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Pretend Play and Social Engagement in Toddlers at High and Low Genetic Risk for Autism Spectrum Disorder

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Abstract

Toddlers with an older sibling with autism spectrum disorder (ASD) and low risk (LR) toddlers with typically-developing older siblings were observed during free play with a parent and elicited pretend with an examiner at 22-months. Functional and pretend play, children's social engagement, and parent sensitivity were assessed during free play. Complexity of play was assessed during the elicited pretend task. Toddlers with an ASD diagnosis showed less pretend play across contexts and less social engagement with parents or the examiner than either LR toddlers or high risk toddlers without a diagnosis (HR-noASD). Lower levels of pretend play and social engagement were associated with symptom severity within the high risk group, reflecting emerging ASD in toddlerhood.

Keywords

High-risk siblings; Functional play; Pretend play; Social engagement; Parent sensitivity; Autism spectrum disorder

Introduction

Typically-developing toddlers spend much of their time engaged in exploratory play with toys and other objects as a way of practicing routines and learning about the world (Lillard 2007; McCune-Nicolich 1981; Piaget 1962). By the middle of the second year, play becomes increasingly complex as toddlers move from reliance on sensorimotor exploration and functional activities with toys to also engage in pretend play, reflecting the ability to imbue objects with imaginary characteristics and functions. In contrast, children with autism

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Conflict of interest The authors have no conflicts of interest.

Ethical approval Informed Consent was obtained from all parents who participated in this study.

spectrum disorder (ASD) show deficits in pretend play in early childhood and beyond (Charman et al. 1997; Hobson et al. 2013; Rutherford et al. 2007), but it is unclear how early these deficits can be assessed reliably. In a recent large-scale prospective screening study of a community sample, delays or deficits in the emergence of pretend play at 24 months were identified as a strong predictor of ASD (Barbaro and Dissanayake 2013). Indeed, these authors note that "although a deficit in pretend play was not a useful marker of AD/ASD at 18 months, it became one of the most important predictors for a diagnosis of AD/ASD at 24 months" (p. 78).

As part of a larger study of social development in toddlers at genetic risk for ASD, we examine the play behavior of 22-month-old toddlers with [high risk (HR)] and without [low risk (LR)] an older sibling with ASD in two different contexts: free play with a parent and elicited pretend play with an examiner. Because pretend play, especially when it is a newly emerging skill, is highly scaffolded by parents (Lillard 2007), we examine parental support and sensitivity as well as child engagement with parents during play. Children's interest in social interaction with a play partner may be particularly important for emerging pretend play in HR toddlers (Hobson et al. 2013), who are not only at increased risk for ASD but also subthreshold social communicative impairments that may be characteristic of the broader autism phenotype (Ozonoff et al. 2014). This is one of the few studies of which we are aware to assess pretend play in HR and LR toddlers just prior to their second birthday when simple pretend play is consolidating in typically-developing toddlers (Belsky and Most 1981), and to compare LR toddlers to HR toddlers with and without a diagnosis of ASD.

Play in Typical Development

Numerous studies have characterized the play behavior of typically-developing children, emphasizing the functions of play (Lillard 2007; McCune-Nicolich 1981; Piaget 1962) and the increasingly sophisticated sequence of activities that characterizes spontaneous play from infancy to toddlerhood (Belsky and Most 1981; Lillard 2007; McCune-Nicolich 1981). Play in early infancy involves sensorimotor exploration, primarily mouthing and simple manipulation (Belsky and Most 1981; Piaget 1962), as infants learn about the properties of objects. By the end of the first year and beginning of the second, play becomes more elaborated and functional, as toddlers use toys in routine ways consistent with their intended uses, for example, stacking blocks or rolling a toy car along the floor. In the second year, pretend play also emerges, primarily at a basic level with acts directed to the self, a doll, or a social partner, such as pretending to feed oneself or a doll in the absence of real food (Lillard 2007). McCune-Nicolich (1981) has argued that early pretend play emerges as toddlers use toy objects to imitate everyday actions and events consistent with their own experiences. More complex symbolic play builds on these basic pretend skills, and language also becomes more symbolic, thereby supporting more complicated pretend play scenarios.

In Belsky and Most's (1981) cross-sectional study of 7–21 month-olds observed during independent play with a standard set of toys, simple pretend play was evident early in the second year and peaked at 21 months. Although both simple manipulation and functional play were observed at each age, simple manipulation declined sharply by 21 months. Moreover, the levels of play showed a hierarchical structure with simple manipulation

emerging prior to functional play which in turn predated the emergence of simple pretend. Thus, 22 months appears to be an optimal time to examine the emergence of simple pretend play in toddlers with and without a genetic risk for ASD.

Although children's spontaneous play provides clues to their level of cognitive functioning, more sophisticated play is often evident when young children have a social partner to scaffold their play either by following the child's lead or suggesting play activities (Lillard 2007). Thus, pretend play often involves imitation, either immediate or deferred, as children engage in routine action sequences with familiar toy objects. In addition, observations of parent–child interaction during play provide a window into their levels of reciprocity and social engagement, which in turn may be associated with the quality of play in a bidirectional feedback loop. Lillard discusses the importance of parental scaffolding, joint attention, and social referencing in facilitating more advanced pretend play sequences in toddlers. In the current study we assess children's levels of play during free play with a parent, while also obtaining ratings of child engagement and parental warmth, sensitivity and scaffolding. In addition, we examine toddlers' ability to imitate increasingly sophisticated play sequences modeled by an examiner to assess more differentiated levels of pretend play.

