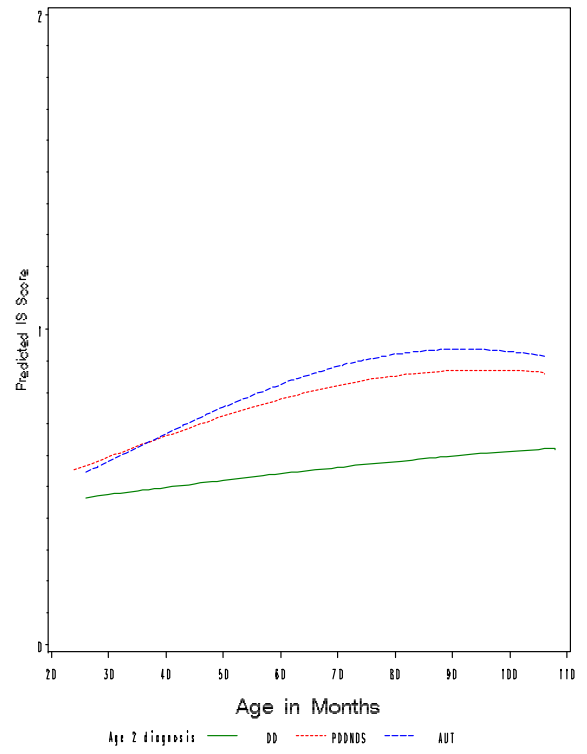
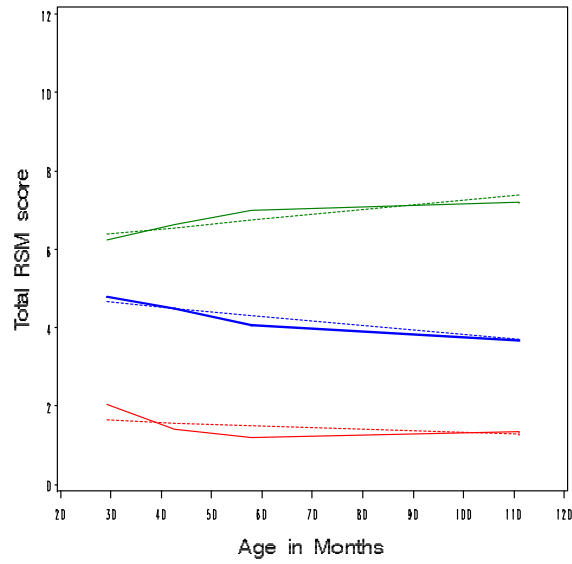
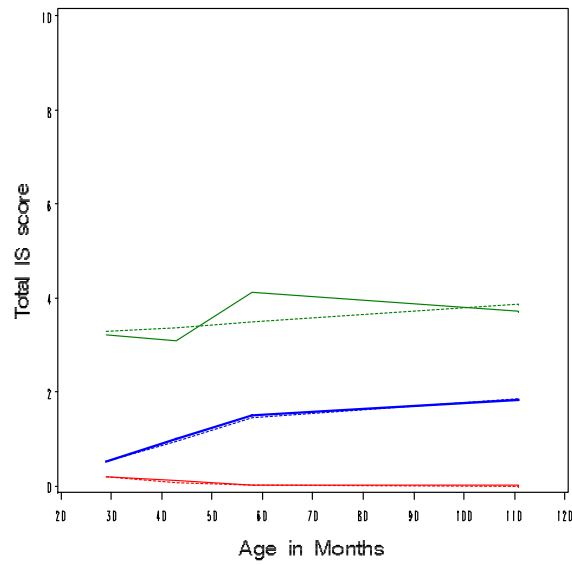


Figure 3.6. Patterns of Change in RSM and IS Scores for ASD only

1. Three-group linear solution for RSM score



2. Three-group quadratic solution for IS score



References

- American Psychiatric Association. (1987). *Diagnostic and statistical manual of mental disorders* (3rd, revised ed.). Washington, D.C.: American Psychiatric Association.
- American Psychiatric Association. (1994). *Diagnostic and statistical manual of mental disorders* (4th ed.). Washington, D.C.: American Psychiatric Association.
- Anderson, D., Lord, C., Risi, S., DiLavore, P., Thurm, A., Shulman, C., & Pickles, A. (in press). Patterns of growth in verbal abilities among children with autism spectrum disorder. *Journal of Clinical Child Psychology*.
- Bishop, S. L., Richler, J., & Lord, C. (2006). Restricted and repetitive behaviors and nonverbal IQ in children with autism spectrum disorders. *Child Neuropsychology*, *12*, 247-267.
- Brune, C. W., Kim, S. J., Salt, J., Leventhal, B. L., Lord, C., & Cook, E. H. (2006). 5-HTTLPR genotype-specific phenotype in children and adolescents with autism. *American Journal of Psychiatry*, *163*, 2148-2156.
- Bryson, S. E., Zwaigenbaum, L., Brian, J., Roberts, W., Szatmari, P., Rombough, V., & McDermott, C. (2007). A prospective case series of high-risk infants who developed autism. *Journal of Autism and Developmental Disorders*, *37*, 12-24.
- Butter, E. M., Mulick, J. A., & Metz, B. (2006). Eight case reports of learning recovery in children with pervasive developmental disorders after early intervention. *Behavioral Interventions*, *21*, 227-243.
- Carcani-Rathwell, I., Rabe-Hasketh, S., & Santosh, P. J. (2006). Repetitive and stereotyped behaviours in pervasive developmental disorders. *Journal of Child Psychology and Psychiatry*, *47*, 573-581.
- Carter, C. S. (2007). Sex differences in oxytocin and vasopressin: Implications for autism spectrum disorders? *Behavioural Brain Research*, *176*, 170-186.
- Charman, T., Taylor, E., Drew, A., Cockerill, H., Brown, J. A., & Baird, G. (2005). Outcome at 7 years of children diagnosed with autism at age 2: predictive validity of assessments conducted at 2 and 3 years of age and pattern of symptom change over time. *Journal of Child Psychology and Psychiatry*, *46*, 500-513.
- Clark, P., & Rutter, M. (1981). Autistic children's responses to structure and to interpersonal demands. *Journal of Autism & Developmental Disorders*, *11*, 201-217.
- Cox, A., Klein, K., Charman, T., Baird, G., Baron-Cohen, S., Swettenham, J., Drew, A., Wheelwright, S., & Nightingale, N. (1999). Autism spectrum disorders at 20 and 42 months of age: Stability of clinical and ADI-R diagnosis. *Journal of Child Psychology and Psychiatry*, *40*, 719-732.
- Cuccaro, M. L., Shao, Y., Grubber, J., Slifer, M., Wolpert, C. M., Donnelly, S. L., Abramson, R. K., Ravan, S. A., Wright, H. H., DeLong, R. G., & Pericak-Vance, M. A. (2003). Factor analysis of restricted and repetitive behaviors in autism using the Autism Diagnostic Interview-R. *Child Psychiatry and Human Development*, *34*, 3-17.

- DiLavore, P., Lord, C., & Rutter, M. (1995). The Pre-Linguistic Autism Diagnostic Observation Schedule. *Journal of Autism & Developmental Disorders*, *25*, 355-379.
- Elliott, C. D. (1990). *Differential Ability Scales*. San Antonio, TX: Psychological Corporation.
- Gabriels, R. L., Cuccaro, M. L., Hill, D. E., Ivers, B. J., & Goldson, E. (2005). Repetitive behaviors in autism: Relationships with associated clinical features. *Research in Developmental Disabilities*, *26*, 169-181.
- Geschwind, D. H., & Levitt, P. (2007). Autism spectrum disorders: developmental disconnection syndromes. *Current Opinion in Neurobiology*, *17*, 103-111.
- Gotham, K., Risi, S., Pickles, A., & Lord, C. (2007). The autism diagnostic observation schedule: Revised algorithms for improved diagnostic validity. *Journal of Autism and Developmental Disorders*, *37*, 613-627.
- Hollander, E. (2005). Obsessive-compulsive disorder and spectrum across the life span. *International Journal of Psychiatry in Clinical Practice*, *9*, 79-86.
- Horner, R. H., Carr, E. G., Strain, P. S., Todd, A. W., & Reed, H. K. (2002). Problem behavior interventions for young children with autism: A research synthesis. *Journal of Autism and Developmental Disorders*, *32*, 423-446.
- Howlin, P., Goode, S., Hutton, J., & Rutter, M. (2004). Adult outcome for children with autism. *Journal of Child Psychology and Psychiatry*, *45*, 212-229.
- Hus, V., Pickles, A., Cook, E. H., Risi, S., & Lord, C. (2007). Using the autism diagnostic interview-revised to increase phenotypic homogeneity in genetic studies of autism. *Biological Psychiatry*, *61*, 438-448.
- Jones, B. L., Nagin, D. S., & Roeder, K. (2001). A SAS procedure based on mixture models for estimating developmental trajectories. *Sociological Methods & Research*, *29*, 374-393.
- Koegel, L. K., Koegel, R. L., Hurley, C., & Frea, W. D. (1992). Improving social skills and disruptive behavior in children with autism through self-management. *Journal of Applied Behavior Analysis*, *25*, 341-353.
- Lord, C., & Pickles, A. (1996). Language level and nonverbal social-communicative behaviors in autistic and language-delayed children. *Journal of the American Academy of Child & Adolescent Psychiatry*, *35*, 1542-1550.
- Lord, C., Risi, S., DiLavore, P. S., Shulman, C., Thurm, A., & Pickles, A. (2006). Autism from 2 to 9 years of age. *Archives of General Psychiatry*, *63*, 694-701.
- Lord, C., Rutter, M., DiLavore, P. C., & Risi, S. (1999). *Autism Diagnostic Observation Schedule*. Los Angeles: Western Psychological Services.
- Lord, C., Rutter, M., & Le Couteur, A. (1994). Autism Diagnostic Interview--Revised: A revised version of a diagnostic interview for caregivers of individuals with possible pervasive developmental disorders. *Journal of Autism and Developmental Disorders*, *24*, 659-685.
- Lord, C., Schopler, E., & Revicki, D. (1982). Sex differences in autism. *Journal of Autism & Developmental Disorders*, *12*, 317-330.
- Lord, C., Shulman, C., & DiLavore, P. (2004). Regression and word loss in autistic spectrum disorders. *Journal of Child Psychology and Psychiatry*, *45*, 936-955.

- McGovern, C. W., & Sigman, M. (2005). Continuity and change from early childhood to adolescence in autism. *Journal of Child Psychology and Psychiatry*, *46*, 401-408.
- Mooney, E. L., Gray, K. M., & Tonge, B. J. (2006). Early features of autism - Repetitive behaviours in young children. *European Child & Adolescent Psychiatry*, *15*, 12-18.
- Moore, V., & Goodson, S. (2003). How well does early diagnosis of autism stand the test of time? Follow-up study of children assessed for autism at age 2 and development of an early diagnostic service. *Autism*, *7*, 47-63.
- Mullen, E. (1995). *The Mullen Scales of Early Learning*. Circle Pines, MN: American Guidance Service Inc.
- Murphy, G. H., Beadle-Brown, J., Wing, L., Gould, J., Shah, A., & Holmes, N. (2005). Chronicity of challenging behaviours in people with severe intellectual disabilities and/or autism: A total population sample. *Journal of Autism and Developmental Disorders*, *35*, 405-418.
- Richler, J., Bishop, S. L., Kleinke, J., & Lord, C. (2007). Restricted and repetitive behaviors in young children with autism spectrum disorders. *Journal of Autism and Developmental Disorders*, *37*, 73-85.
- Rutter, M., LeCouteur, A., & Lord, C. (2003). *Autism Diagnostic Interview-Revised*. Los Angeles: Western Psychological Services.
- Seltzer, M. M., Shattuck, P., Abbeduto, L., & Greenberg, J. S. (2004). Trajectory of development in adolescents and adults with autism. *Mental Retardation and Developmental Disabilities Research Reviews*, *10*, 234-247.
- South, M., Klin, A., & Ozonoff, S. (1999). The Yale Special Interests Interview [Unpublished measure].
- South, M., Ozonoff, S., & McMahon, W. M. (2005). Repetitive behavior profiles in Asperger's syndrome and high-functioning autism. *Journal of Autism and Developmental Disorders*, *35*, 145-158.
- Sparrow, S. S., Balla, D. A., & Cicchetti, D. V. (1984). *Vineland Adaptive Behavior Scales*. Circle Pines, MN: American Guidance Service Inc.
- Starr, E., Szatmari, P., Bryson, S., & Zwaigenbaum, L. (2003). Stability and Change Among High-Functioning Children with Pervasive Developmental Disorders: A 2-Year Outcome Study. *Journal of Autism and Developmental Disorders*, *33*, 15-22.
- Stutsman, R. (1931). Guide for administering the Merrill-Palmer Scale of Mental Tests. In *Mental Measurement of Preschool Children* (L.M. Terman ed., pp. 139-262). New York, NY: Harcourt, Brace & World.
- Szatmari, P., Georgiades, S., Bryson, S., Zwaigenbaum, L., Roberts, W., Mahoney, W., Goldberg, J., & Tuff, L. (2006). Investigating the structure of the restricted, repetitive behaviours and interests domain of autism. *Journal of Child Psychology and Psychiatry*, *47*, 582-590.
- Turner, M. A. (1997). Towards an executive dysfunction account of repetitive behavior in autism. In J. Russell (Ed.), *Autism as an Executive Disorder*. Oxford: Oxford University Press.

