Research Article

Early Play Behaviors of Infants at Elevated Likelihood for Autism Spectrum Disorder

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Importance: Although research has examined early identification of autism spectrum disorder (ASD), few studies have found behavioral markers during midinfancy associated with later ASD diagnosis.

Objective: To examine infants' play behaviors and atypical positions at age 6 mo and later outcome classification among infants at elevated likelihood (EL) and typical likelihood (TL) for ASD. *Atypical positions* refer to movement patterns indicative of motor delays or deviations, including atypical extension and flexion, poor weight shift and rotation, hypertonicity or hypotonicity, and the presence of primitive reflex patterns.

Design: Observational cohort longitudinal design using blinded video analysis.

Participants: Fifty-eight infants (41 EL infants and 17 TL infants) 6 mo of age.

Results: Infants later diagnosed with ASD needed more support to engage in play at age 6 mo compared with infants who did not receive an ASD diagnosis ($U = 130$, $z = -2.29$, $p < .05$, $r = .31$). Atypical positions at 6 mo of age were not associated with a later diagnosis of ASD.

Conclusions and Relevance: Play behaviors may be early indicators of developmental differences for infants later diagnosed with ASD. The results of this pilot study suggest the need to observe the quality of interaction with a caregiver and objects during early play, which may serve as potential early indicators of ASD.

Plain-Language Summary: Few studies have found behavioral markers during midinfancy that are associated with a later diagnosis of autism spectrum disorder (ASD). The results of this study showed that infants who were later diagnosed with ASD were found to have differences in play behaviors compared to infants who were not later diagnosed with ASD. Thus, early play behaviors may be an early indicator of developmental differences for infants who are later diagnosed with ASD.

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Autism spectrum disorder (ASD) is a neurodevelopmental disability characterized by differences in social communication and the presence of restricted and repetitive behaviors and interests (American Psychiatric Association, 2022). The developmental gap between children diagnosed with ASD and children not diagnosed with ASD often widens as they mature (Landa et al., 2013) and contributes to significant difficulties engaging in similar tasks as same-age peers. The effects of ASD (American Psychiatric Association, 2022) vary greatly among children and their families, but early intervention may improve a child's development (Guthrie et al., 2023; Landa et al., 2011; Landa et al., 2013). Early intervention may identify strengths of young children with ASD and exert a powerful influence on their neurological organization, thereby facilitating learning and developmental progress well beyond the early years (Fox et al., 2010). Early identification of ASD may lead to earlier access to intervention.

Although a diagnosis of ASD is not often made before the child is 4 yr of age (Maenner et al., 2023), studies have reported divergence from expected developmental trajectories and emergence of ASD features from late infancy to 2 yr of age among children later diagnosed with ASD (Chawarska et al., 2014; Landa et al., 2007; Zwaigenbaum et al., 2021). Research suggests that subtle signs, including differences in social behaviors, motor behaviors, and sensory processing, may emerge between mid- to late infancy

(Baranek, 1999; Tanner & Dounavi, 2021; Zwaigenbaum et al., 2021).

Few prospective studies of infants in midinfancy have found ASD symptoms. However, prospective studies using brain imaging technology have found white matter abnormalities at 6 mo of age and increased growth rate of the cortical surface area that co-occurs with sensorimotor and visual orienting differences from 6 to 12 mo of age for infants at elevated likelihood (EL) of ASD who are later diagnosed with autism (Hazlett et al., 2017; Klin et al., 2015; Shen et al., 2013; Wolff et al., 2012). Early difficulties with dynamic eye gaze, visual attention, and visual orienting, which can affect later social interaction patterns (Elsabbagh et al., 2012; Jones & Klin, 2013), are linked to the motor system (Flanagan et al., 2012; Leezenbaum & Iverson, 2019).

Movement is the medium in which infants explore, interact with people and objects, respond to environmental demands, and learn about the world (Campbell et al., 1995). Early motor challenges, such as reduced motor control, atypical movement patterns, and atypical general movements (Phagava et al., 2008; Zappella et al., 2015; Zwaigenbaum et al., 2021), are associated with a later ASD diagnosis. Delayed fine motor skills have also been reported among EL infants at 6 mo of age compared with infants with typical likelihood (TL) for ASD and were associated with Autism Diagnostic Observation Schedule (ADOS; Lord et al., 2012) severity scores but were not specific to ASD diagnosis at 3 yr of age (Iverson et al., 2019). Motor delays and atypical movement patterns during infancy may affect infants' exploration and interaction with objects and people in the environment and thus their participation in early occupations (Flanagan et al., 2012). These findings have implications for future development because early exploration and manipulation facilitate learning about object affordances while providing the foundation for more mature play skills (May-Benson, 2010).