Play and ASD

It is well-documented that children with a diagnosis of ASD are less likely to engage in either spontaneous or scaffolded pretend play (Charman et al. 1997; Sigman and Ungerer 1984) than control children. Indeed delays and/or deficits in pretending are a defining characteristic of ASD and are closely linked to cognitive and social deficits (Hobson et al. 2013; Manning and Wainright 2010; Rutherford et al. 2007), including delays in language development and imitation (Charman et al. 1997; Sigman and Ungerer 1984). In their classic study, Sigman and Ungerer compared the play behavior of preschool and school-age children with autism to matched groups of developmentally-delayed children and mental-age matched typically-developing toddlers during both unstructured solitary play and structured play with an examiner. Their definition of functional play included object-directed, selfdirected, and doll-directed acts, thereby categorizing Belsky and Most's more basic levels of pretend as functional. Children with autism spent less time engaged in any of these play acts than children in the other two groups during both the structured and unstructured conditions. Symbolic play, defined as object substitution or the use of the doll as an independent agent, albeit rare, was also less frequent in the children with autism. Children, regardless of diagnosis, engaged in more functional and symbolic play during the structured than unstructured condition. However, in the structured condition, children with autism did not differ from the other two groups in the frequency of doll-directed play. This indicates that some of the children with autism were capable of acts such as feeding the doll when prompted, although they were unlikely to do this spontaneously.

In a more recent study, Rutherford et al. (2007) also compared preschoolers with autism, preschoolers with developmental delay, and younger typically-developing toddlers matched for non-verbal mental age on both spontaneous and structured play tasks. Consistent with prior studies (Charman et al. 1997; Sigman and Ungerer 1984), children with autism

engaged in fewer symbolic acts in either condition than children in the two comparison groups, but children with autism were more likely to engage in some symbolic acts when the play was scaffolded by direct suggestions or modeling. In addition, within the group of children with autism, those who engaged in joint attention were also more likely to show spontaneous symbolic play, implicating social engagement in the emergence of pretend play. This is consistent with Hobson et al.'s (2013) contention that an underlying social impairment reflecting lack of motivation to engage with social partners in shared activities limits opportunities for pretending and may partly explain the lower level of pretend play and imitation observed in children with ASD.

Although a number of recent studies have focused on younger siblings of children with an autism diagnosis, only two studies have specifically examined pretend play in toddler-aged high risk siblings. Landa et al. (2007) observed less play on a structured assessment at 24 months in high risk siblings who received a diagnosis by 14 months, suggesting quite severe impairment. High risk siblings diagnosed at 36 months and siblings with social concerns but no diagnosis did not differ from low risk children in their play behavior. Christensen et al. (2010) observed free play in high risk and low risk toddlers at 18 months. The high risk toddlers who received a diagnosis of ASD by 36 months engaged in less self- and otherdirected play than low risk controls, but not less functional play directed to objects. Group differences in play were no longer significant with verbal mental age controlled. Christensen et al. speculate that the lack of self-directed and other-directed play acts may reflect a lack of social awareness or motivation to engage with play partners. Given the limited data on the play behavior of high risk siblings in toddlerhood, it is unclear whether deficits or delays in pretending are evident only in toddlers who receive a diagnosis of ASD or are also apparent in HR siblings without a diagnosis, some of whom may be showing signs of the broader autism phenotype.

The Current Study

In the current study we examine the play of HR and LR toddlers during free play with a parent and during structured play with an examiner. Toddlers were studied at 22 months and assessed for ASD in the third year of life. LR toddlers were compared to HR toddlers with and without a diagnosis of ASD. In addition to levels of play, we rated the child's engagement with the parent and parental scaffolding and warmth during play, as the quality of the interaction appears to be an important aspect of young children's play behavior. Thus, the current study adds to the literature on early signs of autism in HR siblings by assessing delays or deficits in pretend play in both unstructured and structured contexts that are distinct from standard diagnostic assessments, while also rating social engagement with a play partner. Although limited pretend play is one diagnostic criterion for ASD, standard measures of pretending are only modestly correlated in preschool children with a diagnosis of ASD (Pierucci et al. 2015). Therefore, this study of HR siblings at 22 months provides additional insight into characteristics of both play behavior and social interaction during play, associated with emerging ASD in toddlerhood.

We predicted that children with a diagnosis of ASD would show less pretend when playing with a parent and also demonstrate a lower level of pretend during the elicited play scenarios

with the examiner than LR toddlers. We also expected the toddlers with a diagnosis to be less enthusiastic and interested in sharing play with their parent, although we had no predictions about differences in parent behavior. We also expected the HR toddlers without a diagnosis to fall between the toddlers with ASD and the LR toddlers, but given the limited data on this question, it was unclear whether they would differ from either or both of these groups. Finally, we expected that scores on the two play assessments would predict the severity of autism symptoms within the high risk group.

Method

Participants

Toddlers and their parents are participants in a larger prospective study of children at risk for ASD. The sample includes toddlers with an older sibling with ASD and comparison toddlers with a typically-developing older sibling. The 145 children included in this report were seen at 22 months (M = 22.75, SD = .70) for a play-based assessment of social behavior that included the procedures described below. Most children were seen again at 36 months for a follow-up diagnostic assessment (details below). Groups did not differ in age at the 22-month assessment (see Table 1). Parents signed informed consents prior to participation; the research protocol was approved by the University Institutional Review Board.

