

RESEARCH ARTICLE

Cataloguing and characterizing interests in typically developing toddlers and toddlers who develop ASD

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Funding information

Autism Speaks, Grant/Award Number: 6020; National Institutes of Health, Grant/Award Number: R01 HD055741; NIH, Grant/Award Numbers: K12-HD055887, HD079124, HD86984; Simons Foundation, Grant/Award Number: 140209

Abstract

Intense interests are common in children with and without autism spectrum disorder (ASD), and little research has characterized aspects of interests that are unique to or shared among children with and without ASD. We aimed to characterize interests in a sample of infants at high-familial-risk (HR) and low-familial-risk (LR) for ASD using a novel interview. Participants included HR siblings who were diagnosed with ASD at 24 months (HR-ASD, $n = 56$), HR siblings who did not receive an ASD diagnosis at 24 months (HR-Neg, $n = 187$), and a LR comparison group ($n = 109$). We developed and collected data with the Intense Interests Inventory at 18- and 24-months of age, a semi-structured interview that measures intensity and peculiarity of interests in toddlers and preschool-aged children. Intensity of interests differed by familial risk at 24 months, with HR-ASD and HR-Neg groups demonstrating equivalent intensity of interests that were higher than the LR group. By contrast, peculiarity of interest differed by ASD diagnosis, with the HR-ASD group showing more peculiar interests than the HR-Neg and LR groups at 24 months. At 18 months the HR-ASD group had more peculiar interests than the LR group, though no differences emerged in intensity of interests. This measure may be useful in identifying clinically-relevant features of interests in young children with ASD. We also replicated previous findings of males showing more intense interests at 18 months in our non-ASD sample. These results reveal new information about the nature of interests and preoccupations in the early autism phenotype.

Lay summary: Intense interests are common in young children with autism and their family members. Intense interests are also prevalent among typically-developing children, and especially boys. Here we catalog interests and features of these interests in a large sample of toddlers enriched for autism risk. Children who had family members with autism had more intense interests, and those who developed autism themselves had more unusual interests at 24 months. These results highlight the importance of different aspects of interest in autism.

KEYWORDS

attention, autism spectrum disorder, intense interests, problem behavior, restricted interests, toddlers

INTRODUCTION

Autism spectrum disorder (ASD) is a neurodevelopmental disorder that develops in early childhood and is characterized by difficulties with social communication and interaction and restricted and repetitive behaviors (RRBs; American Psychiatric Association, 2013). One specific form of RRB occurring in individuals with ASD is restricted interests organized around a particular topic. Several terms have been used to describe this phenomenon in ASD in the research literature (e.g., special interests, circumscribed interests, unusual interests, intense interests, restricted interests, unusual preoccupations) with little attempt to operationalize these different terms (Cuccaro et al., 2003; Kanner, 1943; Lewis & Bodfish, 1998; Turner-Brown et al., 2011), or differentiate restricted interests from general interests of childhood (regardless of ASD diagnosis). Many typically developing (TD) young children also demonstrate long-lasting interests which manifest in multiple contexts and are readily observable to others (DeLoache et al., 2007). This begs the question: do interests in ASD differ from those that are characteristic of typical development along some dimensions or dimensions that can be operationalized and measured? Furthermore, are there features of interests that may be unique to children with ASD relative to their peers, or relative to other interests they may have? To the best of our knowledge, no studies have comprehensively characterized the interests of children who go on to develop ASD early in development. The goals of this study were (a) to characterize interests in toddlers at risk for ASD and TD toddlers, (b) to evaluate sex differences of interests in these samples, and (c) to determine what aspects of interests differentiate TD children from those with ASD.

Intense interests are common in young children. DeLoache et al. (2007) found that 30% of their sample of children under 6 years of age demonstrated what they termed “extremely intense interests,” with higher prevalence in males compared with females, and the average age of onset at 18 months. Other studies have also found more intense interests in males compared with females (Leekam et al., 2007; Wolff et al., 2016). Intense interests consumed children’s attention, where parents noted that children frequently searched for their interests in the environment and showed others objects related to their interests. Parents generally responded positively to these interests and provided them with relevant toys, books, and videos. This is in line with theories which posit that interests are functional and are thought to guide attention and learning and promote cognitive development (Berlyne, 1949; Hidi, 2001; Renninger & Wozniak, 1985), as the interests motivate acquisition and organization of specialized knowledge (Johnson & Mervis, 1994).