- Turner, M. A. (1999). Generating novel ideas: fluency performance in high-functioning and learning disabled individuals with autism. *Journal of Child Psychology & Psychiatry & Allied Disciplines*, 40, 189-201.
- Venter, A., Lord, C., & Schopler, E. (1992). A follow-up study of high-functioning autistic children. *Journal of Child Psychology and Psychiatry*, 33, 489-507.
- Walker, D. R., Thompson, A., Zwaigenbaum, L., Goldberg, J., Bryson, S. E., Mahoney, W. J., Strawbridge, C. P., & Szatmari, P. (2004). Specifying PDD-NOS: A comparison of PDD-NOS, Asperger's syndrome, and autism. *Journal of the American Academy of Child and Adolescent Psychiatry*, 43, 172-180.
- Wechsler, D. (1991). *Manual for the Wechsler Intelligence Scale for Children (3rd edition)*. San Antonio, TX: Psychological Corporation.

Chapter IV. The Stability of Restricted and Repetitive Behaviors and Interests in Children with Autism Spectrum Disorders

Background and Significance

The development of restricted and repetitive behaviors and interests (RRBs) in children with Autism Spectrum Disorders (ASD) has received far less attention than the development of symptoms in the other core domains, social interaction and communication. However, several recent studies have examined how RRBs evolve over time in children with ASD and have helped us start to compose a picture of the development of this complex group of behaviors.

Much of the literature on RRB development suggests that some behaviors emerge or become more severe between early and later childhood. Many of these studies have used scores on instruments such as the Autism Diagnostic Interview – Revised (ADI-R: Lord, Rutter, & Le Couteur, 1994) as their measure of RRBs (Cox et al., 1999; Moore & Goodson, 2003). A study by Richler, Bishop, Kleinke, & Lord (2007) also used the RRB items on the ADI-R and found that behaviors characterized by ‘insistence on sameness (IS),’ such as adherence to fixed routines, tend to be relatively uncommon in young children with ASD. However, a follow-up study found that as children reach pre-school and school-age, scores on the ADI-R items that assess these behaviors tended to increase, indicating that these behaviors emerged in children who did not have them at a young age, or became more impairing in those who had them early in development (see Chapter III of this volume). Similarly, Moore & Goodson (2003) found that scores on the ADI-R for some RRB items, such as compulsions and rituals, increased between the ages of 2 and 5. In contrast, behaviors involving ‘repetitive sensorimotor (RSM)’ activities, such as motor mannerisms, tend to persist over time, and sometimes even improve. Similarly, the average number of IS items children with ASD exhibited increased over time, but the number of RSM items remained constant or decreased.

These studies have helped us begin to uncover different patterns of change in RRBs in children with ASD, both as a single category and split into subtypes. However, we still know very little about how individual behaviors change across development in children with ASD. It is particularly important to compare prevalence of individual RRBs at different points in development in children with ASD and children with nonspectrum developmental disorders (DD). It is possible that a pattern of increasing prevalence for certain RRBs is specific to ASD, but also possible that these behaviors increase in prevalence in children with DD. Comparing prevalence of RRBs over time in children with ASD and DD can also help us learn which behaviors are most ‘ASD specific.’ Presumably, a behavior is indicative of ASD if it is very common in children with the disorder, and relatively uncommon in children without the disorder. For example, Richler et al. (2007) found that although IS behaviors like compulsions and rituals were relatively uncommon in young children with ASD, they were still more common in this group than in children with DD of approximately the same age. This would suggest that, such behaviors, if present at a young age, are indicative of ASD. However, we do not yet know if this behavior remains ASD specific as children get older. Because prevalence can change over time in children with ASD and/or children with nonspectrum disorders, a behavior that is indicative of ASD at one age may not be at another age.

A related issue is whether different RRBs persist over time, both in terms of their presence and their severity. If a child has a particular behavior as a young child, does that necessarily mean the child will continue to show the behavior when s/he is older? If so, will the behavior improve over time? Conversely, is the absence of a behavior at a given age predictive of its absence later in development? These questions have not been directly addressed. A prospective case series study by Bryson et al. (2007) suggested that some children who do not show unusual behaviors, such as visual fixation on parts of objects, as infants, acquire these behaviors as they get older. However, we do not know how stable different behaviors are from early childhood to school age. The question of how persistent different RRBs are over time has important clinical implications. Previous literature suggests that parents experience a great deal of stress as a result of RRBs (Gabriels, Cuccaro, Hill, Ivers, & Goldson, 2005; Bishop, Richler, Cain, & Lord,

submitted). They might ask clinicians to make predictions about the child's RRB profile, and it is important that the answers are informed by research.

Stability might also depend on child characteristics. The study by Richler et al. (see Chapter III of this volume) suggests that having a lower NVIQ and a diagnosis of autism (as opposed to milder PDD-NOS) is associated with increasing numbers and severity of RSM behaviors over time. This would suggest that RSM behaviors might remain relatively severe for children with autism and/or low NVIQ scores, but perhaps improve or even cease for children with milder ASD and higher NVIQ scores. Similarly, children at lower levels of functioning who do not exhibit RSM behaviors at young ages, might be more likely to acquire them than higher functioning children, and those who have them to a mild degree when they are young might show some worsening in these behaviors. In contrast, higher levels of functioning, particularly in the social domain, were associated with increases in IS behaviors over time in the aforementioned study. Thus, higher functioning children who have IS behaviors at young ages might be more likely to keep them than lower functioning children, and if they do not have these behaviors early in development, they might be more likely to acquire them.

For children who maintain the same behaviors over the course of development, it is important to determine if the degree to which these behaviors are impairing to the child's and family's functioning changes over time. Richler et al. (see Chapter III of this volume) found that some children who had many RSM behaviors over the course of development showed improving scores on RSM items, indicating that although the behaviors persisted, they became less disruptive, according to parent report. Thus, it is important to look at the stability of scores on RRB items, in addition to the stability of the presence or absence of these behaviors.

It is also important to examine whether children acquire behaviors at the same time that they lose others. An early study by Epstein, Taubman, & Lovaas (1985) reported that children who received intensive behavioral intervention evidenced motor mannerisms like rocking and spinning as young children and later demonstrated circumscribed interests. RSM behaviors have been associated with lower levels of development and IS behaviors with higher levels of development (Turner, 1999; Cuccaro et al., 2003). If this is the case, one might expect that IS behaviors would emerge at the

same time that RSM behaviors are lost. In order to test this, one could determine whether children who have several RSM behaviors and then ‘lose’ some of them are more likely to ‘gain’ IS behaviors than children who do not lose any of their RSM behaviors.

Although simultaneous loss and acquisition of RRBs does not necessarily indicate that one behavior is replacing the other, evidence of such a pattern would indicate that this is a potential phenomenon deserving further study.

The present study uses data from a larger longitudinal study of children referred for early diagnosis of ASD and followed up to the age of 9. The focus of this paper is on trajectories of development of individual behaviors, including prevalence, patterns of change in RRB severity, and stability of the presence and absence of behaviors over time. A sample of children with nonspectrum developmental disorders (DD) is included as a control group. The following hypotheses are made:

- (1) Prevalence: For children with ASD, the prevalence of RSM behaviors will remain consistently high or decrease over time, while the prevalence of IS behaviors will increase over time. The prevalence of RRBs in children with DD will remain more consistent over time. RRBs will be less common in children with DD than in children with ASD.
- (2) Changes in severity: RSM behaviors will be consistently severe for children with a high degree of social impairment and/or low IQ scores and will become somewhat less severe over time in children with milder social impairment and/or higher IQ scores. IS behaviors will be relatively mild or absent at young ages in children with ASD and will become more severe over time, but will not reach the same level of severity as RSM behaviors. Children with higher IQ scores and/or milder social impairment will be more likely to have increasingly severe IS behaviors over time. Children with DD will tend to have relatively mild RRBs that remain mild as they get older, but RSM behaviors will be more severe than IS behaviors.
- (3) Stability: Children with ASD who have RSM behaviors at young ages will maintain these behaviors as they get older. Those who do not have IS behaviors at a young age will acquire them. Children with DD will be more likely than

children with ASD to 'lose' RSM behaviors as they get older and less likely to acquire IS behaviors.

Method

Participants

Data for this study were collected between as part of a longitudinal study of toddlers referred for possible autism (see Lord et al., 2006; Anderson et al., in press). In the first wave of data collection, when children were approximately 2 years of age, there were 214 participants (80% male; 67% Caucasian, 31% African American, 2% other; 34% with mothers who had a college or graduate degree). Of these, 192 children were referred because of concerns about ASD. The North Carolina (NC) ASD referral group consisted of 112 consecutive referrals of children younger than 3 years to four TEACCH centers (state-funded clinics providing services for children with autism and related communication disorders in NC). One child's parents withdrew from participation, leaving a total of 111 NC 'ASD referral' children. The Chicago ASD referral group consisted of 81 consecutive referrals of children under age 3 (except for one child who was 37 months at the time of testing) to an autism clinic at a private university hospital. Exclusionary criteria included moderate to severe sensory impairments or cerebral palsy, known genetic abnormalities, and poorly controlled seizures.

The non-spectrum developmental delay referral group consisted of 22 developmentally delayed children recruited between the ages of 13 and 35 months who met the same exclusionary criteria and who had never been referred for or diagnosed with autism. They were recruited from the three largest sources of referral to the NC autism clinics. This group was primarily comprised of children with mental retardation of unknown etiology (33%), language disorder (33%), or a known genetic disorder (29%).

Procedure

Children in the NC ASD referral groups were assessed at four different times, at approximately ages 2, 3, 5, and 9 years. Children in the DD referral group were only assessed at ages 2, 5, and 9 years, and children recruited from Chicago were not seen at

age 5, with a few exceptions. Because not all families participated at every follow-up appointment, sample sizes and other characteristics vary somewhat for each period of data collection. (Please see Chapter III of this volume and Lord et al., 2006 for further detail about the demographic breakdown at each cohort).

Attrition was not related to original best-estimate diagnosis, gender, verbal and non-verbal IQ, adaptive functioning, and level of language (as measured by the Autism Diagnostic Interview-Revised). However, attrition occurred significantly more often in the non-white families, and significantly more often in families where the mother had attained a lower level of education.

At each point in the study, families underwent a two-part standardized assessment that included a parent interview and a child observation. Parents were administered the Autism Diagnostic Interview-Revised (ADI-R: Lord, Rutter, & LeCouteur) and the Vineland Adaptive Behavior Scales (VABS: Sparrow, Balla, & Cicchetti, 1984), and children were administered the Autism Diagnostic Observation Schedule (ADOS: Lord, Rutter, DiLavore, & Risi, 1999), as well as various cognitive and language measures. (Please see Lord et al., 2006 and Chapters II and III of this volume for a complete description of the measures used and the process for making diagnoses.)