Scholarly work validates a key ideological premise of occupational therapy, which posits that individuals acquire abilities and build proficiencies through a process of active engagement (Mroz et al., 2015). This concept can be observed in play behaviors as early as infancy. Fine and gross motor skills and social communication skills are aspects of early play that involve the infant exploring and interacting with people and objects. Through play, children develop motor, language, cognitive, and social–emotional skills (Rigby & Rodger, 2006). According to the American Academy of Pediatrics, play is critical in promoting safe and nurturing relationships, developing competencies, promoting healthy child development, and improving life course trajectories (Yogman et al., 2018). Variation in play development, including less mature play behaviors during late infancy, may be an early EL indicator of later ASD diagnosis and has been associated with language and social communication differences (Poon et al., 2012; Wilson et al., 2017). In fact, parents of EL infants who were later diagnosed with ASD reported play concerns when their children were as young as 9 mo of age compared with parents of EL non-ASD and TL infants (Sacrey et al., 2015). Observing the quality of infants' interaction with objects and people in their environment through play can serve as a window into their cognitive development (Belsky & Most, 1981).

The literature suggests that play can be a valid and reliable measure of development. However, a gap exists in the literature examining early play skills among infants younger than 9 mo of age at EL for ASD. This suggests the need to examine play behaviors and movement among 6-mo-old infants at EL and TL for ASD. The present pilot study sought to answer the following research questions:

- Do infants who are later diagnosed with ASD demonstrate decreased play abilities at 6 mo of age compared with children who are not later diagnosed with ASD?
- Do infants later diagnosed with ASD exhibit more atypical positions (movement patterns indicative of motor delays or deviations) than infants who do not have a later diagnosis of ASD?

We hypothesized that infants later diagnosed with ASD would demonstrate decreased play abilities and more atypical positions at 6 mo of age compared with infants who were not later diagnosed with ASD.

Method

In this pilot study, we used an observational cohort longitudinal design to examine early play behaviors and atypical positions among 6-mo-old infants at EL and TL of ASD. Archived videos of 6-mo-olds were coded (we were naïve to infant group and to outcome diagnostic status). This study was approved by the Towson University institutional review board (IRB). The infants were part of a larger federally funded longitudinal study approved by the Johns Hopkins University IRB, and the university's IRB-approved informed parental consent was obtained for all participants.

Participants

Participants for this study were drawn from a larger longitudinal study that followed 131 infants ages 5 to 10 mo. through their 36th mo. Inclusion criteria for infants in the larger study were as follows: (1) English as primary language in the home; (2) birth weight ≥2,250 g; (3) gestational age >37 wk; and (4) no severe birth trauma or congenital conditions, brain injury, prenatal illicit drug, or excessive exposure to alcohol. Infants in the larger study were classified as EL if they had an older sibling diagnosed with ASD on the basis of the ADOS (Lord et al., 2012) and expert clinical judgment. Infants were classified as TL if they did not have first- or second-degree relatives with a diagnosis of ASD. At 36 mo, these infants received diagnostic

classifications of ASD, non-ASD social communication delay, or nondelay.

For this pilot study, we selected 41 EL and 17 TL infants from the larger study, all between ages 6 and 7 mo. A research assistant matched the EL infants in the nondelay group by age and sex to EL infants later diagnosed with ASD and EL infants with non-ASD social communication delay. In this study, there were two possible diagnostic classifications: ASD and non-ASD (combining non-ASD social communication delay and nondelay groups because of the small sample size). Outcome classification was based on the results of the ADOS (Lord et al., 2012) and clinical best estimate by expert clinical researchers. Of the 58 infants in the current study, 12 infants (10 EL and 2 TL) were in the ASD group (age range = 6.02–6.61 mo), and 46 infants (31 EL and 15 TL) were in the non-ASD group (age range = 6.02–6.97 mo).

