

# Play and Developmental Outcomes in Infant Siblings of Children with Autism

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**Abstract** We observed infant siblings of children with autism later diagnosed with ASD (ASD siblings;  $n = 17$ ), infant siblings of children with autism with and without other delays (Other Delays and No Delays siblings;  $n = 12$  and  $n = 19$ , respectively) and typically developing controls (TD controls;  $n = 19$ ) during a free-play task at 18 months of age. Functional, symbolic, and repeated play actions were coded. ASD siblings showed fewer functional

and more non-functional repeated play behaviors than TD controls. Other Delays and No Delays siblings showed more non-functional repeated play than TD controls. Group differences disappeared with the inclusion of verbal mental age. Play as an early indicator of autism and its relationship to the broader autism phenotype is discussed.

**Keywords** Autism spectrum disorders · Functional play · Symbolic play · Repetitive behaviors · Play · Infant siblings of children with autism

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## Introduction

Play serves an important role in the social communication impairments that are central to autism spectrum disorders. Play is significantly associated with receptive and expressive language skills and with the development of appropriate social relationships and engagement with peers in children with autism (Charman et al. 2000; Clift et al. 1988; Doswell et al. 1994; Lewis et al. 2000; Mundy et al. 1987; Sigman and Ruskin 1999; Tamis-LeMonda and Bornstein 1994). Accordingly, early play behaviors serve as important predictors as well as points of intervention for later language and social development. Moreover, the study of early play behaviors may elucidate basic impairments in symbolic representation (Lewis 2003) or other common mechanisms that underlie later social communication deficits, and thus, clarify the relationship between play and language in autism.

We observed the play behaviors of infants in a standardized free-play assessment performed at 18 months of age. Infant siblings of children with autism were divided into three subgroups: infant siblings later diagnosed with autism spectrum disorders (ASD siblings), infant siblings

with other deficits in cognition, language and social behavior (Other Delays siblings), and infant siblings later evaluated as typically developing (No Delays siblings). The comparison group consisted of typically developing controls (TD controls) who did not have a family history of autism spectrum disorders.

There are three domains of play defined in the literature: sensorimotor manipulation, functional play, and symbolic or imaginary play. Sensorimotor play involves the simple manipulation of objects, or play focused on the physical attributes of objects (Doherty and Rosenfeld 1984; Lifter et al. 1993; Sigman and Ungerer 1981, 1984). Functional play is the “appropriate use of an object or the conventional association of two or more objects, such as a spoon to feed a doll, or placing a teacup on a saucer” (Sigman and Ungerer 1981). Symbolic play is characterized by an underlying complex representation of objects, and thus the ability to pretend an object is present when it is not or to extend the function of one object to another object (Leslie 1987). Accordingly, symbolic play is often demonstrated through one of three types of actions: substitution (or the use of one object as another); imaginary play (the attribution of false properties to an object or the imagined presence of an absent object); and agent play (in which a doll or similar object becomes the agent of an action) (Leslie 1987; Sigman and Ungerer 1984). In the development of play behaviors, children typically progress from sensorimotor play to functional play and finally, to symbolic or imaginary play (Lifter et al. 1993). Thus, children’s play behaviors reveal the level of sophistication with which they are interacting with their environment and the extent to which they understand the world around them—whether their understanding is purely physical (sensorimotor play) or representational (symbolic play) (Casby 2003).

Children with autism tend not to engage in symbolic play spontaneously and do not produce as many symbolic play actions as typically developing children when prompted (Jarrold 2003; Jarrold et al. 1993; Wulff 1985). Children with autism may engage in symbolic play that is stereotyped and repetitive, for example, acting out scripts with dolls or stuffed animals (Wing et al. 1977). Deficits in symbolic play appear to be specific to individuals with autism and not characteristic of individuals with other developmental disabilities (Mundy et al. 1986). Children with autism also show deficits in functional play (Sigman and Ungerer 1981; Williams et al. 2001). In particular, children with autism appear to perform fewer functional play actions and integrated sequences of functional acts, and spend less time engaging in functional play than children with Down syndrome and typically developing children (Williams et al. 2001).

In addition to previous studies of older children with autism, recent longitudinal studies also find evidence for

deficits in play in infants and toddlers at risk for autism spectrum disorders. Wetherby et al. (2007) examined videotaped behavior samples of children with autism spectrum disorders, children with developmental delays, and typically developing children between the ages of 18 and 24 months and found that the ASD group differed significantly from the TD group in functional actions and pretend play actions directed towards another person or a doll. The ASD and DD groups, however, did not differ on these play variables. Landa et al. (2007) reported differences in the number of action schema sequences and action schemas directed towards others at 24 months between siblings of children with autism diagnosed with autism at 14 months of age, non-affected siblings, and typically developing controls, but found no differences in play variables between siblings of children with autism who had not been diagnosed until 30–36 months and any of the other diagnostic groups. Thus, recent evidence suggests that while children with autism show deficits in play early in childhood, play deficits at this age may appear only in cases where ASD can be identified by 14 months of age and may also appear in children with other developmental delays.

Although there is a growing body of research examining early play behaviors in autism, many of these studies have examined variables that only partially map onto the pre-existing categories of sensorimotor, functional and symbolic play. Wetherby et al. (2007) examined only the symbolic play actions directed toward another person or doll, whereas Landa et al. (2007) examined action schemas, which likely represent a combination of functional and symbolic play behaviors. Accordingly, there is a need for research on early play in children with autism using the same variables (functional and symbolic play) that have been used to document play deficits in studies of older children with autism in order to examine how deficits in functional and symbolic play develop and identify their origins in early childhood. The current study examined functional and symbolic play behaviors at 18 months of age in a sample of infant siblings of children with autism who later did or did not develop autism and a sample of typically developing controls.