Children with ASD display a range of interests, and when these interests cause impairment to social relationships or family routines, they are characterized as a symptom of ASD in the RRB domain. RRB symptoms

(particularly repetitive motor movements, repetitive play) can be characterized reliably as young as 12 months of age (Bryson et al., 2008; Dimian et al., 2017; Elison et al., 2014; Sifre et al., 2021; Wolff et al., 2014; Wolff et al., 2019), with the overall number of repetitive behaviors, as well as stereotypical movements and restricted behaviors differentiating children who go on to develop ASD as early as 12 months (Wolff et al., 2014). By 24 months of age, children with ASD demonstrate more unusual preoccupations than TD children and children with developmental delays (Richler et al., 2007). Studies focusing on later childhood have found that youth with ASD show the same number of interests as their TD peers but their interests are more intense, unusual in content, and cause more functional impairment than everyday interests in TD children (Anthony et al., 2013; Klin et al., 2007; South et al., 2005; Turner-Brown et al., 2011). No studies have investigated whether very young children with ASD demonstrate differences in the form of their interests compared with TD children.

Family members of individuals with ASD also demonstrate features that fall along with a broader autism phenotype (BAP), including personality traits of rigidity, aloofness, anxiety, and hypersensitivity (Piven et al., 1997). Some specific forms of RRB, such as rituals and insistence on sameness in particular, appear to follow familial aggregation (Lam et al., 2008). Preoccupations in youth with ASD have been shown to be associated with their father’s levels of rigidity and aloofness (Smith et al., 2009). Furthermore, family members without ASD in multiplex families (i.e., more than one family member diagnosed with ASD) show a more restricted range of interests than family members of simplex families (i.e., one family member diagnosed with ASD; Gerdtts et al., 2013). Thus, interests appear to fall at the intersection of personality traits of the BAP and ASD symptoms and so may provide a unique avenue for examining factors associated with genetic risk for ASD.

Sharing interests with others can promote social interaction in adolescents with ASD, though it can be challenging to disengage from interests to participate in back-and-forth conversations (Boyd et al., 2007; Gunn & Delafield-Butt, 2016; Winter-Messiers et al., 2007). Furthermore, interests can be a source of pride and self-esteem, and can also serve as a source of comfort and distraction during stressful times and have been shown to reduce anxiety (Lidstone et al., 2014; Mercier et al., 2000; Stratis & Lecavalier, 2013; Winter-Messiers et al., 2007). Understanding the nature of interests in ASD early in development may help identify how to leverage strengths in these interests. Several questionnaires, interviews, and tasks have been developed to assess various types of interests in ASD, though most focus on the types of interests that cause social or family impairment. One of the most common interviews for ASD, the Autism Diagnostic Interview, Revised (ADI-R; Lord et al., 1994), has separate questions assessing unusual preoccupations

(i.e., interest that is odd or unusual in quality) and circumscribed interests (i.e., interests that are unusual in their intensity or nonsocial quality). Historically, the ADI-R did not assess circumscribed interests in children under-age 3 years of age, noting that these interests are more common in children 5 years of age and older. The updated Toddler ADI assesses both unusual preoccupations and circumscribed interests domains for toddlers, with algorithms that include unusual preoccupations as being helpful in differentiating ASD for toddlers with words or phrase speech¹ (Kim et al., 2013). Other measures have also been developed to assess the range of RRBs in early childhood (i.e., Repetitive Behavior Scale for Early Childhood; Sifre et al., 2021; Wolff et al., 2016). However, no measure has been developed for assessing interests broadly in young children with ASD.