Instruments

Revised Knox Preschool Play Scale

We used the Revised Knox Preschool Play Scale (RKPPS; Knox, 2008), an observational rating scale, to code overall developmental play age. The RKPPS describes play in 6-mo increments for an infant's first 36 mo and then in yearly increments from 3 to 6 yr of age. The scale examines four dimensions of play: space management (gross motor skills), material management (fine motor skills), pretense symbolic (early imitation), and participation. The earlier Preschool Play Scale (PPS) version and this more recent RKPPS version have demonstrated adequate psychometric properties (Harrison & Kielhofner, 1986; Lee & Hinojosa, 2010). Although the PPS (Knox, 1974, 1997, 2008) has more studies on its reliability and validity than the RKPPS, the revised scale describes play in more detail for infants. Other infant play outcome measures were reviewed and deemed not appropriate for scoring archived videos or as having limited information on object play at midinfancy. We chose the RKPPS because it assesses motor, imitation, and participation dimensions of play, which are known to be difficult for young children with autism.

We followed the procedures outlined by Knox (2008) to code play behaviors and calculate the play age, derived by calculating the mean of all four dimensions. Before coding, the first and second authors (Joanne E. Flanagan and Barbara B. Demchick) operationalized the coding scheme. For this study, we collaborated with the author of the RKPPS to operationally define play behaviors, which needed clarification (S. Knox, personal communication, January 8, 2012). *Imitation* was defined as an infant demonstrating observed changes in facial expressions, physical movement, or vocalizations or mouth movements in response to the caregiver's or examiner's facial expressions, physical movement, and vocalizations. *Cooperation* was defined as demanding attention and engaging in simple give-and-take interaction with the caregiver or examiner (e.g., tickling, peekaboo). We gave credit if the infant was attending to the adult 75% of the time during a 2-min reciprocal social play protocol. No credit was awarded if gaze aversion was observed or if there was increased effort by the caregiver to engage the infant (e.g., adults repeatedly calling the baby's name, touching the baby, or raising their voices).

The distinction between emerging skills and mastery of skill also needed clarification. Although infants were not given credit for emerging skills, infants who were seen banging, shaking, hitting, handling, or mouthing objects three or more times received credit for that respective manipulation descriptor.

Atypical Positions Subtest of the Toddler and Infant Motor Evaluation

We used the Atypical Positions subtest of the Toddler and Infant Motor Evaluation (TIME; Miller & Roid, 1994) to measure atypical positions at age 6 mo by coding infant movement patterns during the videotaped administration of the gross motor domain of the Mullen Scales of Early Learning (Mullen, 1995). The Atypical Positions subtest has pictorial illustrations of atypical movement patterns, including poor weight shifting, hypertonicity or hypotonicity, and presence of primitive reflexes. This subtest has been found to distinguish between children with and without motor delays or deviations (Miller & Roid, 1994). Further psychometric properties for clinical and research use have been documented for this subtest. We followed the scoring procedures outlined by Miller and Roid (1994) to determine the total raw score and scaled score using available normative data.

Procedure

Archived videotapes of the 6-mo-old infants in this study were previously filmed during standardized tasks that included the Mullen Scales of Early Learning (Mullen, 1995) and a reciprocal social infant–adult play task as part of the original larger study. Videos were coded by two occupational therapists (Flanagan and Demchick), each with more than 15 yr of experience assessing and treating infants and young children with developmental disabilities. When coding videotapes for this pilot study, we were blind to the infants' EL or TL status and the established diagnostic outcome. We used scoring procedures outlined in the TIME manual for the Atypical Positions subtest (Miller & Roid, 1994) and for play behaviors outlined for the RKPPS (Knox, 2008).

We coded 17 (29%) infants' tapes to assess interrater reliability using Cohen's κ. Ten infants (17% of the total tapes) were randomly picked for interrater reliability. Because the primary investigator (Flanagan) knew the EL or TL status of 4 infants and the outcome classification of 3 infants from a previous study (Flanagan et al., 2012), these 7 infants were also included with the number of infants to code for interrater

reliability, for a total of 17 infants. To maintain the rigor of the study, we used the original scores from Demchick, who remained blind to the EL or TL status and outcome classification of these 7 infants, rather than the scores from Flanagan for the data analysis procedures with these 7 infants.

Flanagan's scores were used for the data analysis procedures in this study with the exception of scores for the aforementioned 7 infants. However, if there was a disagreement between the two raters' scores during the process of establishing interrater reliability for the other 10 infants, there was a discussion to reach a consensus; consensus scores were then used for data analysis. The κ value was .63 for total play score and .68 for atypical positions raw score. These values were considered substantial agreements between the two raters (Landis & Koch, 1977).