Repetitive and stereotyped behaviors are one of the defining features of autism. However, there continues to be some disagreement over the age at which repetitive and stereotyped behaviors arise and the specificity of these behaviors to autism spectrum disorders. While some retrospective home video and parent report studies have found evidence for increased levels of repetitive behaviors in children with autism during the first and second year of life (Baranek 1999; Watson et al. 2007), others have not (Werner and Dawson 2005). In addition, many of the studies that support the presence of repetitive behaviors in late infancy and early childhood find similar patterns of

repetitive behaviors in children with other developmental delays. A similar pattern emerges with prospective studies of children later diagnosed with autism spectrum disorders and there is some evidence for increased levels of repetitive behaviors (Watt et al. 2008) and atypical object play (Ozonoff et al. 2008) in the first 2 years of life when compared with children with other developmental delays and those with typical development.

There are differences in how authors have defined repetitive behaviors and the contexts in which they have examined these behaviors. Some studies have included only those atypical motor mannerisms and postures frequently seen in older children with autism (such as hand flapping or head shaking; Loh et al. 2007). Other studies have defined repetitive behaviors more broadly and have included both typical and atypical behaviors (e.g. banging objects together as well as spinning objects; Ozonoff et al. 2008). Few, if any, studies have examined repetitive behaviors in the context of toy play. The current study examines repeated actions during a free play assessment and makes a clear distinction between different types of repetition. In the context of toy play, repeated actions may include repetitive behaviors (e.g. banging a block or a pot), repetitions of functional or symbolic actions with toys (e.g. brushing hair multiple times with a brush or an imaginary brush) and atypical motor behaviors or atypical actions with objects (e.g. hand flapping, postures or shaking/waving objects). Given that the repetition of play actions is observed in typical development, it may be that some repeated behaviors do not distinguish children who are later diagnosed with ASD from children with typical development while other behaviors do. Thus, it is necessary to catalogue what actions are being repeated and distinguish different types of repetitive actions. In particular, it may be important to distinguish purposeful repetitions of functional or symbolic play behaviors (functional repeated play) from purposeless repetitive actions that have the potential to become stereotyped, such as banging or mouthing objects (non-functional repeated play).

The first question of this study is whether the differences in symbolic, functional and repeated play between children later diagnosed with autism and typically developing children are present at 18 months of age. We expand upon previous research demonstrating early deficits in play by using established categories of play and clarifying exactly which aspects of play are impaired at 18 months of age, to ultimately connect early play impairments with later deficits in functional and symbolic play and, thereby, better understand how these impairments emerge. We hypothesized (1) that the ASD siblings would show fewer functional play behaviors than the TD controls; (2) that the ASD siblings would show significantly greater levels of repetitive actions with the potential to become stereotyped

(non-functional repeated play) than the TD controls; and (3) that the groups would not differ on repetitions of previously performed functional or symbolic play acts (functional repeated play).

The second question of the current study is whether the play of siblings of children with autism who do not meet criteria for an autism spectrum disorder is similar to that of children who are later diagnosed with ASD and different from typically developing controls. The current study will expand on previous research by exploring both those infant siblings who show later impairments in general cognition, language and social behavior as well as those who later appear indistinguishable from typically developing controls. Research suggests that close relatives of children with autism are prone to certain autistic characteristics, including deficits in social abilities such as affection, conversation, social play, as well as odd behavior (Bailey et al. 1998; Murphy et al. 2000), abnormalities in language development, and in the use of pragmatics or the inferred meaning of language (Fombonne et al. 1997; Landa et al. 1992). A small minority of relatives of children with autism show evidence of true obsessional and repetitive behaviors (Bolton et al. 1994). Such results lend credence to the concept of a broader autism phenotype in which autistic-like characteristics exist at sub-clinical levels in relatives of individuals with autism. By defining the sub-clinical impairments of “unaffected” siblings (i.e. siblings not displaying the full autism phenotype), especially those apparent early in development, researchers may begin to disentangle the syndrome or full phenotype from underlying inherited behavioral and neurological endophenotypes and thus, begin to clarify the pathway from genotype to autism. It may be that play is a mediator between a genetically determined fundamental insult and the development of language and social communication skills or it may be that play, language, and social communication are all effects of a shared impairment in symbolic representation or another basic deficit.

The question regarding the play behaviors of high-risk siblings who do not develop autism served an exploratory purpose and we did not specify any hypothesis about this group’s play behavior.

## Method

### Participants

Participants were selected from an ongoing study through the Center for Autism Research and Treatment at the University of California, Los Angeles in conjunction with the M.I.N.D. Institute at the University of California, Davis. The larger study recruited infant siblings of children

with autism and typically developing controls at 6, 12, or 18 months of age to participate in developmental assessments with the goal of identifying early predictors of autism. In Los Angeles, infant siblings of children with autism were recruited through the UCLA Autism Evaluation Clinic, through other ongoing studies at the Center for Autism Research and Treatment, and through organizations that provide services for children with autism and their families. Typically developing children were recruited through programs for infants and their mothers and through a mailing to families with an identified child in the appropriate age range. In Davis, participants were recruited through a database maintained by the M.I.N.D. Institute.