At ages 2, 5, and 9, each child was assigned a consensus best estimate clinical diagnosis of autism, Pervasive Developmental Disorder-Not Otherwise Specified (PDD-NOS) or a non-spectrum disorder based on clinical observations, the results of the ADI-R and ADOS, and DSM criteria. All examiners who had seen the child and/or interviewed the caregiver(s) were involved in making the diagnosis. Diagnoses were not given at the age 3 assessment. At age 5, diagnoses were made by examiners blind to the child's history. At age 9, there was always at least one examiner unfamiliar with the child, and about 70% of the time, both examiners were 'blind.' Because children were seen multiple times, diagnoses changed for some children. Even at age 2, the breakdown of ASD vs. non-spectrum diagnoses differed from that of the referral sample of 192 referred for ASD and 22 referred for non-spectrum DD. This is because some of the children referred for ASD were diagnosed as non-spectrum at age 2. In the present study, the child's diagnosis at age 2 is used, unless otherwise specified.

Measures

The Autism Diagnostic Interview – Revised

In the present study, we analyzed all behaviors included in the RRB section of the ADI-R, with the exception of unusual attachments, abnormal/idiosyncratic reactions to sensory stimuli, and circumscribed interests, as these items were not always administered at each cohort. The midline hand movements item was also not examined, as this is a very low-frequency behavior included in the ADI-R to rule out a diagnosis of Rett's Disorder. We added **self-injury**, even though this is not included in the RRB section of the ADI-R, as it is thought to have a repetitive component (see American Psychiatric Association, 1994).

Although this paper focuses on the development of individual RRBs, we also wanted to examine whether RRBs that tend to cluster together in factor analyses also showed similar developmental patterns. Previous studies have consistently found that certain RRBs consistently load on one of two factors: a 'repetitive sensorimotor' (RSM) factor, comprised of **unusual sensory interests, repetitive use of objects, hand and finger mannerisms, and complex mannerisms**; and an 'insistence on sameness' (IS) factor, comprised of **difficulties with changes in routine, resistance to trivial changes in environment and compulsions and rituals**. In a previous study, we confirmed that these behaviors load on two different factors across time, using the same data as in the present study (see Chapter III of this volume for factor loadings at each wave of data collection). In this paper, we consider these behaviors individually and as members of RRB 'subtypes.' The remaining behaviors, **unusual preoccupations, sensitivity to noise, and self-injury**, have not consistently loaded on either factor. We will look at the developmental trends for these behaviors to see if they resemble those of either RRB 'subtype.' Results are organized according to behavior subtype (i.e. RSM, IS, and other) for ease of comprehension.

Analyses

For analyses of prevalence and stability of RRBs across cohorts, we used SAS Proc Genmod (SAS for Windows release 9.1.3). This procedure allows for the inclusion

of a ‘repeated’ statement, which controls for higher correlations among observations from a single subject and is suitable for unbalanced panel data (i.e., where data for some participants is missing at certain time points).

In order to examine different patterns of change in RRB severity over time, we used Proc Traj (Jones, Nagin, & Roeder, 2001), an exploratory procedure written for use in SAS that identifies linear and nonlinear patterns in longitudinal data and classifies the sample into groups based on each individual’s trajectory. We ran a series of models using the censored, normal distribution, to see if distinct groups would emerge. The Bayesian Information Criterion (BIC) was used to determine the optimal number of trajectory groups for each behavior, with a smaller absolute value for the BIC indicating a better fit (see Jones et al., 2001, for the use of the BIC for model selection).

Results

RSM Behaviors

Changes in prevalence over time in children with ASD and DD

As a first step, we calculated the prevalence of each RRB on the ADI-R at cohorts 2, 5, and 9, according to the child’s diagnosis (ASD vs. DD) at age 2 (see Table 4.1). Cohort 3 was omitted, because the DD referral participants were not seen at this age and therefore this was not a representative sample of children with DD. We also omitted cohort 3 from the ASD analyses, to allow for easier comparison with the DD results. Prevalence was calculated separately using only children who were seen at all three cohorts, and results were similar; therefore, only results for the whole sample are reported here.

In order to determine if prevalence significantly increased over time, we ran logistic regressions in which the outcome was whether or not the behavior was present, and cohort was the predictor. These analyses were run separately for children with ASD and DD. Among children with ASD, several patterns were observed. **Unusual sensory interests** decreased in prevalence over time, so that at age 9, the behavior was significantly less common than it had been at age 5. **Repetitive use of objects** was significantly less common at age 9 than it had been at age 2. The two types of motor

mannerisms, **hand and finger mannerisms** and **complex mannerisms**, did not change significantly in prevalence over time.

In the DD group, none of the RSM behaviors changed in frequency significantly. This was partly due to the smaller sample sizes for the DD group. However, as the percentages indicate, patterns of increasing or decreasing prevalence were not as clear as in the ASD subgroup.

In order to compare prevalence of each behavior for children with ASD and DD at each cohort, we also ran logistic regressions with the same outcome, but with diagnosis (i.e. ASD versus DD) as the predictor. NVIQ at age 2 was included as a covariate, since it differed significantly between children with ASD and those with DD. The RSM behaviors were relatively common in the DD group, present in anywhere from one-quarter to one-half of the children. However, all the RSM behaviors were still significantly more common in the ASD sample at ages 2 and 5. For **unusual sensory interests** and **repetitive use of objects**, differences by diagnostic group were not significant at age 9, due to the increase in prevalence between ages 5 and 9 in the DD group.

Changes in severity of behaviors over time

The analyses of prevalence tell us whether behaviors became more or less common over time. However, we do not know from these analyses what kinds of patterns of change in severity children tended to follow. For example, even if overall prevalence did not change over time for a given behavior, there could have been a subgroup of children who showed improvement, as indicated by decreasing scores over time, a subgroup of children who showed worsening, as indicated by increasing scores, and a subgroup of children whose behaviors tended to stay relatively constant in terms of severity, with little change in scores over time.

In order to understand more about patterns of change seen for individual behaviors, we conducted Proc Traj analyses on each behavior, where the child's score on that behavior was the outcome. Analyses were first conducted on the whole sample (i.e. ASD and DD) and then run on the ASD sample only.

(i) Trajectories in the ASD and DD combined sample

Table 4.2 presents the statistical findings for the analyses of the whole sample, and Figure 4.1 provides a visual depiction of the trajectories observed for each behavior. For **repetitive use of objects** (Figure 4.1.1), **unusual sensory interests** (Figure 4.1.2), and **hand and finger mannerisms** (Figure 4.1.3), a three-group solution provided the best fit, based on the BIC. For all three behaviors, there was one group whose scores, on average, consistently remained between 1 and 2, indicating persistent severity. This was the largest trajectory group for **repetitive use of objects** and **hand and finger mannerisms**; for both behaviors, nearly half of the sample fell into this group, compared to just over one-quarter for **unusual sensory interests**. Each behavior also had one group of children who significantly improved over time. This was the largest trajectory group for **unusual sensory interests**; over half of the children were classified in this group, compared to 31% and 19% for **repetitive use of objects** and **hand and finger mannerisms**, respectively. The final group for all three behaviors was comprised of children whose degree of impairment was relatively mild when they were young. For **hand and finger mannerisms**, children in this group showed some worsening over time, but for **unusual sensory interests** and **repetitive use of objects**, scores remained low, indicating consistently mild behaviors.

For **complex mannerisms** (Figure 4.1.4), a two-group solution provided the best fit. Neither group showed significant change over time; there was a group of children with consistently low scores and a group of children with consistently moderate scores, each of which had similar numbers of children.

(ii) Trajectories in the ASD subgroup only

The Proc Traj analyses were re-run on the ASD subgroup, in order to determine if similar trajectory groups would arise. Trajectory groups were similar to those described above. When the analyses were restricted to the ASD sample, a substantial proportion of children fell into the most severe group for **complex mannerisms** (78.9%), **hand and finger mannerisms** (59.0%), and **repetitive use of objects** (57.1%) For **unusual sensory interests**, only 30.4% of the ASD group fell into the consistently severe group.

Child characteristics associated with patterns of change

In order to better understand how variables like diagnosis affected the relative likelihood of group assignment for the RSM behaviors, we then introduced covariates into the Proc Traj analyses (see Table 4.3). Each model was re-run with child gender and race, maternal education, site at which the child was recruited, NVIQ at 2, and diagnosis at 2. Because previous analyses with these data indicated that diagnosis was highly associated with ADOS Social Affect (SA) scores (see Chapter III of this volume) we ran analyses separately replacing diagnosis with SA score, in order to avoid multicollinearity.

(i) Covariates associated with trajectories in the ASD and DD combined sample

For all of the RSM behaviors, having a diagnosis of DD significantly reduced the likelihood of being in the most severe group. For example, based on the estimates reported in the table, the odds of being assigned to the highest-scoring group versus the lowest-scoring group for **repetitive use of objects** were over 15 times greater for children with autism compared to children with DD. In the lowest scoring group, over two-thirds of the children had a diagnosis of DD, and only approximately 10% had autism, with the remaining 20% having PDD-NOS. In contrast, in the highest scoring group, approximately 75% of the children had autism and less than 5% had DD, again with approximately 20% of children having PDD-NOS.

Having a DD diagnosis also reduced the likelihood of being assigned to the highest scoring group relative to the improving group for **repetitive use of objects**. For **unusual sensory interests** and **repetitive use of objects**, DD children were most likely to be assigned to the mildest group, in which scores remained consistently low, rather than the improving group, in which scores started high and decreased over time.

For **hand and finger mannerisms**, as described above, there was a consistently severe group and an improving group, as for the other behaviors. However, the third group was comprised of children whose scores started low but increased, rather than being comprised of children with consistently low scores, as for the other RSM behaviors. Diagnosis did not affect the relative likelihood of assignment to the worsening group relative to the improving group.

When diagnosis was substituted with ADOS SA scores, in order to look at the less confounded relationship between RSM behaviors and social functioning, results were similar, for the most part. Consistent with the findings for diagnosis, children with higher SA scores were significantly more likely to be assigned to more severe groups. For example, for **hand and finger mannerisms**, each 1-point increase in SA score resulted in a 19% increase in the likelihood of being in the highest-scoring group relative to the lowest-scoring group. Results were similar when comparing the highest-scoring group to the improving group. As with diagnosis, SA scores did not affect the likelihood of being assigned to the improving group versus the consistently mild group. The average SA score was 9.7 for the improving group, 10.1 for the worsening group, and 15.5 for the consistently severe group. For **unusual sensory interests** and **repetitive use of objects**, SA scores did not affect the likelihood of assignment to the improving group relative to the consistently mild group, whereas diagnosis had.

NVIQ score at age 2 was also a significant risk factor for all of the RSM behaviors. Having a lower NVIQ score significantly increased the chances of assignment to a higher-scoring group. For example, for **unusual sensory interests**, each 1-point decrease in NVIQ score resulted in a 12% increase in the chances of being assigned to the consistently severe group compared to the consistently mild group and a 7% increase in the chances of being assigned to this group relative to the improving group. Children in the consistently severe group had an average NVIQ score of 54, compared to 73 for the improving group and 84 for the consistently mild group. However, it was only for **unusual sensory interests** that NVIQ affected the likelihood of being assigned to one of the two lower-scoring groups over the highest one.

Of the remaining covariates, gender and site were significant risk factors for **repetitive use of objects**. Males were 31 times more likely to be assigned to the highest-scoring group and 17 times more likely to be assigned to the improving group than the consistently mild group. This was the case even controlling for the fact that a greater proportion of children with ASD were male. Children who were recruited from Chicago were over 9 times more likely to be assigned to the ‘decreasing group’ than the ‘consistently low’ group. Maternal level of education and race were not significant risk factors for any of the behaviors.

(ii) Covariates associated with trajectories in the ASD subgroup only

In order to learn more about which variables were associated with different score trajectories among children with ASD, we re-ran the above analyses on children with an ASD diagnosis (i.e. autism or PDD-NOS) only. In particular, we were interested in whether the child's diagnosis within the spectrum and NVIQ at age 2 would be associated with following a particular trajectory, or whether these effects had been driven by the DD subgroup. In this set of analyses, diagnosis was a significant risk factor for **repetitive use of objects** ($\beta = -2.04, se = .94, p < .05$) Children with autism were nearly 8 times more likely than children with PDD-NOS to be assigned to the 'consistently high' group rather than the 'consistently low' group. In the lowest-scoring group, only approximately one-quarter of the children had autism at age 2, whereas in the highest-scoring group, approximately 80% of the children had this diagnosis.