To date, no study has prospectively investigated differences in interests in children who go on to develop ASD compared with those who do not or examined differences in interests based on the familial increased likelihood of ASD. Furthermore, it remains unknown whether interests are associated with other cognitive and behavioral outcomes, and whether they differ in children who do and do not go on to develop ASD. Further investigation into different aspects of interests and whether they are unique to ASD or follow genetic liability for ASD is warranted.

In this study, we developed a semi-structured parent interview that broadly asks about a child's interests, with follow-up questions to characterize aspects of interests that have been hypothesized to differ in ASD (intensity and peculiarity¹). We administered this interview to a sample of children with higher likelihood of developing ASD by virtue of having an older sibling with ASD (i.e., high-risk; HR²), as well as a comparison sample of children at low risk for ASD (i.e., low-risk; LR). We followed both groups of children from 6 to 24 months of age and conducted a developmental/clinical evaluation for ASD at 24 months of age. Based on results of this evaluation, we categorized participants into three groups: LR; HR without ASD (HR-Neg) and HR with ASD (HR-ASD).

The first aim of our study was to validate the Intense Interests Interview (I-3) within a sample at a higher likelihood for ASD and characterize the nature of interests. For the second aim of our study, we sought to replicate the sex differences in intensity of interest identified by DeLoache et al. (2007) in each outcome group,

hypothesizing that males would demonstrate more intense interests at 18 and 24 months compared with females. Our third aim was to evaluate differences in intensity and peculiarity of interests at 18 and 24 months of age by familial risk and diagnostic outcomes (e.g., HR-ASD vs. HR-negative vs. LR). We hypothesized that the intensity of a child's primary interest would not differentiate ASD-negative from ASD-positive children (Lord et al., 1994) but that the rating of peculiarity would differentiate the groups. We also examined correlates of intense interests in each group, hypothesizing that parent-reported intensity of interests would be associated with elevated developmental level (Smith et al., 2009).

METHODS

Procedures

This study took place in the context of an ongoing longitudinal study of brain and behavioral development, prospectively examining infants at high-familial-risk and low-familial-risk for ASD. The data were collected across four clinical research sites, including the University of North Carolina—Chapel Hill, Washington University in St. Louis, University of Washington in Seattle, and the Children's Hospital of Philadelphia.

Participants

Infants were enrolled as high-risk if they had an older biological sibling with an existing clinical diagnosis of ASD. The community clinical diagnosis was corroborated by scores on the Social Communication Questionnaire (SCQ; Berument et al., 1999) and autism diagnostic interview—revised (ADI-R; Lord et al., 1994). The ADI-R was administered by research-reliable staff. High-risk infants were primarily enrolled at 6 months, with a small proportion enrolled at 12 months. Low-risk children were enrolled at 6 months of age if they had an older sibling who scored below the SCQ threshold for ASD and if they had no first- or second-degree relatives with ASD or intellectual disability according to parent report. Behavioral assessments were conducted at 6, 12, and 24 months. Parents also completed questionnaires and participated in several clinical interviews over the phone when their child was 18 months of age. Exclusion criteria and the number of participants excluded are listed in Supplemental Information (SI).

The sample was selected to include all children with complete I-3 data (described in detail below) at a minimum of one-time point. This form was completed at 18 and 24 months of age. This selection criterion yielded a sample of 375 participants.

At 24 months, toddlers were evaluated to determine whether they met the criteria for ASD. If children met

¹We acknowledge that the term *peculiarity* defines differences in acceptability of interests relative to the neurotypical perspective. We chose to keep this term as indicating that the interest seems different than other children, but do not mean to imply these interests are not acceptable.

²We acknowledge there are differing opinions in the field on how to refer to the younger siblings of children with ASD, with no clear evidence suggesting whether using language of risk or likelihood is preferred (Fletcher-Watson et al., 2016), and differing preferences from varied stakeholders. Given this lack of consensus, we have retained the convention of using group labels of "high-risk" and "low-risk."

diagnostic criteria for autism or pervasive developmental disorder (PDD) using DSM-IV-TR criteria based on expert clinical opinion, they were classified as having ASD (HR-ASD). Children who had an older sibling with ASD, but who did not meet the criteria were classified as high-risk negative (HR-negative). Children with TD siblings who did not meet the criteria for ASD were classified as low-risk (LR). Our final sample included 352 participants with intense interest data, including 109 LR toddlers, 187 HR-negative, and 56 HR-ASD toddlers. See Table 1 for additional information on the sample characteristics.