Most HR toddlers (n = 63, 40 males) were recruited between 6 and 16 months for a study of cognitive and social development, through the Autism Center at the University of Pittsburgh, with the exception of four HR children who joined the study at 22 months. To be eligible for inclusion in the HR group, children had to be born full-term after an uncomplicated pregnancy and delivery, and have an older sibling diagnosed with autism spectrum disorder according to research criteria. The older sibling's diagnosis was confirmed by research reliable staff at the Autism Center who administered the autism diagnostic observation schedule (ADOS; Lord et al. 2000) under the supervision of a licensed psychologist, prior to the younger sibling's enrollment in the study. Infants whose older siblings had known genetic or other anomalies, such as Fragile X, were excluded. Four HR toddlers were half-siblings of the child with ASD.

Low risk (LR) control participants (82, 46 males) were recruited from the local obstetrics hospital, community groups, pediatric offices, and word of mouth. Full-term healthy toddlers with a typically-developing older sibling and negative family history of ASD in first and second degree relatives comprised the LR group. Parents of LR children completed the social communication questionnaire (Rutter et al. 2003) on the older sibling prior to study enrollment; all scored well below the ASD cut-off of 15.

Participant characteristics are summarized in Table 1, including comparisons between the LR toddlers, the HR toddlers without a diagnosis (HR-noASD), and the toddlers with an ASD diagnosis (see below). Participating children are predominantly Caucasian and non-Hispanic (85.5 %). Parent education was scored according to the Hollingshead Scale. Although the majority of parents in both groups had at least a college degree (72.5 %), the LR parents as a group were more highly educated than the parents of the HR toddlers. This

sample overlaps with, but is not identical to those included in two recent publications (Campbell et al. 2015a, b).

Procedure

Toddlers' Play with a Parent

As part of a longer assessment of social development in toddlerhood, children were observed during free play with a parent using a standard set of age-appropriate toys selected to elicit functional and pretend play. These included a dump truck filled with blocks, a school bus with small removable figures, a series of colored shapes and pegs, a farm with animals, a kitchen set, a xylophone, a teddy bear, and a toy airplane with moving parts. Children were given 5 min to explore the toys on their own and then parents were asked to play with their child as they normally would. Because children varied widely in how willing they were to engage with the toys on their own, we focused our coding on the 10-min of children's play with their parent. Moreover, because we were interested in pretend play, given the age of the children, it was more likely that we would observe instances of pretend during play with a parent (Lillard 2007). Observations with the parent also allowed us to assess social engagement with a play partner and parent support for the child's play.

Children's Levels of Play—We used ELAN for Windows, a computer-based time-locked system, to code children's levels of play (Lausberg and Sloetjes 2009). The play period was divided into 30-s bouts, during which the highest level of play was coded. This resulted in 20 bouts across the 10-min observation. Three levels of play were coded, based on Belsky and Most (1981). These included: (1) simple manipulation or exploration (e.g., mouthing, banging, throwing, fingering, shaking, visually inspecting) at the lowest level; (2) functional play (the use of objects in an appropriate manner such as pushing the school bus, stacking blocks or putting them into the dump truck, and playing the xylophone); and (3) pretend play, at the highest level. To be coded as pretend play, the child had to attribute pretend properties to the toys by acting out a sequence or play scenario (for example, pretending to cook by stirring and "tasting"; feeding themselves or their parent while playing with the kitchen set; moving the school bus or other vehicles while making sounds suggesting driving or flying). Although we also included categories reflecting limited engagement in play with the toys, these occurred rarely and are not considered further. Scores were mutually exclusive and hierarchical such that only one play type (the highest level reached) was scored for each 30-s bout. The number of play bouts (out of 20 possible) during which children exhibited each play type as the highest level of play was entered into analyses.

We also rated child engagement with the parent during play on a 4-point scale adapted from the NICHD Study of Early Child Care [NICHD Early Child Care Research Network (ECCRN) 1999]. The first and second 5 min of play were rated separately and the two ratings were averaged. Higher ratings indicated that the child brought the parent into his/her play and/or responded positively to parent suggestions or initiations. Lower ratings indicated that the child ignored the parent, actively rejected the parent by turning away or moving away, or showed a clear preference for solitary play that excluded the parent. Free play data for one LR child are missing because of equipment problems.

Inter-rater reliability was determined by having two independent coders, blind to group assignment and diagnostic status, code 35 % of the DVD's. Intraclass correlation coefficients (ICC) were as follows: simple manipulation (.907), functional play (.938), pretend play (.939), and child engagement with parent (.898).

Parent Behavior during Play—Coders, different from those who scored the child's play, rated parent behavior using 1–4 point rating scales also developed by the NICHD ECCRN (1999). These included ratings of "warmth" (affectionate behavior and enjoyment of the interaction); "sensitivity" (following the child's lead and tuned into the child's interests); "intrusiveness" (the degree to which parents directed or controlled the interaction, following their own agenda rather than the child's lead); and "stimulates cognitive development" (teaching and scaffolding play). ICCs, calculated on 35 % of the DVD's were satisfactory: warmth (.854), sensitivity (.829), intrusiveness (.855), and stimulates cognitive development (.814).