Inclusion criteria for infant siblings of children with autism were based in part on the eligibility of the older siblings (proband). Proband had a previous diagnosis of autistic disorder (not Aspergers Syndrome or Pervasive Developmental Disorder—NOS). In Los Angeles, confirmation of the probands' diagnoses was conducted at the UCLA Evaluation Clinic, based on the DSM-IV criteria (APA 2000), the Autism Diagnostic Observation Schedule (ADOS; Lord et al. 2000), and the Autism Diagnostic Interview-Revised (ADI-R; Lord et al. 1994). At UC Davis, diagnoses of probands were confirmed through a record review and, in cases where records were inconsistent, direct assessment using the ADOS. Exclusion criteria for the proband included medical conditions associated with autistic symptomatology such as Fragile X Syndrome or Tuberous Sclerosis. Both the proband and the infant sibling did not have severe visual, auditory, or motor impairments.

The typically developing control group consisted both of first-born children as well as younger siblings of typically developing children. Inclusion criteria for typically developing first-born children included no history of autism spectrum disorders among first-degree family members (and criteria 2 and 3 below). Inclusion criteria for infant siblings of typically developing children were also based in part on the eligibility of the older sibling (proband). In this case, the proband was typically developing. Inclusion criteria for the typically developing infant siblings included: (1) proband's gestational age of 36–42 weeks; (2) no abnormalities in pregnancy or neonatal period for either the proband or the infant sibling; (3) no chronic health conditions, past hospitalizations or significant injuries for either the proband or the infant sibling; and (4) no diagnosed developmental or learning disabilities, or behavioral disorders in the proband. The typically developing proband must also have scored in the normal range on the parent-completed Social Communication Scale (SCQ; Berument et al. 1999), to rule out autistic symptomatology.

Although play behaviors were examined at 18 months of age, participants were assessed at 24 and 36 months of

age as well. Classification of the groups was based on later outcomes at 36 months of age, except for one participant who was classified based on the assessment at 24 months of age. The play assessments were conducted at the child's 18-month birth date ( $\pm 2$  weeks).

The current study included 77 infants comprising four groups selected from the larger study: (1) infant siblings of children with autism who later met criteria for an autism spectrum disorder (ASD siblings,  $n = 17$ ); (2) infant siblings of children with autism who did not later meet criteria for an autism spectrum disorder, but showed other deficits in cognitive, linguistic and/or social skills (Other Delays siblings,  $n = 12$ ); (3) infant siblings of children with autism who did not later meet criteria for an autism spectrum disorder and did not show deficits in cognitive, linguistic and/or social skills (No Delays siblings,  $n = 29$ ); and (4) typically developing controls (TD controls,  $n = 19$ ). The ASD sibling group included all infants meeting the Group Selection criteria (below). Subjects in the Other Delays sibling, No Delays sibling, and TD control groups were selected randomly from the larger sample, except that the two sites were sampled equally.

#### Group Selection

The ASD sibling group ( $n = 17$ ) was comprised of infant siblings who met criteria for an autism spectrum disorder based on the Autism Diagnostic Observation Schedule (ADOS; Lord et al. 2000) at their outcome assessment (at 36 months of age) and at least one other time point (either 18 or 24 months of age). Infant siblings in this group also showed scores on the Social Communication Questionnaire (SCQ; Berument et al. 1999) consistent with a diagnosis of autism or ASD.

Children were classified as Other Delays siblings ( $n = 12$ ) if they did not meet criteria for ASD on the ADOS at any time point, but showed deficits in general cognition, language, or social behaviors at 36 months of age (with the exception of one child who had not yet been assessed at 36 months of age and was classified based on scores at the 24 month time point). Within this group, 2 of the children had general developmental delays (composite score below 78, one non-language subtest and one language subtest at least 1.5 standard deviations below average on the Mullen Scales of Early Learning [MSEL, Mullen 1995]), 1 had a language delay (at least 2 standard deviations below average on either or 1.5 standard deviations below average on both the receptive and expressive language subtests of the MSEL), 4 had only social deficits (elevated scores on the ADOS Social-Communication algorithm at 36 months, but did not meet criteria for either Autism or ASD on the ADOS or for a language delay or general developmental delay based on the MSEL) and 5

fell into the other concerns category (did not meet criteria for any of the other categories, but parents or examiner noted some concern about the child's development).

Children were classified as No Delays siblings if they were younger siblings of children with autism who did not meet criteria for an autism spectrum disorder based on the aforementioned criteria (including having never met criteria for autism or ASD on the ADOS), did not have deficits in general cognition, language, or social behaviors, did not have any scores on the MSEL more than 2 standard deviations below average and had no more than one score more than 1.5 standard deviations below average.

Children were classified as TD controls if they were not younger siblings of children with autism and they did not meet criteria for an autism spectrum disorder based on the aforementioned criteria, did not show impairments in general cognition, language, or social behaviors, did not have any scores on the MSEL more than 2 standard deviations below average, and had no more than one score more than 1.5 standard deviations below average.

Table 1 shows the demographic characteristics of the participants. Chi-Square ( $\chi^2$ ) analyses, with Fisher's exact test to correct for low expected frequencies, were used to examine group differences in mother's education, family income, and child gender. Group membership was not significantly related to mother's education or family income. There was a significant relationship between child gender and group membership, with a greater percentage of male participants in the ASD sibling group (82.4% male) as compared to the other groups (50.0, 44.8 and 36.8%, respectively). This difference is to be expected given that the ratio of males to females in autism is 4.3:1 (Fombonne 2003).