Diagnosis was not a significant risk factor for **unusual sensory interests**. Upon closer examination, it was clear that this result was due to the fact that the lowest scoring group was comprised exclusively of children with PDD-NOS. Thus, relative likelihoods based on diagnosis could not be accurately estimated. However, it appears that having autism was associated with being in the consistently severe group.

Unusual sensory interests was the only behavior whose score trajectories were associated with SA score, when analyses were run with this variable instead of spectrum diagnosis. The effect of SA was modest: having a higher SA score, indicating greater social impairment, increased the likelihood of being in the consistently severe group relative to the consistently mild group by 28%. The mean SA score was 14.48 in the highest-scoring group and 13.30 in the lowest-scoring group.

NVIQ score at age 2 was also associated with patterns of change among children with autism for **unusual sensory interests**, $\beta = -.09, se = -.04, p < .05$; **hand and finger mannerisms**, $\beta = -.05, se = -.02, p < .01$; and **complex mannerisms**, $\beta = -.04, se = -.02, p < .05$). In all cases, having a lower NVIQ was associated with being in a higher scoring group. For example, the average NVIQ score for children in the consistently moderate group for **complex mannerisms** was 60.9, whereas in the consistently mild group, it was 75.3.

Stability of behaviors over time

The results just presented provide a sense of the patterns of change in severity that emerged for different behaviors and the child characteristics that were associated with these patterns. Another objective of this study was to learn about the stability of different RRBs. If a child starts exhibiting an unusual behavior at a young age, how likely is s/he to continue showing the behavior as s/he gets older? Conversely, is the absence of a particular behavior at a young age predictive of its absence later in development, or are some behaviors commonly acquired when children get older?

In order to address these questions, we ran logistic regressions for each behavior, where the outcome was the probability of having the behavior at Time 2, and the predictor was whether or not the child had the behavior at Time 1. This was done for cohorts 2, 5, and 9, for both ASD and DD. For example, we calculated the probability of having the behavior at age 9 given that it was present at age 5, as well as the probability of having the behavior at age 9 given that it was absent at age 5. If a behavior was relatively stable, then the probability of having it at age 9 given that it was present at age 5 should be greater than the probability of having it at age 9 given that it was absent at age 5. Analyses are not reported for the age 3 cohort for reasons explained above. Regressions were run separately for children with ASD and children with DD.

(i) Stability in the ASD subgroup

When examining stability in children with ASD only, we included NVIQ and diagnosis at age 2 as covariates. We also included the interaction between spectrum diagnosis/NVIQ and the predictor of interest (i.e. whether or not the child had the behavior at the previous cohort), in order to see if stability differed for children with autism and children with PDD-NOS, as well as for children with different levels of cognitive ability.

Results are presented in Table 4.4. In general, children with ASD were very likely to maintain RRBs from one time point to the next. For example, of 48 children who had **hand and finger mannerisms** at age 2, 38 (79.2%) continued to have this behavior at age 5. However, of the 42 children who did not have the behavior at age 5, 15 (35.7%)

had acquired it by age 9. Thus, although the stability for the presence of this behavior was high, the stability for its absence was relatively low. As can be seen in Table 4.4, this pattern also emerged for **unusual sensory interests** and **repetitive use of objects**.

Hand and finger mannerisms was the only RSM behavior for which stability varied according to whether the child had a diagnosis of autism or PDD-NOS at age 2. Stability for the presence of **hand and finger mannerisms** was significantly greater for children with autism than children with PDD-NOS between ages 2 and 5, $\beta = 2.11$, $se = 1.08$, $p < .05$. Of 34 children with autism who had **hand and finger mannerisms** at age 2, 30 (88.2%) continued to have the behavior at age 5. In contrast, of 14 children with PDD-NOS who had the behavior at age 2, only 8 (57.1%) continued to have it at age 5.

(ii) Stability in the DD group

The presence of RSM behaviors tended to be less stable in the DD subgroup than in the ASD subgroup (see Table 4.4). For example, only half of the children who had **hand and finger mannerisms** at age 2 continued to do so at age 5, and the same was true between ages 5 and 9. In contrast, children with DD who did not have RSM behaviors at one time point were less likely to acquire them than children with ASD.

Summary of Developmental Trends

In sum, children with ASD tended to have RSM behaviors that persisted over time. Once a child had a behavior in this category, s/he was likely to continue having it in later years. However, there was evidence of improvement in these behaviors, particularly for children with higher NVIQs and/or milder ASD. Although these behaviors were relatively common in the DD sample, they were significantly more common in the ASD sample in most cases. RSM behaviors were also less stable in the DD sample; children who had them when they were young did not necessarily continue to have them when they were older.

IS Behaviors

Changes in prevalence over time in children with ASD and DD

In contrast to the RSM behaviors, all of the IS behaviors increased in prevalence over time (see Table 4.1). The biggest increase in prevalence for these behaviors occurred between ages 2 and 5. **Difficulties with changes in routine** were present in approximately one-third of children at age 2 and over half of the children by age 5. Similarly, **compulsions and rituals** doubled in prevalence between ages 2 and 5. **Resistance to changes in environment** was relatively uncommon at age 2, present in only 10% of children with ASD. Thus, although it doubled in prevalence by age 5, a significant increase, it was still a relatively uncommon behavior.

In the DD group, **compulsions and rituals** and **difficulties with changes in routine** showed a significant increase in prevalence over time, such that they were more common at age 9 than at age 2. As seen in Table 4.1, these behaviors were present in approximately one-third of the DD sample at age 9. Compared to the RSM behaviors, the IS behaviors tended to be less specific to ASD at age 2, as they were relatively uncommon in both groups. However, at age 5, **compulsions and rituals** and **difficulties with changes in routine** were significantly less common in the DD sample than in the ASD sample.

Changes in Severity over Time

(i) Trajectories in the ASD and DD combined sample

For the IS behaviors, a two-group solution consistently provided the best fit (see Table 4.5). For **difficulties with changes in routine** (Figure 4.2.1), nearly 70% of children maintained consistently high scores over time, while the remainder started with mild symptoms and showed worsening over time. In contrast, for **resistance to changes in environment** (Figure 4.2.2), the largest proportion of children was classified in a group that showed consistently mild symptoms over time. Average scores for children in this group were close to 0 at all ages. Thus, many children in this group never had the behavior at all. The other group of children showed some worsening over time, although behaviors were relatively mild even at their worst. For **compulsions and rituals** (Figure

4.2.3), the larger group was comprised of children whose scores remained consistently very low until approximately 4 years of age, at which point they began to increase. This change in slope explains why there was a significant quadratic effect for this group. Increases were significant, but relatively modest; scores were still substantially lower than 1, even at older ages. The other group was comprised of children whose scores started somewhat higher, increased more sharply until approximately the age of 5, and then reached a plateau.

(ii) Trajectories in the ASD subgroup only

Like the RSM behaviors, trajectories for IS behaviors were similar when we restricted our analyses to the ASD sample. For **difficulties with changes in routine** and **compulsions and rituals**, a greater proportion of children now fell into the more severe group (40% and 41%, respectively). However, even among children with ASD, the worsening trajectory was relatively uncommon for **resistance to changes in environment**; only 22% of children fell into this group.

Child characteristics associated with patterns of change

(i) ASD and DD combined sample

Diagnosis was a significant risk factor for **resistance to changes in environment** (see Table 4.6). Children with DD were approximately half as likely as children with autism to be in the higher scoring group. Children in the group who consistently had scores of 0 for this behavior were relatively evenly distributed across all three diagnostic groups, whereas in the worsening group, over two-thirds of the children had autism and only 7% of children with DD. Diagnosis did not affect the likelihood of group assignment for **difficulties with changes in routine** and **compulsions and rituals**. When SA score was substituted for diagnosis, it was not a significant risk factor. Although having a diagnosis of autism increased the likelihood of being assigned to the higher scoring group for this behavior, having a higher SA score did not.

Of the remaining variables, gender was a significant risk factor for **difficulties with changes in routine**; males were over 3 times more likely to be in the consistently

severe group. Maternal level of education was a significant risk factor for **compulsions and rituals**. Having a mother with a lower level of education significantly decreased the likelihood of assignment to the group whose scores increased between ages 2 and 5. Race, recruitment site, and NVIQ, were not significant risk factors for any of the IS behaviors.

(ii) ASD subgroup only

Diagnosis within the spectrum was not associated with the likelihood of group assignment. There were no significant effects for SA or NVIQ scores at 2 when analyses were restricted to the ASD subgroup.

Stability of Behaviors over Time

(i) Children with ASD

Difficulties with changes in routine was the only IS behavior that had clearly stable presence over time (see Table 4.7). The other IS behaviors were less stable for their presence. For example, of the 15 children with ASD who had **resistance to changes in environment** at age 5, only 6 (40.0%) continued to have the behavior at 9. In contrast, this behavior clearly had stable absence. Of the 51 children who did not have the behavior at age 5, 42 (82.3%) continued not to have it. Stability for the absence of this behavior remained high between ages 5 and 9.

Among children with ASD, stability for the absence of **difficulties with changes in routine** was greater for children with PDD-NOS than for children with autism between the ages of 5 and 9, $\beta = -8.02$, $se = 2.25$, $p < .001$. In the PDD-NOS subgroup, of 12 children who did not exhibit the behavior at age 5, 11 (91.6%) continued not to exhibit the behavior at age 9. In contrast, 11 of 19 (57.9%) children with autism who did not have this behavior at age 5 acquired it by age 9. For **compulsions and rituals**, stability was higher in the PDD-NOS group, both in terms of the presence and the absence of the behavior, $\beta = -2.60$, $se = 1.19$, $p < .05$. Of the 20 children with autism who had this behavior at age 5, only 8 (40.0%) maintained it until age 9. In contrast, 7 of 9 children with PDD-NOS maintained this behavior during the same period. Children

with autism were also more likely than not to acquire this behavior by age 9, if they had not had it at age 5. Of the 20 children with autism who did not have the behavior at age 5, 13 (65.0%) acquired it. Children with PDD-NOS were not as likely to acquire this behavior; of the 19 children who did not have it at age 5, 7 (36.8%) acquired it.

(ii) Children with DD

Resistance to changes in environment appeared to be highly unstable for children with DD (see Table 4.7). However, so few children had this behavior at any time that stability is difficult to establish. **Compulsions and rituals** had a level of stability comparable to the ASD sample, while the presence of **difficulties with changes in routine** was less stable than in the ASD sample, particularly between ages 2 and 5.

Summary of Developmental Trends

Overall, IS behaviors were relatively uncommon in young children with ASD and became more common with time. This was true both for children with ASD and DD. However, increases in prevalence were more dramatic for children with ASD. Despite this general trend for increased prevalence, there was still a substantial minority of children with ASD who had consistently mild IS behaviors. **Resistance to trivial changes in environment** was consistently absent over time, even among many children with ASD. In contrast to the RSM behaviors, trajectories of IS behaviors were not closely related to child characteristics like diagnosis and NVIQ.

‘Other’ Behaviors

We wanted to look at the same patterns in **unusual preoccupations**, **self-injury** and **sensitivity to noise**, which do not consistently load on the RSM or IS factor, in order to see if they shared developmental trends with either factor.

Prevalence of behaviors over time

Unusual preoccupations and **self-injury** were both consistently present in a substantial minority of children with ASD. Rates of **self-injury** in the DD sample were similar to those in the ASD sample, whereas **unusual preoccupations** was significantly

more common in ASD at ages 2 and 5. In contrast, **sensitivity to noise** clearly increased in prevalence between ages 2 and 5 in children with ASD and was not specific to ASD, as it was consistently present in many children with DD (see Table 4.1).