Data Analysis

We analyzed data using IBM SPSS Statistics for Windows (Version 28). Although there were no missing data for the atypical positions variable, data were missing for some play behaviors for 4 infants because of poor video quality or missing sections (e.g., caused by infant fussiness). For these cases, we performed the analysis without mean replacements for the missing data. Because this was a pilot study with a small sample size, we used nonparametric tests in the analysis (Field, 2009). Descriptive statistics were performed on all variables. We estimated group comparisons for age, sex, race, and socioeconomic status (SES; Hollingshead, 1975). We used Fisher's exact test for analyses involving sex and race because the expected count was <5 in each category; we used Mann–Whitney *U* tests for analyses involving SES and age and for testing group differences in play scores. Because research has demonstrated differences between infants in the TL and EL groups (Iverson et al., 2019), we did an additional analysis comparing the ASD group with the TL non-ASD and EL non-ASD groups using a Kruskal–Wallis test and Bonferroni correction to adjust for multiple pairwise comparisons. We used Fisher's exact test to test for between-groups differences in atypical positions.

Results

Sample Characteristics

Descriptive statistics showed no significant group differences for age, sex, race or ethnicity, or SES between ASD and non-ASD groups (see Table 1).

Play Behaviors

Total play scores for infants later diagnosed with ASD at age 30 to 36 mo were lower at 6 mo compared with infants without a later ASD diagnosis ($U = 130$, $z = -2.29$, $p = .022$, $r = .31$; see Table 2). There were no significant differences among the three groups when the ASD group was compared with the EL non-ASD and TL non-ASD group using a Kruskal–Wallis test and Bonferroni correction ($H = 5.85$, $p = .054$). However, there was a trend toward lowest scores for the ASD group, followed by the EL non-ASD group; the TL non-ASD group tended to have the highest play age (see Figure 1).

Movement

The presence of atypical positions at age 6 mo (see Table 2) was not significantly different for infants in the ASD group compared with infants in the non-ASD groups ($p = .166$). However, 42% (5 out of 12) of infants in the ASD group were observed to display asymmetrical tonic neck reflex (ATNR) position compared with 19% (6 out of 31) of infants in the EL non-ASD group and 20% (3 out of 15) in the TL non-ASD group.

Discussion

This study examined whether overall play behaviors and atypical positions of infants at age 6 mo were associated with a diagnosis of ASD at 30–36 mo. Results

Table 1. Demographic Characteristics of Infants in the ASD and Non-ASD Groups

Characteristic	Total (*N* = 58)	ASD Group (*n* = 12)	Non-ASD Group (*n* = 46)	*p*
Age, mo., *M* (*SD*)	6.37 (0.25)	6.26 (0.20)	6.38 (0.26)	.16
Male gender, *n* (%)	31 (53)	9 (75)	22 (48)	
Race/ethnicity, *n*				.23
White	48	9	39	
Black/African American	1	0	1	
Asian	2	0	2	
Multiracial	3	2	1	
Unknown/not reported	4	1	3	
Socioeconomic status, *M* (*SD*)	57.92 (7.34)	60.41 (3.11)	57.20 (8.06)	.21

Note. All comparisons were made using Student's *t* test for age and socioeconomic status; chi-square was used for race and gender. ASD = autism spectrum disorder.

Table 2. Median Scores of Play Age and Percentage of Atypical Positions in ASD and Non-ASD Groups

Infant Measure	ASD (n = 12)	Non-ASD (n = 46)	p
Play age score, mo.	3.5	5.4	.022*
Atypical positions, %	66	46	.166

Note. Play age in months was compared using a U statistic; atypical positions were dichotomized (presence–absence) and compared using Fisher's exact test. ASD = autism spectrum disorder. *$p < .05$.