One-way analyses of variance (ANOVA) were used to examine group differences in the chronological age, verbal mental age, and non-verbal mental age of the groups based on participants' scores on the MSEL at 18 months of age. The groups differed significantly on both verbal and non-verbal mental age. Between group contrasts found that all of the groups differed from one another in their verbal mental ages, with the exception of the Other Delays and No Delays sibling groups. The ASD group differed from all of the other groups in their non-verbal mental ages; the other groups did not differ from one another.

## Measures

### *Autism Diagnostic Observation Schedule (ADOS; Lord et al. 2000)*

The ADOS was administered at 18, 24 and 36 months of age and used in classifying the children as having an autism spectrum disorder. The ADOS is a structured observational

assessment with modules designed for different levels of expressive language that measure the social and communication behaviors indicative of autism. The assessment provides opportunities for interaction and play and "presses" for certain target behaviors within these interactions. An algorithm is used with cut-offs for autism and autism spectrum disorders. The ADOS has high test-retest reliability as well as good internal consistency (Lord et al. 2000). However, because the ADOS only examines a 30-min sample of behavior, it cannot be used in isolation to diagnose autism spectrum disorders (Lord et al. 2000). As such, the Social Communication Questionnaire (SCQ) was also used to categorize the participants into groups.

### *Social Communication Questionnaire (SCQ; Berument et al. 1999)*

The SCQ is a 40-item parent-report questionnaire that addresses the child's social functioning and communication skills. It is based on the DSM-IV criteria for autism spectrum disorders and is highly correlated with the ADI-R (Berument et al. 1999). Using a cut-off score of 15, the SCQ has a sensitivity of 85% and a specificity of 67% (Berument et al. 1999). The SCQ was administered at 24 and 36 months of age as a measure of autism symptomatology.

### *Mullen Scales of Early Learning (MSEL; Mullen 1995)*

The Mullen is a standardized, normed developmental assessment of verbal and non-verbal IQ for children under 6 years of age that was administered at each time point. It provides an overall index score as well as verbal subscale scores (Receptive Language and Expressive Language) and non-verbal subscale scores (Visual Reception and Fine Motor). The Mullen has good test-retest reliability and high internal consistency (Mullen 1995).

## Procedure

During the 18-month visit, each child was administered a 4-min free-play assessment. The assessment involved the presentation of a standard set of toys the child had not yet seen during the visit and was administered in the middle of the full assessment protocol after the MSEL and before the ADOS. Toys included a play stove, pot with a lid, some sponges (3), a play sandwich (that came apart into the different components—2 slices of bread, 1 slice of cheese, tomatoes and cucumbers/pickles), a brush, a cup, a plate, a spoon, a fork, a square block, a cylindrical block and two Ernie dolls. During the administration of the assessment, the child was allowed to play with the toys with little intrusion from the test administrator or the child's

**Table 1** Demographic information by group

|                              | Autism spectrum siblings ( <i>n</i> = 17) | Other delays infant siblings ( <i>n</i> = 12) | No delays infant siblings ( <i>n</i> = 29) | Typically developing controls ( <i>n</i> = 19) | $\chi^2$ and <i>F</i> ( <i>df</i> ) |
|------------------------------|---|---|--|--|-------------------------------------|
| Age at testing (mo)          | 18  | 18  | 18   | 18   |                                     |
| Age at outcome grouping (mo) | 33.95 (4.69)                              | 33.75 (4.76)                                  | 34.54 (5.12)                               | 35.56 (2.72)                                   | <i>F</i> (3,73) = .55               |
| Verbal MA                    | 11.74 (2.69) <sup>a</sup>                 | 15.29 (3.65) <sup>b</sup>                     | 17.16 (3.86) <sup>b</sup>                  | 20.18 (3.59) <sup>c</sup>                      | <i>F</i> (3,73) = 17.96**           |
| Non-verbal MA                | 16.62 (1.64) <sup>a</sup>                 | 18.68 (2.21) <sup>b</sup>                     | 18.88 (2.13) <sup>b</sup>                  | 19.83 (1.91) <sup>b</sup>                      | <i>F</i> (3,70) = 8.12**            |
| Gender (% male)              | 82.4%                                     | 50.0%   | 44.8%                                      | 36.8%  | $\chi^2$ (3, <i>N</i> = 77) = 8.64* |
| Income                       |   |   |  |  |                                     |
| \$75,000–\$125,000 +         | 5 (29.4%) <sup>1</sup>                    | 5 (41.7%)                                     | 20 (69.0%)                                 | 11 (57.9%)                                     | $\chi^2$ (3, <i>N</i> = 75) = 6.05  |
| Mother’s education           |   |   |  |  |                                     |
| College degree +             | 11 (64.7%) <sup>2</sup>                   | 6 (50.0%) <sup>2</sup>                        | 24 (82.8%)                                 | 15 (78.9%) <sup>2</sup>                        | $\chi^2$ (3, <i>N</i> = 74) = 4.45  |

Group contrasts are indicated by the letters a and b where different letters mean significant differences between groups at the *p* < .05 level

<sup>1</sup> Two individuals in this group failed to report their income level. The percentage reported is out of 100% of the total *n*

<sup>2</sup> One individual in each of these groups failed to report the mother’s education level. The percentages are out of 100% of the total *n*

\* *p* < .05

\*\* *p* < .001

guardian. The assessment was videotaped and coded, beginning either when the experimenter finished placing all toys on the table or when the child first interacted with the toys (if the child began playing with the toys before they had all been placed on the table).