Changes in severity over time

The trajectory analyses indicated that children tended not to show significant changes in severity for **unusual preoccupations** and **self-injury** (see Table 4.8). For both behaviors, there was a group of children with consistently low scores and a group of children with consistently higher scores. This was somewhat similar to the trajectories for **complex mannerisms**, in the RSM category. For **unusual preoccupations** (Figure 4.3.1), over half of the children were in the higher scoring group, whereas for **self-injury** (Figure 4.3.2) the majority of children were in the consistently mild group. For **sensitivity to noise** (Figure 4.3.3), there was a group of children whose behavior substantially worsened only between ages 2 and 5, and a group with more constant, but milder worsening, over time, similar to **compulsions and rituals** in the IS category. Children were evenly distributed between the two groups

Child characteristics associated with patterns of change

Diagnosis was a significant risk factor for **unusual preoccupations** and **sensitivity to noise**, but not **self-injury**. Of the children in the consistently mild group for **unusual preoccupations**, 17.3% had autism, while in the ‘consistently moderate group, 66.9% did. When SA score replaced diagnosis, it was not a significant risk factor for any behaviors. NVIQ was a significant risk factor for **unusual preoccupations** and **self-injury**. In both cases, lower NVIQ scores increased the likelihood of assignment to the higher-scoring group. For example, for **self-injury**, the average NVIQ score was 72.1 in the consistently absent group and 60.7 in the worsening group. Gender was also significant for **unusual preoccupations**, with boys about 5 times more likely to be assigned to the moderate-scoring group rather than the low-scoring group. Of the children in the ‘consistently low’ group, 60% were male, whereas in the consistently moderate group, over 90% were male. For **sensitivity to noise**, lower levels of maternal education

were associated with a greater likelihood of assignment to the higher-scoring group. (See Table 4.9).

When the trajectory analyses were restricted to the ASD sample, **unusual preoccupations** was the only behavior whose score trajectories were affected by diagnosis. Children with autism were almost 7 times more likely to be in the higher-scoring group than in the mild group.

Stability of behaviors over time

Finally, the presence of **unusual preoccupations** and **self-injury** was relatively unstable for both children with ASD and those with DD. The stability for the absence of these behaviors was higher; children who did not have these behaviors at one time point tended to continue not to have them. The presence of **sensitivity to noise** has highly stable between 2 and 5 in children with ASD, but less stable between 5 and 9. In the DD sample, the presence of this behavior was never very stable. (See Table 4.10).

Summary of Developmental Trends

The behaviors that fell into the ‘other’ category did not consistently share features with either RSM or IS behaviors, in terms of developmental patterns. **Unusual preoccupations** and **self-injury** shared some similarities with RSM behaviors, in that they had relatively stable prevalence, and scores, on average, tended to remain stable over time. Additionally, trajectories were associated with diagnosis and NVIQ. However, the stability analyses indicated that on an individual level, **unusual preoccupations** was not very stable; children who had the behavior at one time point often lost it as they got older. This is in contrast to most of the RSM behaviors, which children tended to maintain over time. Also, although **unusual preoccupations** tended to be more common in ASD than in DD, like the RSM behaviors, **self-injury** was equally prevalent in both groups. **Sensitivity to noise** shared some features with IS behaviors, in that prevalence tended to increase over time, and some children showed worsening in this behavior, particularly between the ages of 2 and 5. However, this behavior was relatively common in children with DD, in contrast to the IS behaviors.

Concurrent Changes in RSM and IS Behaviors

Another objective of this study was to examine whether losses of some behaviors were contemporaneous with the acquisition of others. Based on the notion that IS behaviors are associated with higher levels of development while RSM behaviors are associated with lower levels of development, we might expect that, as children get older, they acquire IS behaviors at approximately the same time that they lose RSM behaviors. In order to examine this hypothesis, we categorized children based on whether their total scores on the RSM and IS items increased, decreased, or remained the same between cohorts. We then used cross-tabulations to determine how likely children were to have an increase in IS score between two time points, given that they had a decrease in RSM score in the same period. In particular, we were interested in whether children were more likely to show worsening IS behaviors if they had had improving RSM behaviors during the same period than if they did not. The same procedure was followed with total number of RSM and IS items (i.e., to determine if ‘gaining’ IS behaviors was associated with ‘losing’ IS behaviors). Similar results were obtained; therefore, only results for RSM and IS scores are reported here.

Overall, children whose RSM scores decreased between two cohorts were not more likely to have an increase in IS score. In fact, children whose RSM score *increased* between two cohorts were more likely to have an increase in IS score during the same time period than children whose RSM score decreased. For example, of 58 children whose RSM score decreased between ages 2 and 9, 36 (62.1%) had IS scores that increased during the same period. In contrast, of 48 children whose RSM score increased between ages 2 and 9, 41 (85.4%) had increases in IS scores. A chi-square test found this difference to be significant, $\chi^2 = 7.2$, $p < .01$. When we ran the cross-tabulations on the ASD sample only, results were similar. Again, children whose RSM scores increased were generally more likely to have an increase in IS scores than children whose RSM scores decreased. However, because some cell sizes were relatively small when we ran analyses on the ASD subsample, chi-square tests could not be run.

Just as increases in RSM scores were generally accompanied by increases in IS scores, so were decreases in RSM scores associated with decreases in IS scores. For example, of 24 children whose RSM scores decreased between ages 5 and 9, 15 (62.5%)

had decreases in IS scores, while of 26 children whose RSM scores increased during this time, only 7 (26.9%) had contemporaneous decreases in IS scores.

These results suggested that RSM and IS scores from the same age were positively correlated. We ran Pearson correlations on RSM and IS scores for each age (e.g. RSM score at age 2 and IS score at age 2). As expected, RSM and IS scores were positively and significantly correlated for most pairs ($r = .32, p < .001$, for scores at age 2; $r = .29, p < .001$, for scores at age 3; $r = .36, p < .001$, for scores at age 9.) At age 5, the correlation between RSM and IS scores did not reach significance ($r = .16, p = .07$). Correlation coefficients were similar when run on children with ASD only. At age 5, the correlation was even lower, ($r = .07, ns$).

In order to determine why the correlation at age 5 was lower than at others, we then ran correlations separately for children with autism, children with PDD-NOS, and children with DD. Correlation coefficients were similar for the three groups at all ages, except at age 5, where there was a negative, but nonsignificant, correlation between RSM and IS scores for children with autism ($r = -.15, ns$), a positive correlation between scores for children with PDD-NOS, which approached but did not reach significance due to small sample size ($r = .30, p = .08$), and a negligible correlation between scores for children with DD ($r = -.02, ns$). Thus, the low correlation between scores at age 5 for the whole sample resulted from combining the three diagnostic groups, each of which showed a different relationship between RSM and IS scores at this age.

The most obvious difference between cohort 5 and the other cohorts is that it was almost exclusively comprised of children from North Carolina. This means that the ‘DD referral’ children (i.e., those for whom there had never been a concern about ASD), who were recruited from NC, made up the majority of the DD group at age 5. In contrast, in other cohorts, the DD sample was comprised both of children from the ‘DD referral’ group as well as children from the ‘ASD referral’ group (i.e., children for whom there had been concerns about ASD, but who were diagnosed with DD at 2). The DD children in the age 5 cohort might have been relatively less impaired, overall, than the DD children at other cohorts, given that these were children for whom ASD had never been a concern. When we compared children in the DD subgroups at each cohort, children with DD had a mean SA score of 3.7 at age 5, compared to 6.5, 5.7, and 6.6 for the age 2, 3,

and 9 cohorts, respectively, $F(3,155) = 2.86, p < .05$, supporting the hypothesis that the age 5 DD children were less impaired.

Given the positive correlation between RSM and IS scores at most cohorts, we wanted to see what proportion of children with ASD had high scores on both factors. We calculated the median RSM and IS score at each cohort, and considered a child to be ‘high’ on both if the child exceeded the median for both scores. For cohorts 2, 3, and 9, we found that approximately 20% of children with ASD had high scores on both the RSM and IS factors.

In sum, children who lost or improved in RSM behaviors were not more likely to gain or worsen in IS behaviors than children who did not. Rather, gaining or worsening in behaviors in one group was associated with similar trends in the other group. Among children with ASD, there was a subgroup that had several and/or severe behaviors in both the RSM and IS subdomains.

Discussion

Heterogeneity in the Development of Individual RRBs

Most studies of RRBs in children with ASD have focused on the category of RRBs as a whole. Some recent studies have further subdivided the category and have considered behaviors as part of RRB subtypes. Here, we explored RRBs and their development on an even finer-grained level, that of individual behaviors. Our findings indicate that, within this group of behaviors that is commonly thought of as a category, there is a good deal of variability in development, including how common these behaviors are at different points in time, how their severity changes as children get older, and how commonly behaviors are lost or acquired. The variables that affect patterns of development are also different depending on the behavior.

This degree of heterogeneity highlights the importance of looking at RRBs ‘under the microscope.’ Parents might want a prognosis of their child’s RRB profile as they get older, and the findings here suggest that prognoses might differ substantially depending on the behavior. A young child who has a severe unusual sensory interest, such as visually examining objects for long periods of time, might show some improvement in

this behavior with age. This is particularly true if the child has milder autism, specifically milder social impairments. However, a child who has severe difficulties with changes in routine is likely to continue to have similarly severe difficulties at older ages, even if that child is relatively high functioning. In some cases, not having a behavior at a young age is a good prognostic indicator. For example, a child who is relatively flexible regarding small changes in his or her environment is likely to continue tolerating such changes as s/he gets older. However, the absence of some behaviors early in development, such as hand and finger mannerisms, is not predictive of their absence several years later, and parents of children who display this behavior to a mild degree when they are young might find that this behavior becomes more disruptive as their child gets older, particularly if the child has a clear diagnosis of autism, as opposed to PDD-NOS. Thus, when parents ask clinicians how likely it is that their child will continue to display RRBs and whether these behaviors are likely to improve, the answer will depend on which behavior the parent is asking about and the characteristics of that particular child.

Evidence for RRB Subtypes

Although the findings from this study indicate considerable variability in the development of individual RRBs, they nevertheless provide further evidence for the idea that there are RRB ‘subdomains,’ one characterized by behaviors that have repetitive sensory and/or motor components, and the other by behaviors that involve adherence to fixed sequences and/or routines. As predicted, behaviors in the former category tended to remain very common in children with ASD over time. The results from the trajectory analyses indicated that, for many children with ASD, RSM behaviors remain present and severely impairing over time. Having a low NVIQ and a clear diagnosis of autism, even at a young age, increased the likelihood that these behaviors would be persistent and severe across development, also consistent with our predictions. In contrast, as hypothesized, behaviors in the IS category were often absent or very mild in young children with ASD, but tended to emerge and/or increase in severity as children get older. This was true regardless of the child’s level of cognitive and social ability.

Despite the many differences in the ways that RSM and IS behaviors changed over time in children with ASD, there was no evidence that IS behaviors “replaced” RSM

behaviors, contrary to our prediction. In fact, many children with ASD in our sample had both kinds of behaviors, particularly in later childhood. Thus, there appears to be some relationship between the two kinds of behaviors. Perhaps some underlying mechanism, such as early disruption to brain development, gives rise to further difficulties, such as impaired executive function and lack of social interest, which each, in turn lead to different kinds of RRB. It is also important to consider whether behaviors within a factor, or even behaviors that are scored on the same ADI-R item, ‘replace’ each other over time. For example, a child might stop lining objects up at the same time that s/he starts spinning objects. Both of these are considered example of repetitive use of objects, but they take on different forms. Prospective longitudinal studies would be most appropriate for addressing the question of whether behaviors replace each other, as it would be possible to determine whether the acquisition or worsening of one behavior closely followed the loss or improvement of another.

The behaviors that were not part of either the RSM or IS group of behaviors, namely unusual preoccupations, sensitivity to noise, and self-injury, did not consistently follow the trends of either group. Given that these behaviors have not consistently loaded on a particular factor in other studies (Cuccaro et al., 2003; Szatmari et al., 2006), it is not surprising that they are difficult to categorize based on developmental patterns. It is important to consider the implications of this finding. It could be that some of these behaviors have both repetitive sensorimotor and inflexible aspects to them, and therefore share characteristics with both RSM and IS behaviors. For example, a child who has a preoccupation with fans might enjoy watching fans spin around, which is a sensory interest. At the same time, the child might insist on looking at the fan as the first thing they do every time they visit their grandmother’s house, which is a compulsive behavior. It is also possible that these behaviors fall into other RRB ‘subtypes’ that have not been identified yet. Finally, sensitivity to noise and self-injury have been found to be relatively common in children with nonspectrum disorders, both in the present study as well as in previous work. Such behaviors might be related to disruptions in brain development that are associated with general cognitive impairment, rather than with ASD specifically.