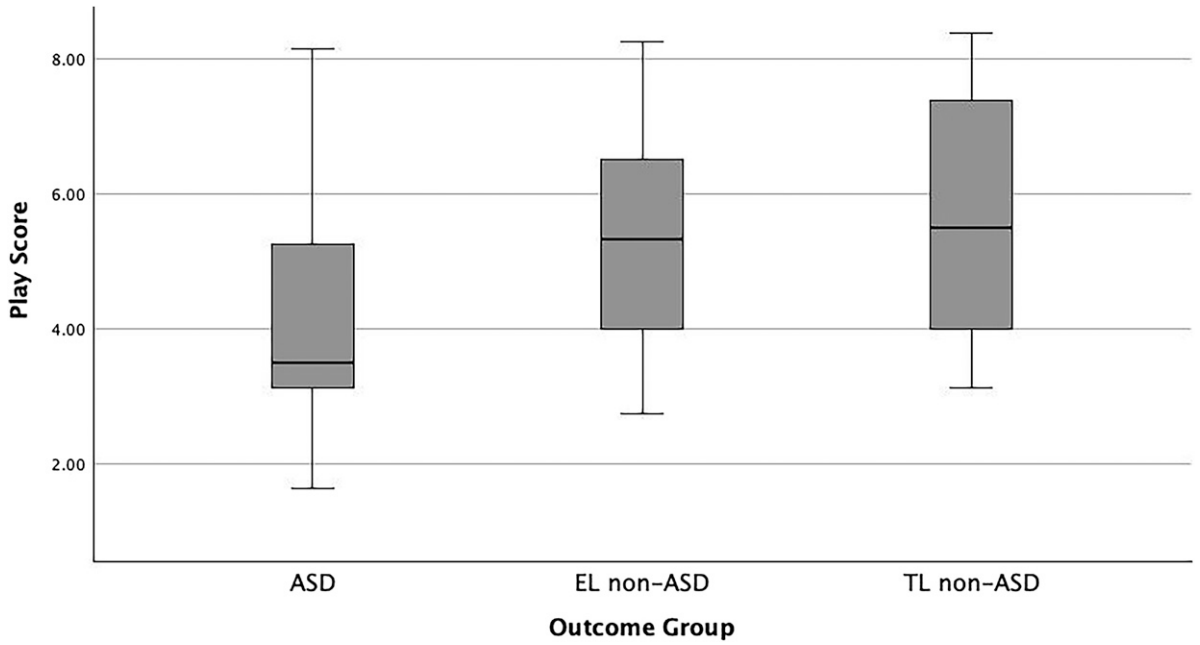
indicated that total play scores differentiated infants with ASD from infants with non-ASD at age 6 mo but atypical positions did not. To our knowledge, this study is the first to show play behaviors among infants as young as 6 mo of age associated with a later diagnosis of ASD.

The RKPPS measured overall play age during participation in early play with objects and the caregiver or examiner in the environment. The total score of the RKPPS is composed of items that relate to gross motor, fine motor, early imitation, and participation dimensions. Because of the pilot nature of our study and the limited sample size, we refrained from analyzing scores across various domains to mitigate the risk of inflating Type I error. Nonetheless, clinically significant behaviors were observed in the ASD group, including decreased environmental exploration, limited variety of play with toys, reduced imitation, and restricted reciprocal social interaction with adults.

Because of the heterogeneity of ASD, it is unlikely that a single domain will be a strong indicator of ASD at 6 mo of age, but the application of several items to complete an integrated task for an infant, such as playing peekaboo with a caregiver, may yield important information of early occupational challenges. Playing peekaboo requires the complex integration of eye contact with smiles and vocalizations when engaging with others, reciprocal relationships, attention to task, anticipation of the adult's actions, and coordination of body movements, which all enable participation in early play occupations. Therefore, it is not surprising that the infants later diagnosed with ASD showed fewer play behaviors at 6 mo compared with the infants not later diagnosed (Libertus et al., 2014). The literature suggests that early social and motor signs of ASD emerge between the ages of 6 and 12 mo (Tanner & Dounavi, 2021, Zwaigenbaum et al., 2021). Our pilot study supports this by suggesting their emergence at 6 to 7 mo when examining play behaviors.

Our findings are supported in the literature in studies that have examined play skills in older infants, toddlers, and preschoolers. Restall and Magill-Evans (1994) found lower total play scores among preschoolers with ASD compared with typically developing children using the PPS (Knox, 1974). EL infants at 9 to 12 mo of age presented with less mature play behaviors compared with those in the typically developing control group (Wilson et al., 2017). Furthermore, object play skills and early social communication skills (imitation and joint attention) in late infancy were predictive of intellectual and communication functioning among children with ASD at 3 to 7 yr of age (Poon et al., 2012). The importance of early developing motor and manipulation skills for cognitive, social, and language development highlights the need for

Figure 1. Distribution of infant play scores for the ASD, EL non-ASD, and TL non-ASD groups.