In the coding system, the children’s play behaviors were divided into functional, symbolic, and repeated play. The categories of functional and symbolic were defined according to the parameters set out by Sigman and Ungerer (1984). Functional play acts included four different subgroups of actions—object-directed (e.g. placing a cup on a plate), self-directed (e.g. brushing his/her hair), doll-directed (e.g. brushing the doll’s hair), and other-directed (e.g. putting a spoon to the experimenter’s mouth). For the purpose of this study, doll-directed and other-directed functional play were combined and labeled “other-directed” since both behaviors had low base rates. Symbolic play included three subgroups of actions: substitution play, or using one object as another (e.g. putting a plate on his/her head as a hat), imaginary play, or the attribution of pretend properties to actual objects/existence of pretend objects (e.g. making cooking sounds while cooking), and doll-as-agent play, or the use of a doll to perform independent actions (e.g. having the doll brush its own hair). The category of repeated play was created for this study and constituted all non-novel actions in which the child performed the same action on the same object multiple times. Repeated play was then divided into functional repeated acts and non-functional repeated acts. Functional repeated play was defined as non-novel actions that were repetitions of previously performed functional or symbolic play acts and thus, continued to

manifest the same functional or symbolic understanding of the object (e.g. putting a spoon to one’s mouth multiple times as this continues to illustrate a concrete or functional understanding of the use of a spoon). Non-functional repeated play, on the other hand, included non-novel actions that did not reflect a functional or representational understanding of the object when repeated (e.g. repeatedly putting objects into a pot and then taking them out as this does not illustrate the function of the pot as a cooking tool). Non-functional repeated play also included actions that were repetitive in nature and had the potential to become stereotyped (e.g. banging and chewing on toys). However, atypical or stereotyped behaviors (e.g. hand-flapping, twirling and toe-walking) were not included as non-functional repeated play as such actions often do not involve interaction with toys.

We did not include a specific category for sensorimotor play behaviors because the objects provided to the children in the play assessment were not appropriate for sensorimotor play (unlike balls, Silly Putty, etc.) and instead, pulled for more developmentally sophisticated behaviors. Given the functional nature of the toy set used in this study, children’s sensorimotor exploration was classified as non-functional repeated play. The rationale behind this was that when performed with the provided toys, sensorimotor actions such as mouthing or banging toys together are repetitive in nature, appear purposeless and have the potential to become stereotyped.

Reliability was calculated for a team of two coders blind to the group status of the participants and to the study hypotheses. Reliability was established separately for frequencies within each of the categories of play. Coders were

trained on a sample of typically developing children at ages 18 months, 24 months and 36 months. Intraclass correlation coefficients (ICCs) were used to evaluate interrater reliability. ICCs for the training sample for each of the play categories ranged from .80 to .97. The coding data generated by both coders was averaged for the analyses reported here. The purpose of averaging the codes was to account for the subjective quality of child behavior and to include both coders' estimates of behavior frequencies rather than choose one person's count over the other. ICCs for the two coders for the whole data set ranged from .84 to .95.

## Results

### Control Variables Associated with Play

Given the group differences in gender, we examined the relationship between gender and play behaviors. Girls showed significantly more total functional play ( $t(75) = -2.66, p = .01$ ), self-directed play ( $t(75) = -2.09, p = .04$ ), and other-directed play ( $t(75) = 3.19, p = .002$ ), and fewer non-functional repeated play behaviors ( $t(75) = 3.20, p = .002$ ) than boys. However, given the ratio of boys to girls in autism is 4:1, these results should be interpreted with caution. Such group differences in gender may represent an artifact of the grouping variable or vice versa.

Verbal mental age was positively related to object-directed ( $r = .25, p = .03$ ), self-directed ( $r = .26, p = .02$ ), other-directed ( $r = .27, p = .02$ ), functional ( $r = .41, p < .001$ ), and symbolic play ( $r = .26, p = .02$ ), and negatively related to non-functional repeated play ( $r = -.37, p < .001$ ). Non-verbal mental age was positively related to other-directed ( $r = .37, p = .001$ ), functional ( $r = .32, p = .006$ ), and symbolic play ( $r = .27, p = .02$ ). Non-verbal mental age was negatively related to non-functional repeated play ( $r = -.32, p = .005$ ). Given that children with autism show deficits in language and that many also have other cognitive deficits, the relationships between verbal mental age, non-verbal mental age, and play may represent artifacts of the grouping variables. In other words, the diagnostic groups differ in verbal and non-verbal mental age and, thus, the correlations may be the result of simultaneous group differences in all of the variables. However, it is also possible that the diagnostic groups are proxies for differences in verbal and non-verbal mental age.

Due to variability in children's engagement with the toy set, duration of play sessions coded for each child also

varied. None of the play variables was correlated with duration of the coded play session (all  $p$  values  $>.20$ ).

### Group Differences in Play Behaviors

Given that these are count data, a negative binomial regression was used to examine group differences in play at 18 months of age. The negative binomial distribution accounts for the positive skew of count data as well as the overdispersion (in which the variance exceeds the sample mean) seen in the data. An exposure (an adjustment to the model to account for different observation times) was used to capture the different lengths of time subjects played with the toys. We entered three dichotomous grouping variables to represent the ASD infant sibling, Other Delays infant sibling and No Delays infant sibling groups. We then compared each of the dummy coded sibling groups to the TD control group (included in the model as the reference group) on each of the play variables. The coefficients and significance test results for the negative binomial regressions are presented in Table 2. The coefficients shown represent the difference in each of the play variables for the three infant sibling groups compared to the TD control group.