The Role of Child Characteristics in ASD

Among children with ASD, children who had lower NVIQ scores at age 2 were more likely than children with higher NVIQs to have RSM behaviors that remained consistently severe over time. Having autism (versus PDD-NOS) was also a risk factor for persistently severe RSM behaviors, as well as unusual preoccupations. This was partly driven by the fact that children with autism had a greater degree of social impairment than children with PDD-NOS. However, the association between having an early diagnosis of autism and having consistently severe RRBs was likely due to factors beyond degree of social impairment, as suggested by the finding that, for some behaviors, having autism increased the likelihood of having a consistently severe trajectory, but having a higher social affect score on the ADOS did not. It is important, then, to consider what it is about having an early diagnosis of autism, other than being socially impaired, that is related to having severe RRBs that persist over time. A child who has many and/or severe RRBs at a young age is likely to be diagnosed with autism rather than PDD-NOS, and, as the present study has shown, RRBs that are severe in early childhood often remain severe as children get older. Thus, if early diagnosis is associated with having consistently severe RRBs, this relationship could be driven by the overall stability of severe RRBs in autism. The predictive value of early diagnosis can be assessed using a technique such as growth curve analysis. It makes sense for early RRBs to at least partly predict trajectories of RRB development. However, there may be other symptoms associated with an early diagnosis of autism that have predictive value that goes beyond what is accounted for by early RRBs, such as stereotyped or idiosyncratic language

Previous studies have suggested that behaviors characterized by insistence on sameness should be thought of as ‘higher order’ behaviors (Turner, 1999). In a recent study (see Chapter III of this volume), we found some evidence for this claim, as children with milder social impairments were more likely to exhibit IS behaviors (see Chapter III of this volume). In the present study, there was little evidence that having milder social impairment and/or having PDD-NOS increased the likelihood of following a more severe trajectory for any one behavior. Perhaps the impact of having milder ASD only emerges when IS behaviors are aggregated together, as in the earlier paper. Consistent with previous studies, having persistent/increasing IS behaviors over time was also not

associated with having a higher NVIQ score (Szatmari et al., 2006; Hus, Pickles, Cook, Risi, & Lord, 2007). There is insufficient evidence, at this point, to say that these behaviors truly are associated with higher levels of functioning. It might be useful to look at other indices of functioning, such as Verbal IQ scores or scores on measures of adaptive functioning or aberrant behavior.

Autism Specificity of RRBs Across Development

By taking developmental patterns into account, we were able to learn more about the autism specificity of RRBs. What seems to be particular to ASD is not the presence of any particular behavior, but rather specific patterns of change in certain behaviors. For example, although RSM behaviors were more common in ASD than in DD at all ages, they were nevertheless quite prevalent in the DD sample, particularly in later childhood. None of these behaviors on its own, then, was highly indicative of ASD. However, the consistent severity of these behaviors over time *was* particular to the ASD sample. Very few children with DD fell into the consistently severe trajectory that emerged for the RSM behaviors. Children with DD also commonly lost RSM behaviors over time, whereas this pattern was less common in the ASD sample. IS behaviors tended to become more common over time both in children with ASD and DD. Thus, although the presence of such behaviors early in development was indicative of ASD, their absence at young ages was not particularly meaningful. When considering whether a particular behavior is a ‘red flag’ for ASD, then, various issues need to be considered, including what kind of behavior it is, how impairing it is, and how consistently it has been present.

Limitations and Future Directions for Research

The findings on RRBs presented here are based solely on parent report. This presents a number of limitations, as scoring on the ADI-R is dependent, to some extent, on the manner in which the parent describes the behavior. In a longitudinal study, scores for some behaviors might improve, in part, because the parent has adjusted to the child’s behavior and no longer sees it as severely impairing the child’s or family’s functioning. Although the ADI-R includes probes designed to obtain detailed information about behaviors so that the examiner can score behaviors based more on evidence than on

parent perspective, it is nevertheless crucial to conduct longitudinal studies of RRBs that involve direct observation of these behaviors, in order to see if similar results are obtained. In the present study, there was some evidence that maternal education might be associated with RRBs, although the nature of this relationship was unclear. If it can be established that this association holds even when RRBs are measured through observation, then this would suggest that there is a real effect that goes beyond differences in reporting style that might relate to education level.

We also found a relationship between gender and RRB development that needs to be replicated with observational data. Males were more likely to have relatively severe behaviors that persisted over time, compared to females. This is consistent with a study by Lord, Schopler, & Revicki (1982), which found that males were more likely to show unusual visual interests and repetitive play, based on direct observation. These findings are particularly interesting given the consistent trend for females with ASD to have greater cognitive impairment than males (Volkmar, Szatmari, & Sparrow, 1993). If certain ‘low level’ RRBs are indeed associated with being male, this seems to be in spite of cognitive functioning, rather than because of it. The relationship between gender and RRBs, particularly those involving a repetitive sensory and/or motor component, clearly deserves further study.

The degree to which our sample is representative of current samples of children referred for ASD is also an issue. This study began at a time when it was rare for children to be referred for a diagnosis of autism at a young age, whereas today, children are increasingly referred for diagnosis at young ages. Consequently, it is likely that a higher proportion of the children with ASD in the present study were more severely affected than children referred for a diagnosis today. Similarly, the DD subgroup is not necessarily representative of children with a particular nonspectrum developmental disorder. Rather, it is a heterogeneous group of children whose common characteristic is that they were not diagnosed with ASD. Some of the children diagnosed with DD at age 2 were children who had been referred due to concerns about ASD. Although it was decided that these children did not have ASD, the fact that there were concerns suggests these children might not be prototypical of children who are not on the autism spectrum. The finding that RSM and IS behaviors were generally correlated in children with DD, as

they were in children with ASD, is consistent with this claim, particularly since this was true at all ages except age 5, when the DD group was comprised mostly of children referred for DD, and therefore a ‘cleaner’ DD sample. It is essential to conduct studies of RRBs with ASD that include relevant control groups, such as children with mental retardation of unknown etiology, or children with Down Syndrome. Children with disorders that involve RRBs, such as OCD and Tourette Syndrome might also be appropriate control groups for studies of RRBs in children with ASD who are not cognitively impaired.

Finally, we were not able to assess the effects of treatment was limited in the present study. Future work should examine both whether the intensity of treatment, as well as the modality (e.g. Applied Behavioral Analysis versus play-based therapy) have any effect on trajectories of RRB development. Ideally, researchers interested in the effects of treatment should conduct prospective, longitudinal studies in which data about RRBs are collected both through parent report and careful observation, and are measured both before and after treatment.

Conclusion

The overall objective of this paper was to enhance our understanding of RRBs by considering how development affects each behavior that falls into this eclectic category. Given the variability both within the same behavior over time and between different RRBs, taking the role of development into account and focusing in on individual behaviors helped us identify patterns that otherwise would not have been apparent. The relatively narrow focus of this paper thereby helped to clarify the ‘big picture’ of RRBs in ASD. Focused examinations of the development of specific social and communication difficulties in ASD would likely reveal similarly important patterns that, taken together with the findings from the present study, would contribute to our overall understanding of ASD.

Table 4.1. Prevalence of RRBs on the ADI-R by Cohort and Diagnosis at Age 2

ADI-R RRB item	ASD			DD		
	2 (n = 161)	5 (n = 96)	9 (n = 129)	2 (n = 53)	5 (n = 39)	9 (n = 42)
RSM Behaviors						
Unusual sensory interests	76.4	72.2	65.3 ^a	50.9 ^d	39.4 ^d	53.7
Repetitive use of objects	78.6	63.2 ^a	58.7 ^a	44.2 ^d	26.3 ^d	37.5
Complex mannerisms	60.0	50.0	56.1	26.4 ^d	36.8	29.3 ^d
Hand and finger mannerisms	53.8	58.9	64.2	22.6 ^d	24.3 ^d	36.6 ^d
IS Behaviors						
Difficulties with changes in routine	30.0 ^{bc}	52.2	55.6	17.3 ^c	16.2 ^{cd}	36.6
Compulsions/rituals	18.8 ^{bc}	37.8	49.2	11.3 ^c	15.8 ^d	29.3 ^d
Resistance to trivial changes in environment	9.4 ^{bc}	22.2	23.8	5.8	8.1	9.8
Other Behaviors						
Unusual preoccupations	40.6	43.3	34.4	15.1 ^d	10.5 ^d	22.0
Sensitivity to noise	35.6 ^{bc}	57.8	55.7	23.1	35.1 ^d	39.0
Self-injurious behaviors	28.6 ^b	32.2	37.4	37.7	36.8	34.1

^a less than cohort 2, $p < .05$

^b less than cohort 5, $p < .05$

^c less than cohort 9, $p < .05$

^d less than ASD same cohort, $p < .05$

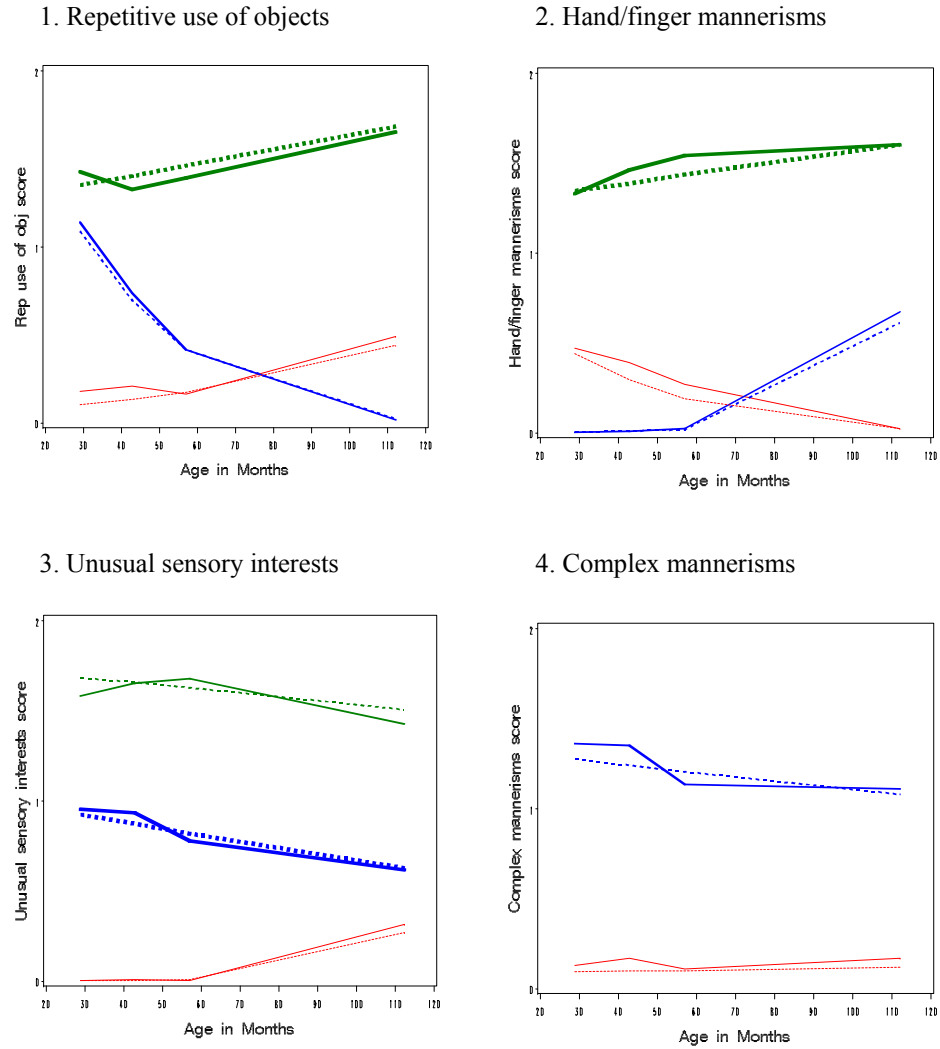
Table 4.2. Trajectory groups for RSM Behaviors: ASD and DD Combined Sample

	Trajectory descriptions (N / % of sample)	Parameter Estimates (SE)
Unusual sensory interests	Consistently Mild (32/15.0%)	.04 (.02)
	Improving (120/56.1%)	-.006 (.003)*
	Consistently Severe (62/29.0%)	-.005 (.004)
Repetitive use of objects	Consistently Mild (42/19.6%)	.01 (.01)
	Improving (67/31.3%)	-.04 (.01)***
	Consistently Severe (105/49.1%)	.005 (.002)*
Hand/finger mannerisms	Improving (40/18.7%)	-.03 (.01)*
	Worsening (68/31.8%)	.04 (.02)**
	Consistently Severe (106/49.5%)	.004 (.002)
Complex mannerisms	Consistently Mild (78/36.4%)	.002 (.008)
	Consistently Severe (136/63.6%)	-.004 (.002)

Note Unless otherwise indicated, significant estimates are for linear terms.