Note. Boxes represent the interquartile range with the median indicated by a central line. Whiskers extend to the highest and lowest values. ASD = autism spectrum disorder, EL = elevated likelihood, TL = typical likelihood.

research focusing on the relationship between early qualitative aspects of movement and early play among EL infants (Iverson et al., 2019). Through engagement in occupations such as exploration in the environment, play, and social interactions, children acquire, integrate, and refine skills and optimize their development (Humphry, 2002). Our findings suggest the need to move beyond standard assessments that focus primarily on the accomplishment of gross or fine motor tasks to examine the quality of play during social interaction and environmental exploration, which may together serve as potential early indicators of ASD. Specifically, the findings of this study indicate that assessing infants' early play skills at 6 mo of age in a social context may reveal early signs of differences in motor-based functional skill development among EL infants later diagnosed with ASD.

Regarding our second research question, we did not find differences in atypical positions between the infants in the ASD and non-ASD groups. However, many of the infants in this study demonstrated ATNR, although not in the obligatory manner associated with neuromotor disorders (e.g., cerebral palsy). Twice as many infants in the ASD group displayed ATNR positions compared with the infants in the non-ASD groups. The presence of ATNR after 6 mo of age is a possible indicator of concern about the developing nervous system (Hamer & Hadders-Algra, 2016). As reported in the results, 19% of EL non-ASD infants also demonstrated this atypical position. This is consistent with literature reporting motor differences among EL infants (e.g., Flanagan et al., 2012; Leezenbaum & Iverson, 2019). Surprisingly, the TL non-ASD infants showed a similar percentage of ATNR as the EL non-ASD group. Studies have reported that families are more likely to participate in research studies when they have concerns about their children, so infants in the control groups in our study may not be representative of the general population (Hadders-Algra et al., 2010).

This presentation of atypical movement patterns among infants later diagnosed with ASD has been reported in the literature (Phagava et al., 2008) and may have predictive significance for neurological signs (Hamer & Hadders-Algra, 2016). Atypical movement may restrict active exploration and limit infants' interaction with objects and people and the subsequent development of foundational early infant occupations (Flanagan et al., 2012). Such missed opportunities can have negative cascading effects on motor, perceptual, language, and cognitive development (Leezenbaum & Iverson, 2019).

Limitations

This study had a small sample size, with a relatively small number of infants receiving an ASD diagnosis and a few infants with missing data. We recommend that additional studies be conducted with larger samples. Although there had been several reliability and validity studies of the PPS version, including with infants, the RKPPS had not previously been used to evaluate play behaviors among infants who have an increased likelihood for ASD. The videos of most of the children in this study took place in a clinical setting using a standardized protocol. Videos obtained in less structured conditions in a more natural context may provide more insight into infant play. However, the standardized items involved the presentation of toys as well as playful interactions with adults, allowing for sensorimotor exploration, object manipulation, early imitation, and social exchanges typical of infants that age. The relation between play and motor skills also needs to be further examined using more sophisticated motion analysis.

Implications for Occupational Therapy Practice

The results of this study have important clinical implications for early identification and intervention. Given the role early intervention can play in enhancing infant development (Guthrie et al., 2023; Landa et al., 2011), evaluating and intervening at earlier ages, even before diagnosis, may help improve outcomes for infants and young children at EL of ASD (Green et al., 2017). This study has the following implications for occupational therapy practice:

- Evaluating early play behaviors among infants at EL of ASD may provide valuable contributions to early indicators of ASD.
- The importance of early play skills for cognitive, language, and social development highlights the need for practitioners to focus on qualitative aspects of early play among infants at EL of ASD.
- When challenges in early play behaviors are identified, early intervention should be recommended to improve outcomes.

Conclusion

Given the high incidence of ASD and its impact on child development and family functioning, it is imperative to identify early indicators so that intervention can begin early for the best chance to enhance outcomes (Guthrie et al., 2023). This pilot study showed that even at 6 mo of age, there are differences in infants' play skills that can be identified by a skilled practitioner that may predict later diagnosis of ASD. One of the earliest indicators of ASD may be related to occupational behaviors of how an infant engages in moving within the environment and interacting with objects and others during early childhood occupations. Findings may lead to earlier detection and intervention and guide practice to improve outcomes.

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