Repetitive Behavior in 12-Month-Olds Later Classified With Autism Spectrum Disorder

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Objective: As compared to the utility of early emerging social communicative risk markers for predicting a later diagnosis of autism spectrum disorder (ASD), less is known about the relevance of early patterns of restricted and repetitive behaviors. We examined patterns of stereotyped motor mannerisms and repetitive manipulation of objects in 12-month-olds at high and low risk for developing ASD, all of whom were assessed for ASD at 24 months. **Method:** Observational coding of repetitive object manipulation and stereotyped motor behaviors in digital recordings of the Communication and Symbolic Behavior Scales was conducted using the Repetitive and Stereotyped Movement Scales for 3 groups of 12-month-olds: low-risk infants (LR, n = 53); high–familial-risk infants who did not meet diagnostic criteria for ASD at 24 months (HR-negative, n = 75); and high−familial-risk infants who met diagnostic criteria for ASD at 24 months (HR-ASD, n = 30). **Results:** The HR-ASD group showed significantly more stereotyped motor mannerisms than both the HR-negative group ($p = .025$) and the LR group ($p = .001$). The HR-ASD and HR-negative groups demonstrated statistically equivalent repetitive object manipulation scores ($p = .431$), and both groups showed significantly more repetitive object manipulation than the LR group ($p < .040$). Combining the motor and object stereotypy scores into a Repetitive and Stereotyped Movement Scales (RSMS) composite yielded a disorder-continuum effect such that each group was significantly different from one another (LR < HR-negative < HR-ASD). **Conclusion:** These results suggest that targeted assessment of repetitive behavior during infancy may augment early ASD identification efforts. J. Am. Acad. Child Adolesc. Psychiatry, 2014;53(11):1216–1224. **Key Words:** autism, repetitive behavior, motor stereotypies, infant siblings, development

Accumulating evidence suggests that a number of social communicative risk markers observed as early as 12 months of age distinguish infants who will later meet diagnostic criteria for autism spectrum disorder (ASD).[1-5] Comparatively less is known about whether restricted and repetitive behaviors (RRBs) represent similar levels of risk for a later diagnosis. Comprehensive characterization of restricted and repetitive behavior at 12 months has important diagnostic implications[6] and is also relevant for developmental models of pathogenesis.

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In a cohort of 18- to 24-month-old toddlers, ascertained via a general population screening of health care and childcare agencies, Wetherby *et al.*[7-9] observed that repetitive object manipulation and stereotyped motor behaviors differentiated toddlers with ASD from typically developing toddlers and toddlers with developmental delays. These data complement and extend evidence based on parent-report and observational diagnostic tools that highlight the importance of repetitive behaviors for distinguishing toddlers and preschool-aged children with ASD from comparison groups.[10-14] Lower-order repetitive behaviors[15] observed in clinically ascertained samples and repetitive use of objects in particular represent early emerging and persistent features of ASD

present from 2 to 9 years of age,[14] and the severity of these behaviors at 2 years of age predicts prognosis 7 years later.[16]

Comparing children with developmental delays to children with ASD has elucidated aspects of RRBs that are specific to the ASD phenotype. However, additional comparison groups are needed to determine whether detailed characterization of RRBs might inform the underlying biological architecture of ASD. For example, Wolff *et al.*[17] observed differences in repetitive behavior profiles between preschool-aged boys with idiopathic autism and boys with fragile X syndrome who also met diagnostic criteria for ASD. Evidence from family studies of affected sibling pairs suggests that higher-order repetitive behaviors tend to aggregate in families.[18,19] Further evidence from a dense, extended pedigree study suggests unique linkage signals for RRBs.[20] Another family study examining a dimensional index of motor functioning found striking similarities in the degree of motor impairment among affected sibling pairs, 83% of whom had scores of at least 1 SD below the general population mean.[21] However, only 6% of unaffected siblings of probands with ASD performed below 1 SD of the population mean.

Several prospective studies of infants at high familial risk for developing ASD have examined RRBs. In these studies, high risk is defined as having an older sibling diagnosed with ASD, and low risk is defined as having a typically developing older sibling and no first- or second-degree relatives with autism. Results from a micro-behavioral coding analysis in which 4 objects were briefly presented to high- and low-risk infants suggested that those who later developed ASD (n = 9) showed specific patterns of unusual visual and manual exploration of objects at 12 months of age.[22] The comparison groups in this study included an "other delays" group (n = 10) and a "no concerns" group (n = 47). In another study, high-risk 18-month-olds later diagnosed with ASD (n = 17), high-risk siblings who showed other delays (n = 12), and high-risk siblings who showed no developmental delays (n = 19) demonstrated more nonfunctional repetitive play acts with objects than low-risk toddlers.[23] Although comparisons were not conducted between the high-risk subgroups, this analysis suggests that nonfunctional repetitive play may be a familial marker of ASD. Atypical motor mannerisms (e.g., arm waving) have differentiated 18-month-olds who later developed ASD (n = 8) from low-risk infants and high-risk infants who did not develop ASD; however, this pattern was not observed at 12 months of age.[24] An additional preliminary study examining the rate and inventory of object and motor stereotypies in 15-month-olds points to the possibility that high-risk infants who do not develop ASD (n = 12) show higher rates of these behaviors than low-risk infants.[25] In sum, findings to date suggest that some features of repetitive behavior may emerge as early as 12 or 18 months of age in children who develop ASD. However, small sample sizes, investigations of isolated features (i.e., either repetitive object manipulation or stereotyped motor behaviors), and decisions to merge high-risk and low-risk groups have limited the impact of findings to date.

In the current study, we used a standardized behavioral coding scheme[7] to examine patterns of motor stereotypies and repetitive object manipulation across 3 groups of 12-month-olds that maintains the integrity of the family design: high-risk infants later diagnosed with ASD; high-risk infants who did not meet diagnostic criteria for ASD; and low-risk infants. The primary objective was to characterize patterns of RRBs representative of either a disorder-specific deficit or a pattern representing familial liability for ASD in a large sample of 12-month-olds (N = 158).

METHOD

This study took place in the context of an ongoing Autism Center of Excellence Network study (the Infant Brain Imaging Study [IBIS]) prospectively investigating longitudinal brain and behavioral trajectories in high- and low-risk infants. The institutional review boards at all sites approved the research protocol, and parents provided informed consent for their infants to participate. Additional information on the study design is provided by Elison *et al.*[26] and Wolff *et al.*[27]

Participants

All infants were assessed at 1 of 4 clinical sites: the University of North Carolina at Chapel Hill, the University of Washington, the Children's Hospital of Philadelphia, and Washington University in St. Louis. The study sample included children with a digital video recording of the Communication and Symbolic Behavior Scales–Developmental Profile (CSBS-DP)[28] collected at the 12-month visit, and clinical outcome data at the 24-month visit. Exclusion criteria for both high- and low-risk infants included the following: history of known genetic conditions or syndromes