Following the NICHD Study and because ratings of warmth and sensitivity were highly correlated with each other [r(144) = .725] and were negatively correlated with intrusiveness [r(144) = -.344, -.425], we derived a sensitivity composite score by adding the averaged warmth and sensitivity ratings and subtracting the averaged rating of intrusiveness. Stimulation of cognitive development was considered separately because it dealt specifically with scaffolding play.

Elicited Pretend Play

We also obtained a measure of children's ability to imitate structured, examineradministered pretend scenarios with a doll and toy props (Brownell and Carriger 1990). Children were seated at a small table with the examiner who modeled four scenarios at four levels of complexity. Examiners were trained to administer this task prior to data collection and they followed a standard script for each story. They modeled each story twice and then asked the child to repeat the story. At level 1, the self is the recipient of the actions (e.g., the examiner pretends to feed herself with a spoon). At level 2, the self is the agent and a doll is the recipient (e.g., the examiner pretends to feed the doll with a spoon). At level 3, the doll is both the agent and the recipient (the examiner has the doll feeding herself). At level 4, the doll is the agent and a toy teddy bear is the recipient (the examiner has the doll feeding the teddy bear). Boy and girl dolls were used to match the child's gender. Each story (making breakfast; getting ready for bed; making tea; getting ready for school) included three actions in fixed order, but the order of presentation of each scenario and the level of difficulty were counterbalanced, with the exception that the easiest level (self as recipient) was always presented first.

Scoring was done from DVDs by a new set of coders blind to group membership. For each pretend scenario the number of different actions imitated was scored (0–3), regardless of the sequence. In addition, the level of complexity was scored. For example, if the examiner modeled a level 3 scenario (the doll is making breakfast by pouring cereal, stirring it, and feeding herself), but the child imitated these actions by acting on the doll (i.e., the child feeds the doll), she would be credited with a level 2 response. If the child acted on himself,

he would be credited with a level 1 response. For the purposes of data analysis we calculated a weighted composite score that included the number of actions imitated at each level. In the example above, the child who imitated three actions at level 2 would receive a score of 6 for that scenario. Inter-observer agreement, calculated on 32 % of the DVD's, was high: total score (ICC = .962); highest level reached (ICC = .922). We also noted the proportion of children who reached level 2 and the proportion who reached either level 3 or 4, as few children reached these two higher levels. If children attended to the demonstrations, but did not attempt the tasks, they received a score of 0.

In addition to coding the child's performance on this task, we rated children's interest (engagement, attention, enthusiasm vs. disengaged, inattentive, bored), compliance (cooperative, able to take turns vs. grabbing toys, throwing toys, not awaiting turn), and activity level (able to sit at the table or on floor, even if moving around between stories vs. running around the room, frequently on the move) on 4-point scales, as children's interest and cooperation might vary widely in 22-month-olds and especially in toddlers with emerging autism. All three ratings were scored so that higher scores reflected better attention and compliance, and less inappropriate activity. Inter-rater agreement was high: ICC's = . 882 for interest, .829 for compliance, and .838 for activity level. Because these ratings were quite highly correlated (r's from .584 to .751), they were composited into an overall engagement score (range 3–12).

Developmental and Autism Assessments

Mullen Scales of Early Learning—The Mullen scales of early learning (MSEL; Mullen 1995), a standardized measure of cognitive development, was administered at 24 months. An early learning composite (ELC) score was derived from performance on the Receptive Language and Expressive Language scales and on the Visual Reception and Fine Motor scales. We substituted the 16 month Mullen score for eight children who were missing one or more subtest scores at 24 months or who missed the 24 month visit, but had been seen earlier (4 LR, 2 HR-noASD, 2 ASD). The correlation between the 16 and 24 month Mullen scores, available for 75 children, was r = .64, p < .01). Another seven children are missing Mullen scores at both 16 and 24 months (2 LR, 4 HR-noASD, 1 ASD) because of missed appointments, scheduling difficulties, or the child's refusal.

Evaluations for Autism—Both HR and LR toddlers were evaluated for an ASD diagnosis at follow-up, using the autism diagnostic observation schedule (ADOS; Lord et al. 2000). The ADOS, a semi-structured observational assessment, includes play-based activities that are meant to elicit reciprocal social interaction, communication, and stereotyped behaviors and provides scoring rules for a diagnosis of ASD. All children received either Module 1 or 2. Evaluations were conducted by a research-reliable tester from the Autism Center under the supervision of a licensed clinical psychologist with extensive experience assessing children with ASD; different examiners conducted the diagnostic assessments and the play visits. Diagnostic decisions were based on the 36 month assessment for most of the HR children (n = 41); two HR children who missed their 36 month visits were seen at 48 months; 18 HR toddlers were assessed at 24 months. One HR child with a diagnosis was assessed at a local developmental clinic specializing in autism,

but the ADOS score was not available. Although 36 month outcome data are preferred, data suggest that when diagnoses are clear at 24 months, they are likely to be stable (Chawarksa et al. 2009). Most LR children (n = 62) were assessed at 36 months, 18 were assessed at 24 months, and one was seen at 48 months.