For ease of presentation, Table 3 presents the mean rates per minute and standard deviations for each of the four groups for each of the play behaviors examined (functional, symbolic, functional repetitive and non-functional repetitive) as well as the total number of play acts. Functional play was divided into the sub-categories of object-directed, self-directed, and other-directed play. Table 3 superscripts also demonstrate the group differences in play from the negative binomial regression analyses (coefficients, standard errors and  $z$  tests from the negative binomial regression are presented in Table 2).

The first question of this study was how the ASD siblings would compare to the TD controls on functional, symbolic and repetitive play. The negative binomial regression results suggest that the ASD sibling group shows significantly fewer novel functional play behaviors than the TD control group, supporting the first hypothesis. The ASD sibling group performed an average rate of 1.28 (SD = 1.80) novel functional play actions per minute, while the TD control group performed an average rate of 1.42 (SD = .82) functional play actions per minute. With regard to the sub-categories of functional play (object-directed, self-directed and other-directed), the ASD sibling group showed significantly less self-directed ( $M = .27, SD = .25$ ) and other-directed functional play ( $M = .01, SD = .03$ ) than the TD control group ( $M = .49, SD = .38$ ) and ( $M = .18, SD = .34$ ), respectively).

**Table 2** Group differences in play behaviors—negative binomial regression

| Variable                               | <i>B</i> | SE of <i>B</i> | <i>z</i> |
|--|----------|----------------|----------|
| <b>Total functional play</b>           |          |                |          |
| ASD                                    | −.59     | .21            | −2.78*   |
| Other delays                           | −.25     | .22            | −1.12    |
| No delays                              | −.08     | .17            | −0.49    |
| Constant                               | 1.87     | .13            | 13.98**  |
| <b>Object-directed functional play</b> |          |                |          |
| ASD                                    | −.31     | .27            | −1.14    |
| Other delays                           | .02      | .29            | 0.09     |
| No delays                              | .04      | .23            | 0.17     |
| Constant                               | 1.22     | .18            | 6.74**   |
| <b>Self-directed functional play</b>   |          |                |          |
| ASD                                    | −.70     | .29            | −2.42*   |
| Other delays                           | −.79     | .34            | −2.32*   |
| No delays                              | −.35     | .23            | −1.55    |
| Constant                               | .79      | .16            | 4.80**   |
| <b>Other-directed functional play</b>  |          |                |          |
| ASD                                    | −3.26    | 1.49           | −2.19*   |
| Other delays                           | −.27     | .62            | −0.43    |
| No delays                              | −.02     | .48            | −0.04    |
| Constant                               | −.27     | .37            | −0.73    |
| <b>Symbolic play</b>                   |          |                |          |
| ASD                                    | −.24     | .66            | −0.37    |
| Other delays                           | .20      | .67            | 0.30     |
| No delays                              | .30      | .54            | 0.56     |
| Constant                               | −.81     | .43            | −1.88*** |
| <b>Functional repetitive play</b>      |          |                |          |
| ASD                                    | −.44     | .35            | −1.27    |
| Other delays                           | −.53     | .39            | −1.37    |
| No delays                              | −.47     | .31            | −1.53    |
| Constant                               | 2.02     | .23            | 8.66**   |
| <b>Non-functional repetitive play</b>  |          |                |          |
| ASD                                    | 1.23     | .37            | 3.37*    |
| Other delays                           | .88      | .41            | 2.18*    |
| No delays                              | 1.02     | .33            | 3.07*    |
| Constant                               | .81      | .27            | 3.00*    |

TD control group served as the reference group

\*  $p < .05$

\*\*  $p < .001$

\*\*\*  $p < .1$

There were no group differences in symbolic play. Rates of symbolic play were low with group means between .08 and .12 symbolic acts per minute, and none of the groups differed significantly from the TD control group.

The second hypothesis, that the ASD sibling group would show greater levels of non-functional repeated play than the TD control group was supported. On average, the ASD sibling group performed 2.30 (SD = 2.26)

non-functional repeated play acts per minute while the TD controls performed an average of .54 (SD = .98).

The third hypothesis of no group differences in functional repeated play was supported as none of the sibling groups differed significantly from the TD control group on functional repeated play. All of the groups showed between .77 and 1.64 functional repeated play acts per minute.

The second question of this study focused on the play of infant siblings of children with autism who are not later diagnosed with autism and addressed how these infant sibling groups would differ from typically developing controls. Results suggest that neither the Other Delays sibling group nor the No Delays sibling group differed significantly from the TD control group on novel functional play.<sup>1</sup> When categories of functional play were considered, the Other Delays sibling group showed significantly less self-directed functional play ( $M = .24$ ,  $SD = .17$ ) than the TD controls ( $M = .49$ ,  $SD = .38$ ), while the No Delays sibling group ( $M = .44$ ,  $SD = .55$ ) did not differ from the TD control group. In addition, both the Other Delays sibling ( $M = 1.23$ ,  $SD = .92$ ) and No Delays sibling groups ( $M = 2.11$ ,  $SD = 4.02$ ) showed significantly more non-functional repeated play acts per minute than the TD controls ( $M = .54$ ,  $SD = .98$ ).