* $p < .05$; ** $p < .01$; *** $p < .001$

Figure 4.1. Trajectory Groups for RSM Behaviors: ASD and DD Combined Sample



Note Dashed line – predicted trajectory; solid line – observed trajectory.
 Thickness of line indicates relative proportion of sample in trajectory group.

Table 4.3. Child Characteristics Affecting Likelihood of Group Assignment for RSM Behaviors

	Trajectory descriptions	Grp 3 vs Grp 1 (SE)	Grp 3 vs Grp 2 (SE)	Grp 2 vs Grp 1 (SE)
Unusual sensory interests	Grp 1: Consistently Mild Grp 2: Improving Grp 3: Consistently Severe	Dx: -2.72 (.75)*** NVIQ: -.11 (.03)*** SA: .25 (.08)**	Dx: -1.25 (.53)* NVIQ: -.07 (.02)** SA: .14 (.06)*	Dx: -1.57 (.51)** NVIQ: -.05 (.02)**
Repetitive use of objects	Grp 1: Consistently Mild Grp 2: Improving Grp 3: Consistently Severe	Dx: -2.72 (-.85)** NVIQ: -.08 (.02)** Gender: 3.47 (1.38)* SA: .33 (.09)***	Dx: -1.04 (.45)* NVIQ: -.05 (.02)* SA: .16 (.08)*	Dx: -1.68 (.80)* Gender: 2.86 (1.28)* Site: 2.25 (.99)*
Hand/finger mannerisms	Grp 1: Improving Grp 2: Worsening Grp 3: Consistently Severe	Dx: -.99 (.37)** NVIQ: -.05 (.02)** SA: .19 (.07)**	Dx: -1.32 (.36)*** NVIQ: -.05 (.02)** SA: .18 (.06)**	
Complex mannerisms	Grp 1: Consistently Mild Grp 2: Consistently Severe	n/a	n/a	Dx: -1.16 (.29)*** NVIQ: -.04 (.01)** SA: .13 (.04)**

Note Dx = Age 2 Diagnosis; NVIQ = Age 2 Nonverbal IQ Score; SA = Age 2 ADOS Social Affect Score

* $p < .05$; ** $p < .01$; *** $p < .001$

Table 4.4. Stability of RSM behaviors for Children with ASD and DD

	ASD		DD	
	2 to 5	5 to 9	2 to 5	5 to 9
unusual sensory interests	80.6/47.8*	78.4/35.3	52.6/26.3	72.7/45.0
repetitive use of objects	69.8/45.8	73.8/45.5	33.3/18.2	55.6/33.3
hand/finger mannerisms	79.2/35.7	81.1/46.7*	50.0/19.4	50.0/31.8
complex mannerisms	58.0/40.0	73.5/39.4	77.8/21.4	50.0/5.9

Note Number before slash = Percentage of children who had the behavior at Time 1 and continued to have it at Time 2
 Number after slash = Percentage of children who did not have the behavior at Time 1 but had the behavior at Time 2

* $p < .05$

Table 4.5. Trajectory Groups for IS Behaviors: ASD and DD Combined Sample

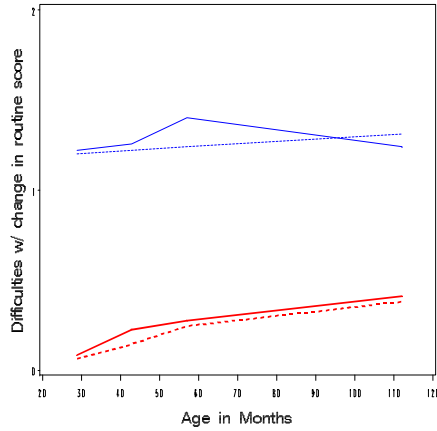
	Trajectory Descriptions (N / % of sample)	Parameter Estimates
Difficulties with changes in routine	Worsening (146/68.2%)	.07 (.02)*** -.0003 (.0001)* (quadratic)
	Consistently Severe (68/31.8%)	.002 (.003)
Resistance to changes in environment	Consistently absent (178/83.2%)	-3.50 (.61)*** (intercept)
	Worsening (36/16.8%)	.02 (.006)*
Compulsions/rituals	Worsening (135/63.1%)	.21 (.09)* -.001 (.0005) (quadratic)*
	Worsening between 2 and 5 years (79/36.1% 214)	.06 (.02)** -.0004 (.0002)*

Note Unless otherwise indicated, significant estimates are for linear terms.

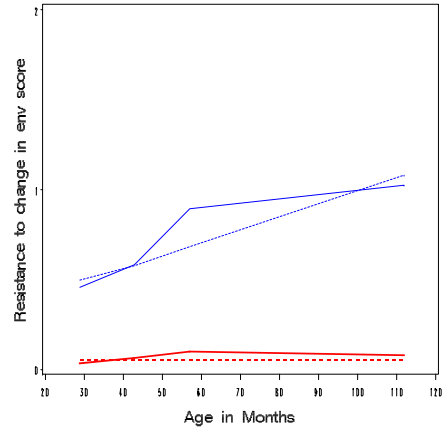
* $p < .05$; ** $p < .01$; *** $p < .001$

Figure 4.2. Trajectory Groups for IS Behaviors: ASD and DD Combined Sample

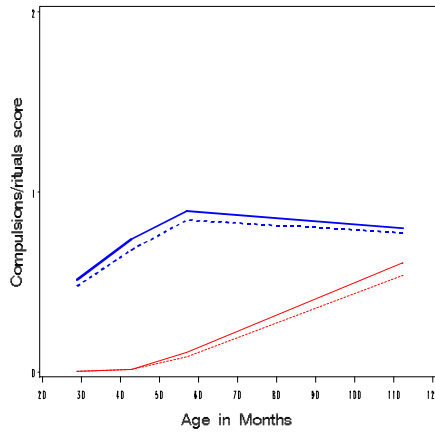
1. Difficulties with changes in routine



2. Resistance to changes in environment



3. Compulsions and rituals



Note Dashed line – predicted trajectory; solid line – observed trajectory.
 Thickness of line indicates relative proportion of sample in trajectory group.

Table 4.6. Child Characteristics Affecting Likelihood of Group Assignment for IS Behaviors

	Trajectory Descriptions	
Difficulties with changes in routine	Grp 1: Worsening Grp 2: Consistently Severe	Gender: 1.15 (.57)*
Resistance to changes in environment	Grp 1: Consistently zero Grp 2: Worsening	Dx: -.78 (.34)*
Compulsions/rituals	Grp 1: Worsening Grp 2: Worsening between 2 and 5 years	Educ: -.99 (.49)*

Note Dx = Age 2 Diagnosis; Educ = Maternal Level of Education

* $p < .05$

Table 4.7. Stability of IS Behaviors for Children with ASD and DD

	ASD		DD	
difficulties with changes in routine	72.4/41.7	80.6/38.7***	33.3/14.7	60.0/28.0
resistance to changes in environment	66.7/19.3*	40.0/17.7	0.0/8.57	0.0/7.41
compulsions/rituals	68.4/28.6	51.7/51.3	66.7/11.4	50.0/32.0

Note Number before slash = Percentage of children who had the behavior at Time 1 and continued to have it at Time 2

Number after slash = Percentage of children who did not have the behavior at Time 1 but had the behavior at Time 2

* $p < .05$; ** $p < .01$; *** $p < .001$

Table 4.8. Trajectory Groups for ‘Other’ Behaviors: ASD and DD Combined Sample

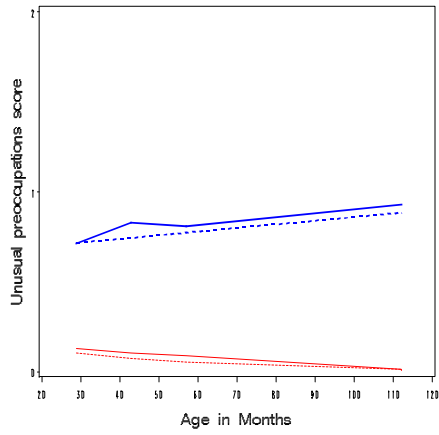
	Trajectory descriptions	Parameter Estimates (SE)
Unusual preoccupations	Consistently Mild (n= 89)	-.02 (.02)
	Consistently Moderate (n = 125)	.004 (.003)
Self-injury	Consistently absent (n=131)	-1.44(.23) (intercept)***
	Consistently moderate (n=83)	.004 (.002)
Sensitivity to noise	Worsening (n = 108)	.01 (.004)*
	Worsening between 2 and 5 years (n = 106)	.09 (.02)*** -.0006 (.0001) *** (quadratic)

Note Unless otherwise indicated, significant estimates are for linear terms.

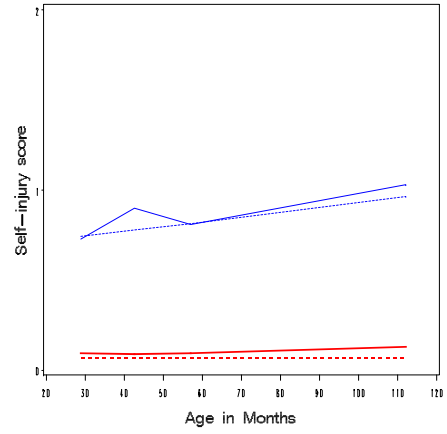
* $p < .05$; ** $p < .01$; *** $p < .001$

Figure 4.3. Trajectory Groups for ‘Other’ Behaviors: ASD and DD Combined Sample

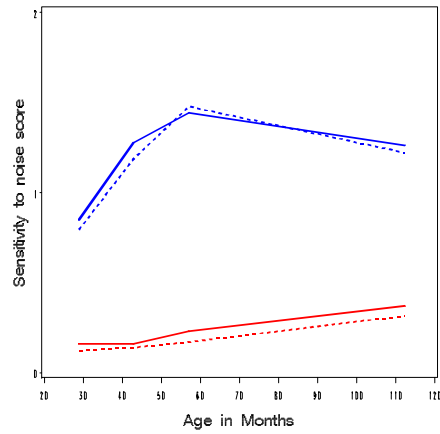
1. Unusual preoccupations



2. Self-injury



3. Sensitivity to noise



Note Dashed line – predicted trajectory; solid line – observed trajectory.
Thickness of line indicates relative proportion of sample in trajectory group.