Children were classified as ASD if they met cut-off scores on the ADOS (Lord et al. 2000) and also met DSM-IV criteria for an autism spectrum disorder (American Psychiatric Association 2000), as determined by interview and observation. Parents of children with elevated ADOS scores and/or serious clinical concerns were interviewed using the autism diagnostic interview-revised (ADI-R; Lord et al. 1994) to provide further information. Final diagnoses were made by a licensed clinical psychologist, based on a combination of structured diagnostic measures, DSM-IV criteria, and clinical judgment; 14 HR toddlers (12 boys) received a diagnosis of ASD. One LR boy with an earlier language delay and some social concerns received a diagnosis of ASD at 48 months and is included with the ASD group.

In addition to providing a diagnosis, ADOS scores were converted to severity scores using the algorithm provided by Gotham et al. (2009) to allow for comparability across age; severity scores are summarized in Table 1. As expected, there was a main effect of group status, F(2, 139) = 87.88, p < .001, partial $\eta^2 = .56$. The children with a diagnosis of ASD had substantially higher severity scores than the LR children (p < .001, d = 3.13) and the HR-noASD children (p < .001, d = 2.96). LR and HR-noASD children did not differ from one another.

Data Analysis Plan

Group differences were analyzed with ANOVA's and followed up with Games-Howell tests, given the unequal sample sizes across the three groups (LR, HR-noASD, ASD). Effect sizes are reported using partial eta squared (η^2) for main effects and Cohen's *d* for paired comparisons. Some analyses were also conducted controlling for the Mullen ELC or other covariates as described below. In addition, the proportion of children in each group who engaged in any pretend during free play and who reached a higher level during elicited pretend were compared with χ^2 . Correlations between behavior ratings and pretend play scores were examined separately by group. Finally, correlations were calculated between ADOS severity scores and pretend play measures within the HR group as a whole, including the one LR child with an ASD diagnosis.

Results

Preliminary analyses were conducted to test for gender differences during free play and elicited play. Only the children with no diagnosis were included in this analysis, as gender and ASD were confounded (13 of 15 children with a diagnosis of ASD were boys). There was trend for girls to receive higher ratings than boys on child engagement with parent during free play, t(127) = 1.93, p = .056. Because girls and boys did not differ on other play measures, child gender is not considered further. Correlations between parent education and play measures were not significant, so parent education is not controlled in the analyses. In addition, 14 children were observed with fathers (7 LR, 5 HR, 2 ASD), so we examined

whether children's free play behavior differed as a function of which parent served as the play partner. Children engaged in significantly less pretend play with fathers than with mothers, t(142) = 3.49, p = .001, so parent is included as a covariate in the analysis of pretend play. No other child variables differed by parent play partner. Similarly, mothers and fathers did not differ on the sensitivity composite score or the rating of "stimulates cognitive development."

There were significant group differences on the Mullen Early Learning Composite, F(2, 135) = 21.99, p < .001, partial $\eta^2 = .25$ (see Table 1). Children with ASD had significantly lower scores than the LR (p < .001, d = -1.73) and HR-noASD (p = .001, d = -1.39) toddlers. The HR-noASD children also had lower scores than the LR children (p < .02, d = -.50).

Group Differences in Play

Free Play—Children did not differ by group in the number of play bouts during which they engaged in either simple manipulation, F(2, 141) = 2.36, p = .098, or functional play, F(2, 141) < 1. However, group differences were evident in pretend play, with play partner (mother or father) controlled, F(2, 140) = 3.30, p = .04, partial $\eta^2 = .045$. Post hoc contrasts indicated that the toddlers with ASD were significantly lower in pretend play than either the LR (p = .014, d = -.86) or the HR-noASD toddlers (p = .019, d = -.94), who did not differ from each other. The group difference in pretend play was no longer significant once the Mullen ELC was controlled, F(2, 133) = 1.43.

Ratings of child engagement with parent also varied by group, F(2, 141) = 15.44, p < .001, partial $\eta^2 = .18$. The children with an ASD diagnosis were significantly less engaged with their parent during play than either the LR (p < .001, d = -1.61) or HR-noASD (p < .001, d = -1.41) toddlers. Although this difference was somewhat attenuated with the Early Learning Composite covaried, group differences remained significantly lower on engagement than either the LR (p = .003) or the HR-noASD (p = .001) children. HR-noASD toddlers and LR toddlers did not differ from one another on child engagement with parent in either analysis. Descriptive statistics are summarized in Table 2.

Finally, we examined the proportion of toddlers in each group who exhibited *any* pretend play during the 10-min observation. Whereas the majority of LR (77.5 %) and HR-noASD (73.5 %) toddlers engaged in one or more bouts of pretend, only 27 % of the children with a diagnosis of ASD showed *any* pretend play. A Chi square test was significant, $\chi^2(2) = 14.45$, p = .001. Fisher's exact tests indicated that significantly fewer children with ASD engaged in pretend play than either LR or HR-noASD children (p < .01).

Parent Ratings during Free Play—Significant group differences were evident for both the composite sensitivity score, F(2, 141) = 3.54, p = .032, partial $\eta^2 = .048$, and ratings of stimulates cognitive development, F(2, 141) = 5.34, p = .006, partial $\eta^2 = .07$. Parents of children with ASD did not differ from parents of the LR or HR-noASD children on either rating. However, parents of HR-noASD children received significantly higher ratings than parents of LR children on both measures (p < .015, d = .49 for sensitivity; p < .003, d = .59

for stimulates development), indicating that they were especially engaged in supporting their children's play. Results remained the same when the Mullen ELC was controlled.