In light of the significant correlations between play variables and verbal mental age reported earlier, the analyses were rerun adding verbal mental age at 18 months of age as a covariate. After covarying verbal mental age, most of the aforementioned effects were no longer significant. There continued to be trends for the ASD sibling group to show fewer other-directed novel functional play behaviors than the TD control group and for the Other Delays sibling group to show fewer self-directed novel functional play behaviors than the TD control group. The No Delays sibling group continued to show significantly more non-functional repeated play than the TD control group, even though parallel effects for the ASD and Other Delays sibling groups dropped out. This suggests that, while language and non-functional repeated play are strongly related in the ASD and Other Delays sibling groups, they are not in the No Delays group.

With regard to the group differences that were no longer significant after covarying for verbal mental age, it is possible either that verbal mental age accounts for all of the variance in play attributed to group membership or that there was insufficient power to detect an effect of group

<sup>1</sup> We compared each of the sibling groups (ASD, Other Delays and No Delays) to the TD control group in a negative binomial regression model for each play variable. Comparisons among the sibling groups were not performed to reduce the number of total analyses and because there were no a priori hypotheses as to how these sibling groups would differ.



**Table 3** Rates per minute of play behaviors by group

|                              | ASD sibling group<br>(mean, SD) ( <i>n</i> = 17) | Other delays sibling groups<br>(mean, SD) ( <i>n</i> = 12) | No delays sibling groups<br>(mean, SD) ( <i>n</i> = 29) | TD control group<br>(mean, SD) ( <i>n</i> = 19) |
|------------------------------|--|--|---|---|
| Functional play              | 1.28 (1.80) <sup>a</sup>                         | .91 (.64) <sup>b</sup>                                     | 1.58 (1.50) <sup>b</sup>                                | 1.42 (.82) <sup>b</sup>                         |
| Object directed              | 1.00 (1.84)                                      | .54 (.52)  | .86 (.77)   | .73 (.54)                                       |
| Self directed                | .27 (.25) <sup>a</sup>                           | .24 (.17) <sup>a</sup>                                     | .44 (.55) <sup>b</sup>                                  | .49 (.38) <sup>b</sup>                          |
| Other directed               | .01 (.03) <sup>a</sup>                           | .12 (.21) <sup>b</sup>                                     | .26 (.55) <sup>b</sup>                                  | .18 (.34) <sup>b</sup>                          |
| Symbolic play                | .08 (.21)  | .11 (.19)  | .12 (.22)   | .11 (.22)                                       |
| Functional repeated play     | 1.39 (1.56)                                      | .77 (1.05)   | 1.22 (1.81)   | 1.64 (1.44)                                     |
| Non-functional repeated play | 2.30 (2.26) <sup>a</sup>                         | 1.23 (.92) <sup>a</sup>                                    | 2.11 (4.02) <sup>a</sup>                                | .54 (.98) <sup>b</sup>                          |
| Total play acts              | 5.05 (3.65)                                      | 3.02 (1.99)  | 5.03 (6.75)   | 3.74 (2.74)                                     |

Group contrasts are indicated by the letters a and b where different letters mean significant differences between groups at the  $p < .05$  level. Each sibling group was only compared to the TD control group

membership above and beyond verbal and non-verbal mental age.<sup>2</sup>

## Discussion

We examined the play behaviors of three groups of 18-month-old siblings of older children with autism: children later diagnosed with an autism spectrum disorder (ASD siblings), children later diagnosed with other deficits (Other Delays siblings), and children with apparent typical development (No Delays siblings). We contrasted these groups with typically developing controls.

Our first question addressed differences in functional, symbolic and repetitive play between the ASD siblings and the TD controls. Consistent with our first hypothesis, the ASD sibling group performed fewer novel functional play acts than the TD control group. This finding suggests that the deficits in functional play observed in older children with autism (Sigman and Ungerer 1981; Williams et al. 2001) are observable by 18 months of age and thus, is consistent with previous research findings that play impairments are evident early in development (Landa et al. 2007; Wetherby et al. 2007).

Examination of the subtypes of functional play revealed that the ASD sibling group showed fewer self-directed and other-directed play behaviors than the TD controls. However, the ASD sibling group did not show fewer object-directed functional play acts. This finding is of particular interest because it suggests that children with ASD may not understand people as potential recipients of a play action

and/or are not motivated to direct play behaviors to people (self or other) even before many of them are diagnosed.

Symbolic play did not differ among the groups due to floor effects, with few participants in any of the groups displaying symbolic play behaviors. Accordingly, results from the current study contrast with findings by Wetherby et al. (2007) of differences in pretend or symbolic play directed towards another person or doll. Wetherby and colleagues observed play when participants were between 18 and 27 months of age, while the present study examined children at only 18 months of age. It is likely that the lack of group differences in symbolic play in the present study are due to the younger age of the sample, following previous findings that children with a verbal mental age lower than 20 months do not yet consistently engage in symbolic play (Wing et al. 1977). Taken together, these results suggest that deficits in functional play appear prior to deficits in symbolic play, in a trajectory similar to that observed in typical development (Casby 2003).

In support of our second hypothesis, the ASD sibling group also showed significantly more non-functional repeated play than the TD controls. Given that the two groups do not differ in the total number of play acts performed, it appears that the ASD sibling group is engaging in non-functional repeated play at the expense of performing novel actions. This finding supports previous research suggesting that repetitive and stereotyped behaviors and/or their precursors are observable in the second year of life among children subsequently diagnosed with autism. The increased frequency of these behaviors suggests that by 18 months of age, children who are later diagnosed with autism are already interacting with and exploring their environment in a way that is atypical. Moreover, because these repeated behaviors are performed at the expense of novel actions, children with autism may fail to receive the benefits of fully exploring their environment and thus, negatively impact their cognitive and language development.