Table 4.9. Child Characteristics Affecting Likelihood of Group Assignment for ‘Other’ Behaviors

	Trajectory Groups	Significant Covariates (Grp 2 vs. Grp 1)
Unusual preoccupations	Grp 1: Consistently Mild Grp 2: Consistently Moderate	Dx: -1.25 (.34)*** NVIQ: -.03 (.01)* Gender: 1.67 (.64)**
Self-injury	Grp 1: Consistently zero Grp 2: Consistently moderate	NVIQ: -.03 (.01)**
Sensitivity to noise	Grp 1: Worsening Grp 2: Worsening between 2 and 5 years	Dx: -.68 (.27)* Educ: 1.10 (.44)*

Note Dx = Age 2 Diagnosis; NVIQ = Age 2 Nonverbal IQ score; Educ = Maternal Level of Education

* $p < .05$; ** $p < .01$; *** $p < .001$

Table 4.10. Stability of ‘Other ’ Behaviors for Children with ASD and DD

	ASD		DD	
unusual preoccupations	47.2/40.7	43.3/36.1	50.0/3.1	25.0/18.5
self-injury	59.6/26.3	52.6/31.3*	50.0/29.2	58.3/21.1*
sensitivity to noise	76.3/43.1	56.4/41.4	30.0/37.0	54.6/21.1

Note Number before slash = Percentage of children who had the behavior at Time 1 and continued to have it at Time 2
Number after slash = Percentage of children who did not have the behavior at Time 1 but had the behavior at Time 2

* $p < .05$

References

- American Psychiatric Association. (1994). *Diagnostic and Statistical Manual of Mental Disorders* (4th ed.). Washington DC: American Psychiatric Association.
- Anderson, D., Lord, C., Risi, S., DiLavore, P., Thurm, A., Shulman, C., & Pickles, A. (in press). Patterns of growth in verbal abilities among children with autism spectrum disorder. *Journal of Clinical Child Psychology*.
- Bishop, S. L., Richler, J., Cain, A. C., & Lord, C. (submitted). Parental perceptions of raising a child with Autism Spectrum Disorder (ASD).
- Bryson, S. E., Zwaigenbaum, L., Brian, J., Roberts, W., Szatmari, P., Rombough, V., & McDermott, C. (2007). A prospective case series of high-risk infants who developed autism. *Journal of Autism and Developmental Disorders*, *37*, 12-24.
- Cox, A., Klein, K., Charman, T., Baird, G., Baron-Cohen, S., Swettenham, J., Drew, A., Wheelwright, S., & Nightingale, N. (1999). Autism spectrum disorders at 20 and 42 months of age: Stability of clinical and ADI-R diagnosis. *Journal of Child Psychology and Psychiatry*, *40*, 719-732.
- Cuccaro, M. L., Shao, Y., Grubber, J., Slifer, M., Wolpert, C. M., Donnelly, S. L., Abramson, R. K., Ravan, S. A., Wright, H. H., DeLong, R. G., & Pericak-Vance, M. A. (2003). Factor analysis of restricted and repetitive behaviors in autism using the Autism Diagnostic Interview-R. *Child Psychiatry and Human Development*, *34*, 3-17.
- Epstein, L. J., Taubman, M. T., & Lovaas, O. I. (1985). Changes in self-stimulatory behaviors with treatment. *Journal of Abnormal Child Psychology*, *13*, 281-293.
- Gabriels, R. L., Cuccaro, M. L., Hill, D. E., Ivers, B. J., & Goldson, E. (2005). Repetitive behaviors in autism: Relationships with associated clinical features. *Research in Developmental Disabilities*, *26*, 169-181.
- Hus, V., Pickles, A., Cook, E. H., Risi, S., & Lord, C. (2007). Using the autism diagnostic interview-revised to increase phenotypic homogeneity in genetic studies of autism. *Biological Psychiatry*, *61*, 438-448.
- Jones, B. L., Nagin, D. S., & Roeder, K. (2001). A SAS procedure based on mixture models for estimating developmental trajectories. *Sociological Methods & Research*, *29*, 374-393.
- Lord, C., Risi, S., DiLavore, P. S., Shulman, C., Thurm, A., & Pickles, A. (2006). Autism from 2 to 9 years of age. *Archives of General Psychiatry*, *63*, 694-701.
- Lord, C., Rutter, M., DiLavore, P. C., & Risi, S. (1999). *Autism Diagnostic Observation Schedule*. Los Angeles: Western Psychological Services.
- Lord, C., Rutter, M., & Le Couteur, A. (1994). Autism Diagnostic Interview--Revised: A revised version of a diagnostic interview for caregivers of individuals with possible pervasive developmental disorders. *Journal of Autism and Developmental Disorders*, *24*, 659-685.
- Lord, C., Schopler, E., & Revicki, D. (1982). Sex differences in autism. *Journal of Autism & Developmental Disorders*, *12*, 317-330.

- Moore, V., & Goodson, S. (2003). How well does early diagnosis of autism stand the test of time? Follow-up study of children assessed for autism at age 2 and development of an early diagnostic service. *Autism, 7*, 47-63.
- Richler, J., Bishop, S. L., Kleinke, J., & Lord, C. (2007). Restricted and repetitive behaviors in young children with autism spectrum disorders. *Journal of Autism and Developmental Disorders, 37*, 73-85.
- Sparrow, S. S., Balla, D. A., & Cicchetti, D. V. (1984). *Vineland Adaptive Behavior Scales*. Circle Pines, MN: American Guidance Service Inc.
- Szatmari, P., Georgiades, S., Bryson, S., Zwaigenbaum, L., Roberts, W., Mahoney, W., Goldberg, J., & Tuff, L. (2006). Investigating the structure of the restricted, repetitive behaviours and interests domain of autism. *Journal of Child Psychology and Psychiatry, 47*, 582-590.
- Turner, M. (1999). Annotation: Repetitive behaviour in autism: a review of psychological research. *Journal of Child Psychology and Psychiatry, 40*, 839-849.
- Volkmar, F., Szatmari, P., & Sparrow, S. S. (1993). Sex differences in pervasive developmental disorders. *Journal of Autism and Developmental Disorders, 23*, 579-591.

Chapter V

Conclusion

The findings from this series of studies highlight the importance of identifying subtypes and considering development in understanding RRBs in children with Autism Spectrum Disorders (ASD). Ultimately, these two concepts may prove crucial to identifying the causes of RRBs and the most effective ways of reducing them.

The Importance of Subtyping

The studies that have been presented here add to the mounting evidence that RRBs should be thought of as being comprised of at least two major subtypes, rather than as a single category. At first glance, subcategorizing RRBs might seem to be merely a different way of grouping the same behaviors. However, the way in which we classify RRBs is far from trivial. Understanding the common features that RRBs share will likely prove instrumental in identifying common etiological mechanisms for these behaviors in children with ASD. For example, Rett's Disorder, another one of the Pervasive Development Disorders, is characterized by very distinctive hand mannerisms, such as hand-wringing and clapping. Researchers have identified a mutation in the MECP2 gene that is responsible for most cases of this disorder, and have found that mice expressing a truncated version of the MECP2 protein exhibit hand stereotypies very similar to those in Rett's Disorder (Lewis, Tanimura, Lee, & Bodfish, 2007). Identifying behaviors that behaviors 'go together' developmentally in ASD might help us isolate the causes of such behaviors.

RRBs that have similar causes might also be treated most appropriately using similar forms of intervention. There is substantial evidence that humans and animals raised in restricted environments and/or subjected to early social deprivation suggests are prone to stereotyped behaviors, such as repetitive motor mannerisms (Mason, 1991). Such studies are relevant to children with ASD, who likely experience a restricted,

socially-deprived environment from a young age, given their social and communication deficits. One implication for treatment is that stereotyped behaviors might be reduced by enriching the child's environment, particularly in terms of social inputs (Lewis et al., 2007). Behaviors characterized by insistence on sameness, such as adherence to non-functional routines, might be more closely associated with deficits in executive function, particularly cognitive rigidity (Lopez, Lincoln, Ozonoff, & Lai, 2005). Interventions that focus on increasing cognitive flexibility might be most appropriate for addressing these problematic behaviors.

The growing trend for early identification of ASD might further the goal of identifying the causes of RRBs, which in turn might aid in the development of appropriate intervention. If we can ascertain children at risk for ASD at very young ages and follow their development prospectively, we might be able to identify behaviors that precede RRBs and determine if such behaviors are predictive of RRBs. We might even be able to identify 'proto-RRBs,' behaviors that are not currently thought of as belonging to this category, but that seem to share features with RRBs, such as difficulty with disengagement of visual attention, which has been found to distinguish infants later diagnosed with ASD from infants with typical development or nonspectrum disorders, such as language delay (Zwaigenbaum et al., 2005). In this way, we might be able to further efforts for even earlier identification and treatment of ASD.

The consistent finding that there are subdomains of RRBs also points to a need to revise diagnostic criteria for ASD. The current diagnostic criteria state that one behavior from the entire category of RRBs is sufficient for a diagnosis of autism, provided deficits in social impairment and communication are also present (American Psychiatric Association, 1994). However, we know from this series of studies, as well as from previous work, that certain behaviors, particularly those in the 'repetitive sensorimotor' category, are not specific to children with ASD. For this reason, no one RRB should 'rule in' a diagnosis. Rather, if we are going to use RRBs to inform diagnosis, we need to look at more specific features of the behaviors, such as their severity and persistence over time.

The Importance of Development

The findings from these studies also point to the need to think of RRBs as dynamic, changing along with other aspects of development. Consideration of developmental trajectories is particularly important for the identification of phenotypes within ASD. Behaviors such as repetitive use of objects are common in children with ASD at young ages, regardless of factors such as IQ and degree of social impairment. However, as children with ASD get older, some begin to show some improvement in these behaviors, experiencing less impairment from them, while others show little change. Similarly, although certain RRBs tend to be rare in young children with ASD and only emerge later in development, some children have them even when they are young. Perhaps these different ‘profiles’ constitute different phenotypes of ASD that might be more meaningful than classifications such as ‘autism’ and ‘PDD-NOS.’ Isolating relatively homogeneous subgroups within ASD is essential for identifying the genes associated with the disorder, but must be done within a developmental framework.

Taking a developmental perspective on RRBs is also necessary if we want identify factors, such as experience, gene expression, and neurological structure that give rise to these behaviors. More specifically, we need to understand how these different factors interact to affect development. It is commonly accepted in the field of developmental psychopathology that there is no single underlying cause for atypical development, but rather a combination of variables that leads an individual down a particular developmental pathway (Cicchetti & Sroufe, 2000). As Walden and Hurley (2006) point out, children with autism might have a genetic predisposition to display certain behaviors, such as stereotypies. These behaviors, in turn, elicit a particular social response from others, which might actually alter gene expression, thereby changing the developmental course of the atypical behavior. Understanding how behaviors such as RRBs change over time is only the first step. Next, we must learn what factors cause this change. Cicchetti and Sroufe (2000) provide the example of studies of depression, which have consistently found that negative attributional biases are both correlates of depression and predictors of later depressive symptoms. This is an important contribution to our understanding of depression, but researchers now need to determine how such biases develop in the first place.

Summary

Despite advances in our understanding of ASD, much about the disorder remains a mystery. The eclectic mix of symptoms known as restricted and repetitive behaviors and interests is an intriguing part of the enigma of ASD. These behaviors are core to the disorder, but it is not clear exactly how they fit with the other core features. They change over the course of development, but we do not know what causes these changes. They are among the most disruptive behaviors in ASD, but we know very little about how to reduce them. The hope is that this group of studies provides a small step in addressing these larger questions, by describing RRBs and their development in rich detail. Now that the ‘what’ of RRBs has been addressed, we can begin to tackle the ‘why.’

References

- Cicchetti, D., & Sroufe, L. A. (2000). The past as prologue to the future: The times, they've been a-changin' [Editorial]. *Development & Psychopathology*, *12*, 255-264.
- Lewis, M. H., Tanimura, Y., Lee, L. W., & Bodfish, J. W. (2007). Animal models of restricted and repetitive behavior in autism. *Behavioural Brain Research*, *176*, 66-74.
- Lopez, B. R., Lincoln, A. J., Ozonoff, S., & Lai, Z. (2005). Examining the relationship between executive functions and restricted, repetitive symptoms of Autistic Disorder. *Journal of Autism and Developmental Disorders*, *35*, 445-460.
- Mason, W. A. (1991). Effects of social interaction on well-being - development aspects. *Laboratory Animal Science*, *4*, 323-328.
- Walden, T. A., & Hurley, J. J. (2006). A developmental approach to understanding atypical development. In T. Charman & W. Stone (Eds.), *Social and Communication Development in Autism Spectrum Disorders* (pp. 269-295). New York: Guilford.
- Zwaigenbaum, L., Bryson, S., Rogers, T., Roberts, W., Brian, J., & Szatmari, P. (2005). Behavioral manifestations of autism in the first year of life. *International Journal of Developmental Neuroscience*, *23*, 143-152.