Given these differences in parent behavior, we re-examined group differences in child engagement with parent controlling for the parent sensitivity composite. Results remained as reported above: both LR and HR-noASD children were significantly more engaged with their parents during play than children with a diagnosis of ASD, indicating that the difference in child engagement was not accounted for by parent behavior.

Elicited Play—Groups differed significantly in both the total score, F(2, 142) = 10.80, p < .001, partial $\eta^2 = .13$, and the highest level of play reached, F(2, 142) = 5.57, p = .005, partial $\eta^2 = .07$. Post hoc Games-Howell tests indicated that the children with ASD received lower total scores on the elicited pretend tasks than either the LR or HR-noASD groups (p < .001 with d's of -1.36 and -1.29 respectively). LR children also reached a significantly higher level of play than the children with ASD (p = .03, d = .83) and the HR-noASD children reached a marginally higher level of play than the children did not differ from each other on either score. This pattern of results was maintained when analyses were recalculated controlling for the Mullen ELC and contrasts were run between the children with ASD and the two comparison groups [total score: F(2, 134) = 4.24, p = .016, LR vs. ASD, p = .006, HR-noASD vs. ASD, p = .007; highest level: F(2, 134) = 3.03, p = .052, LR vs. ASD, p = .016, HR-noASD vs. ASD, p = .03].

In addition, we examined the proportion of children in each group who reached level two (i.e., child as agent acting on the doll). In all three groups, the majority of children were able to imitate the examiner and pretend to act on the doll. However, in both the LR and HR-noASD groups, over 90 % did, whereas only 60 % of the children with ASD imitated at level 2, $\chi^2(2) = 14.32$, p = .002, with both the LR and HR-noASD groups differing from the ASD group on follow-up Fisher's exact tests (p < .01). Very few children reached levels 3 or 4 (the doll acting on herself, the doll acting on the teddy bear) at 22-months: only 11 % of LR, 4 % of HR-no-ASD, and 0 % of children with ASD scored above a level 2 at this age. This is consistent with what one would expect in 22-month-olds.

Finally, we examined the engagement composite score reflecting interest and compliance during task administration. As expected, the children with an ASD diagnosis were less engaged, F(2, 142) = 24.48, p < .001, partial $\eta^2 = .26$, than either the LR or HR-noASD groups (p < .01, with d's of -1.76 and -1.21 respectively). There was also a marginal difference between the LR and HR-noASD groups, with the HR-noASD group somewhat less engaged (p < .054, d = -.45). Group differences on this variable remained significant when the ELC was controlled. Thus, the children with ASD were less engaged with the task than either the LR or HR-noASD children (p < .001), and lower task engagement was not explained by group differences in cognitive functioning.

Associations between Child and Parent Behavior Ratings and Measures of Play

Group differences in child engagement and parent behavior underscore the links between social interest and pretending in toddlers. Thus, we examined correlations between

children's scores on pretend in each context and the relevant rating of child engagement, separately by group. Correlations between parent behavior ratings and free play pretend were also calculated. As can be seen in Table 3, the correlations between behavioral ratings of child engagement and pretend were uniformly positive, with children who were more engaged with their parent and the examiner scoring higher on both measures of pretend. The correlations were consistently higher in the group with ASD; tests for the differences between correlations indicated that child engagement with parent mattered more for children with a diagnosis (ASD vs. LR, Z = 2.50, p = .013; ASD vs. HR-noASD, Z = 1.84, p = .066). A similar pattern was evident for the elicited pretend, but the differences between pairs of correlations were not significant. Ratings of parent sensitivity and stimulation were also more highly correlated with pretend play scores in the ASD group, but correlations did not differ significantly between groups. These data suggest the importance of social engagement and parental support, and especially for the toddlers with a diagnosis of ASD.

Associations between ADOS Severity Scores and Measures of Play

Pearson correlations were calculated between the play measures and the ADOS severity score, within the HR group as a whole and including the one LR toddler with a diagnosis. As expected, ADOS severity scores were negatively correlated with pretend play during free play, r(61) = -.263, p = .041 and with total score during elicited pretend, r(61) = -.557, p < .001. In addition, children with higher severity scores were less engaged with their parents during play, r(61) = -.527, p < .001, and less engaged and cooperative with the examiner during elicited pretend, r(61) = -.577, p < .001. These correlations are consistent with the group differences already reported, but highlight the associations between symptom severity on the ADOS and social engagement and pretend play skills across these two different contexts.

Discussion

We examined pretend play in HR and LR 22-month-old toddlers during free play with a parent and on a structured task requiring them to reproduce pretend play scenarios modeled by an examiner. We also assessed children's social engagement with their parent during play and parent sensitive responsiveness and stimulation of development. Finally, during the elicited pretend task, we rated children's cooperation with the examiner and interest in the task.