<sup>2</sup> Non-verbal mental age was also added to the negative binomial regression models, but was dropped because it did not significantly predict any of the play behaviors once verbal mental age and group had been accounted for. When only non-verbal mental age was added to the negative regression models, it did not significantly predict any of the play behaviors and findings of group differences persisted.

In line with our third hypothesis, the groups did not differ significantly in functional repeated play. This suggests that all of the groups, regardless of their later developmental status, engage in some form of repetition. Thus, as suggested by the literature on typically developing children, repetition of actions is not abnormal in and of itself. Rather, it is the content or what is being repeated that is predictive of atypical development. Moreover, typically developing children engage in both repetitive and novel actions, whereas children who are later diagnosed with autism spectrum disorders do not.

Our second question addressed the play of children at risk for autism spectrum disorders and this study examined two groups of children with a known genetic risk for autism who do not develop the disorder—one with later deficits in cognition, language and social behavior (Other Delays siblings) and one with no observable deviations from typical development (No Delays siblings). Neither group differed from TD controls in their total number of play acts or in the number of novel functional or symbolic play acts they performed (with the latter likely due to floor effects). Within the category of novel functional play, the Other Delays sibling group showed deficits in self-directed play compared to the TD control group. This echoes the aforementioned finding of decreased self-directed play in the ASD sibling group and may reflect a similar difficulty in understanding social partners and/or sharing attention that may impact later social development.

Interestingly, both the Other Delays and No Delays sibling groups performed significantly more non-functional repeated play acts than the TD controls. This result is consistent with findings by Bailey et al. (1998) that obsessional and repetitive behaviors may be observed in relatives of children with autism and suggests that these behaviors may arise early in development. More importantly, these results suggest that children at-risk for autism spectrum disorders may display atypical behaviors at 18 months of age even if they do not appear delayed later on. Both the Other Delays and No Delays siblings had lower verbal mental ages than the TD controls at 18 months. Especially in the case of the No Delays siblings (who show no language or other delays compared to TD controls at 36 months of age), these findings document atypical development in siblings at risk for autism.

Notably, after covarying the effects of verbal ability, many of the aforementioned group differences in play behaviors were no longer significant. This raises the question of whether, given that language is one of the core deficits in autism, it is appropriate to use language as a covariate. To the extent that deficits in language in children with ASD may act as a proxy for the disorder itself, by covarying verbal mental age, we may be removing the variance in early development we are trying to explain. In

light of the relationships between play and language in typically developing children and children with autism (Lyytinen et al. 1999; Mundy et al. 1987), and the possibility that in ASD, deficits in play and language may represent behavioral manifestations of an underlying deficit such as symbolism (Lewis 2003), it may be inappropriate to control for one when looking at the impact of the other. This seems especially true given that language skills are not fundamentally necessary to engage in play and particularly in non-symbolic forms of play. Thus, it is still important to characterize deficits in play even if their predictive ability may be ultimately overshadowed by language skills. For that reason, we have presented group differences in play without covarying verbal mental age and have discussed the significance of these results.

Nevertheless, when we did covary language, there continued to be trends for the ASD sibling group to show fewer other-directed novel functional play behaviors than the TD control group and for the Other Delays sibling group to show fewer self-directed novel functional play behaviors than the TD control group.

One limitation of this study is that the 4-min play assessment was relatively short, such that measures of observed play may have underestimated participants' true abilities. While time spent playing did not differ across the groups, the length of the assessment may differentially impact scores such that children who are slow-to-warm-up do not have adequate time to acclimate to the task/situation and demonstrate their true abilities. To verify the accuracy of these results, future investigators might observe play for a longer period of time and thus, obtain a more representative sampling of participants' play.

Another possible limitation of this study was the classification of the participants into the different diagnostic groups. Almost all participants were classified based on assessments through 36 months of age, with one having data only through 24 months of age. However, it is possible that participants from the non-ASD groups (Other Delays siblings, No Delays siblings and TD controls) may develop ASD after 36 months of age. However, it is unlikely that a significant percentage will later develop autism given the prevalence of autism spectrum disorders in infant sibling populations is still only 2–6% (Newschaffer et al. 2003).

Overall, we replicated and expanded upon prior research on deficits in play by examining children at risk for autism spectrum disorders prior to the age at which clinicians typically confer diagnoses. As such, findings of group differences represent potential predictors of developmental and diagnostic outcomes rather than characteristics of an already diagnosed condition. Our results suggest that deficits in functional play are evident by 18 months of age. Our results suggest that few children, even those that are typically developing, display symbolic play behaviors at

18 months of age. However, it may be that our assessment failed to capture the children's symbolic play, which may occur with more frequency when interacting with caregivers in a naturalistic setting. Likewise, our results suggest that children who are later diagnosed with autism show repeated behaviors during play that have the potential of becoming stereotyped. These behaviors appear to prevent children from fully exploring their environment and this may impact later development by acting as a mechanism through which later deficits in functioning appear. Moreover, we contributed to the literature on the broader autism phenotype by showing that deficits in play similar to those seen in children who are subsequently diagnosed with autism are also present in high-risk children who do not develop this disorder and that these deficits in play occur regardless of whether or not the children experience delays subsequently.

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