The results clearly demonstrate that toddlers who received a diagnosis of ASD were less likely to exhibit pretend play with a parent during an unstructured play interaction than either the LR toddlers or HR toddlers without a diagnosis and they also received lower scores on the highly structured elicited pretend task. Thus, across two different situations meant to support pretend play, the toddlers who received a diagnosis of ASD obtained lower scores than the LR and HR-noASD groups. Importantly, the HR-noASD toddlers did not differ significantly from the LR toddlers on either of these measures of pretend play. Whereas over 70 % of the HR-noASD and LR toddlers showed at least one bout of pretend during free play with their parent, only 27 % of the toddlers with ASD did. Even on the highly structured elicited play task, one in three children with an ASD diagnosis failed to

imitate simple actions requiring them to act on the doll, in contrast to fewer than 10 % of the children in the other two groups. However, a higher proportion of children with ASD were able to imitate some pretend sequences in the structured context, but were less likely to do so during free play, consistent with earlier studies of pretend play in preschool and school-age children with ASD (Rutherford et al. 2007; Sigman and Ungerer 1984). Thus, these results extend the data on deficits in pretend play in preschool-age children with ASD to the toddler period. They also add to the limited data on pretend play in HR toddlers who receive a diagnosis (Christensen et al. 2010) as the deficit in pretend play was evident across two contexts, scaffolded play with a parent and imitation of pretend play scenarios after a demonstration. Furthermore, consistent with the recent findings of Barber and Dissanayake (2013), they highlight early pretend play deficits as an important marker of emerging autism, especially in view of the finding that HR-noASD toddlers did not differ from LR toddlers on either of these pretend play measures.

Consistent with observations of typical development, however, these 22-month-old toddlers, regardless of group and diagnostic status, spent most of their time in functional play and showed only a few instances of simple manipulation. Thus, the children with ASD had moved beyond simply manipulating the toys to show age-appropriate levels of functional play. Furthermore, the children with ASD were clearly interested in the set of toys provided, but even when scaffolded by a parent, they showed few instances of pretend play. This delay or deficit in pretending in the children with ASD is likely to reflect their concomitant lack of social engagement with their parent as well as their lower level of cognitive and language development, as the group difference in pretending was no longer significant with Mullen scores controlled.

It has been suggested that social engagement and shared enjoyment facilitate pretend play in both typically developing toddlers (Lillard 2007) and in children with ASD (Hobson et al. 2013; Sigman and Ungerer 1984). In the current study, the HR-noASD children were similar to the LR children, not only in their pretend play, but also in their engagement with their parents. In contrast, children with ASD showed the lowest level of shared enjoyment and social engagement during play with their parent. Moreover these group differences in engagement with their parent were not accounted for by differences in cognitive functioning. At the same time, the correlations presented in Table 3 indicate that child engagement with parent mattered most for the pretend play of children with ASD. Children with a diagnosis were more likely to exhibit some pretend play when they were also more socially engaged with their parent during the free play observation. A similar pattern of correlations during the elicited pretend play sequence also suggests that interest in the task and cooperation with the examiner, which also are likely to reflect some degree of social engagement, are associated with higher scores. Although the correlations between cooperation and total score are positive across groups, they are especially high in the group with ASD. It is not clear whether this is because children's social interest and engagement with others, i.e., both parent and examiner, is especially important in the early development of pretense in this group, or whether it is because children in this group who can pretend are more likely to elicit engagement from others. Nevertheless, the combination of less social engagement with play partners and less pretend play, primarily simple pretend like making believe one is

feeding a doll or washing her face, are associated with a diagnosis of ASD in HR siblings and correlated significantly with ADOS severity scores.

In two prior reports from this study, with overlapping samples (Campbell et al. 2015a, b), we found that HR infants with a diagnosis were less engaged with parents during free play at 11 months than either the LR or HR-noASD infants. At 22 months, they were also less likely to solicit their parents during an empathy probe when they were confronted with a "crying baby." Thus, across three different types of laboratory situations and at two ages, HR siblings who received a diagnosis of ASD showed less interest in social interaction with their parents.

Despite the group differences in children's social interest and shared enjoyment with parents, the parents of the diagnosed children did not differ from the parents of LR children at either 11 (Campbell et al. 2015a) or 22 months. At both ages, we rated parent behavior during a free play interaction using well-validated ratings of parent sensitivity and warmth as well as stimulation of cognitive development (NICHD ECCRN 1999). Ratings were made by a separate team of coders who were blind to group and diagnostic outcome and did not code the child's pretend play. It is noteworthy, albeit not surprising, that the parents of the LR children and the parents of the children with an ASD diagnosis were generally rated as sensitive and stimulating. The parents of the HR toddlers without a diagnosis, however, received even *higher* ratings and they differed significantly from the parents of the LR toddlers.

Thus, the parents of HR toddlers without ASD were seen as especially tuned into their children's needs during the play session and they provided a higher level of stimulation and scaffolding of their child's play than parents of LR toddlers. This suggests that they were strongly invested in supporting their toddler's development in view of their prior history raising a child with ASD, their toddler's level of social engagement, and possibly their awareness of the importance of early intervention and participation in parent-administered treatment regimens. This is also likely to reflect the fact that their children were more engaged with them than were the HR toddlers with a diagnosis. That is, this may be parentchild reciprocity at work, an example of a bidirectional feedback loop. Because the toddlers with ASD were less socially engaged and presumably harder to draw into reciprocal play than the HR-noASD children, their parents likely had fewer opportunities to provide the high level of stimulation and scaffolding seen among parents of HR toddlers without ASD. In a recent paper examining mothers' use of gestures and children's later language acquisition in HR and LR infant siblings, Talbott et al. (2015) reported that mothers of HR infants who did not receive a later diagnosis used more gestures to scaffold language development at 12 months than mothers of LR infants. Furthermore, maternal gesture use at 12 months predicted language development at 18 months. These authors also suggest that the mothers of the HR-noASD infants were especially attuned to the developmental functioning of their infants, an observation consistent with the current results.

The strengths of this study include the collection of prospective data on a carefully ascertained sample of HR and LR siblings, most followed from infancy to toddlerhood, the careful follow-up by independent testers, and the use of age-appropriate measures of play,

derived from studies of typical development. In addition, the use of two independent assessments of pretend play along with ratings of child engagement across these two play situations, add to the robustness of the findings. Finally, the ratings of parent behavior during a typical, unscripted play interaction provide new information about diagnostic status, children's social engagement, and parental sensitivity and scaffolding.

The limitations of this study include the relatively small number of toddlers with a diagnosis, missing data on the Mullen scales of early learning at 24 months, and the fact that we did not have the resources to conduct more fine-grained coding of either the specifics of toddler free play or parent involvement in specific play acts. However, continued follow-up through 36 months will allow us to examine trajectories of pretend play at 22, 28, and 34 months and to obtain more complete outcome data at 36 months. In addition, it is difficult, if not impossible, to tease apart the contributions of cognitive and language delay from those of lower social engagement in explaining the group differences in pretend play. Our follow-up data may provide a clearer picture of the relative roles of social engagement and developmental delay in accounting for these group differences.

Although more research is needed with larger samples of HR toddlers to further validate and extend these findings, we can tentatively conclude that the combination of low levels of social engagement, pretend play, and empathic concern (Campbell et al. 2015b), evident at 22 months, are clear signs of emerging ASD. Importantly, the HR toddlers without a diagnosis did not differ from the LR toddlers in either social engagement or pretend play at 22 months, suggesting some degree of specificity. From a clinical perspective, early interventions that encourage spontaneous social engagement with parents and other familiar adults appear important (Dawson 2008) and our data indicate that parents are already actively scaffolding play and social interaction with their toddlers. These data also underscore what is already known about typically developing toddlers (Brownell and Kopp 2007; Lillard 2007), that parents are an especially important source of support for young children's ability to engage in pretend play, a skill that also has implications for social interactions with peers by the preschool years. Taken together, these results suggest that social interest and engagement motivate participation in shared activities, as reflected early in joint attention (Rutherford et al. 2007), and then in shared play sequences that give rise to and promote the continued development of imitation and pretending in both HR and LR toddlers (Hobson et al. 2013; Lillard 2007).

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Table 1

Sample characteristics

	ASD (n = 15)	HR-noASD $(n = 49)$	LR(n = 81)
Gender, % male	87 %	57 %	57 %
Ethnicity, Caucasian	80 %	84 %	88 %
Parent education			
High school/some college	50 %	36 %	14 %
College degree	29 %	34 %	42 %
Graduate degree	21 %	30 %	44 %
Age at 22 month assessment, M (SD)	22.78 (.70)	22.72 (.73)	22.76 (.68)
Mullen early learning composite, M (SD)	77.71 (17.14)	98.96 (13.04)	106.16 (15.63)
ADOS severity score, M (SD)	5.07 (1.39)	1.60 (.92)	1.41 (.91)

Seven children are missing Mullen scores (one ASD, four HR-noASD, two LR); one ASD child is missing an ADOS severity score. See text for explanations

ASD toddlers with a later ASD diagnosis, HR-noASD high risk toddlers without a later ASD diagnosis, LR low risk toddlers

Table 2

Means and standard deviations of free play and elicited pretend play measures

	ASD (n = 15) M (SD)	HR-noASD (n = 49) M (SD)	LR (n = 81) M (SD)
Free play			
Simple manipulation	2.60 (3.76)	1.18 (1.75)	1.51 (2.09)
Functional	15.20 (4.87)	14.98 (3.41)	14.39 (4.10)
Pretend	.73 (1.49) ^a	3.16 (3.33) ^b	3.18 (3.74) ^b
Engagement w/parent	1.70 (.77) ^a	2.83 (.83) ^b	2.97 (.81) ^b
Parent ratings			
Sensitivity composite	5.33 (1.58)	6.10 (1.12) ^a	5.38 (1.77) ^b
Stimulates cognitive development	2.73 (1.08)	3.32 (.73) ^a	2.81 (.97) ^b
Elicited pretend play			
Total score	5.33 (4.50) ^a	11.69 (5.03) ^b	12.44 (5.59) ^b
Highest level	1.47 (.74) ^a	1.96 (.50)	2.04 (.64) ^b
Engagement composite	5.47 (2.88) ^a	8.71 (2.48) ^b	9.67 (1.75) ^b

One LR child is missing data on free play

ASD high risk toddlers with a later ASD diagnosis, HR-noASD = high risk toddlers without later ASD diagnosis, LR = low risk toddlers

Means with different superscripts differ significantly at p < .05 or better. See text

Table 3

Correlations between pretend play scores and behavior ratings

	ASD (n = 15)	HR-noASD (n = 49)	LR (n = 81)
Free play	-		
Child engagement with parent	.763 **	.386***	.225*
Parent sensitivity	.528*	.340*	.113
Stimulates cognitive development	.547*	.205	.349 **
Elicited pretend			
Child engagement in task	.750 **	.402 **	.582 **

One LR child is missing data on free play