Measures

The I-3 builds on previous studies assessing intense interests in young children (DeLoache et al., 2007) and characterizing developmental features of interests as they relate to ASD (Bodfish et al., 2000; Lam et al., 2008; Miranda et al., 2010; Turner-Brown et al., 2011). The I-3 was completed in a semi-structured interview format, such that the clinician asks the parent to list his or her child's primary interests (i.e., "Are there objects or activities that your child prefers to others, or shows an intense interest in? By interest, I mean what objects or activities does s/he seek out and spend time holding onto, talking about, or playing with?"). Parents were able to report up to seven interests. For each interest reported and described, the clinical interviewer queried the parent as to the age of onset and asked questions to determine intensity and peculiarity ratings. Specifically, the examiner asked how distressed the child is if the interest/activity is not available (intensity), and whether others have noticed or commented on it (peculiarity).

Following the administration of the I-3, the clinical interviewer rated the level of intensity on a 1–5 scale with the following probe: When the child is prevented from the interest or the interest is blocked, can the child be

redirected? The intensity rating was based on the following key: 1 = easily; 2 = with mild distress, but can redirect him/herself; 3 = with moderate distress, child needs help to redirect; 4 = with severe distress, child requires intense help to redirect; 5 = with severe distress, child requires intense help to redirect and may perseverate for long periods of time. The following probe anchored the clinical interviewer's rating of peculiarity: How unusual or peculiar is the interest? Is it noticeable by others? Is it embarrassing to the parent? The peculiarity ratings included: 1 = not peculiar, interest is developmentally appropriate; 2 = mildly unusual, only obvious to people who know the child well or watch the child carefully; 3 = moderately unusual, noticeably unusual; 4 = severe, clearly unusual, or peculiar, but not present in more than one setting; 5 = extreme, clearly unusual or peculiar and occurs often in more than one setting. Research staff was trained on the measure over regular meetings. Staff from different sites were given instructions and protocol documentation and allowed to ask questions of the measure author.

Variables of interest on the I-3 included the clinician's assessment of intensity of the child's most intense interest, clinician's assessment of peculiarity of their most peculiar interest, as well as the age of onset of the most intense interest, and the total number of interests. The I-3 was conducted as part of a phone call visit when the child was 18 months-old, and in-person during the 24-month behavioral visit.

We developed a coding scheme to characterize the interests parents reported their children exhibiting. Two independent coders who were blind to the participant's risk or diagnostic status rated the child's most intense interest at each time point. Categories included balls, vehicles, dolls, books, electronic objects/digital media, domestic pretend play, construction toys, outdoor or active activities, stuffed animals, crafts, live animals, explicitly social activities, idiosyncratic (unusual or repetitive), and miscellaneous. Raters showed good reliability,

TABLE 1 Participant characterization by group

	LR		HR-Neg		HR-ASD	
	M (SD)	Range	M (SD)	Range	M (SD)	Range
Sex	61 M, 48 F	-	101 M, 86 F	-	45 M, 11 F	-
V18 age visit start	18.33 (0.58)	18–20	18.36 (0.72)	17–21	18.75 (1.39)	18–25
V24 age visit start	24.69 (1.07)	23–29	24.68 (1.13)	23–31	25.04 (1.85)	23–32
V12 Mullen ELC	105.43 (14.14)	51–134	100.82 (12.82)	64–142	92.36 (15.25)	51–122
V24 Mullen ELC	110.89 (17.04)	49–146	101.39 (16.05)	66–139	81.11 (17.89)	49–134
V24 vineland ABC	104.02 (8.55)	62–131	100.27 (8.94)	75–152	88.33 (10.68)	58–107
V24 RBS-R total score	1.59 (2.37)	0–16	4.24 (6.61)	0–34	10.4 (10.33)	1–48
V24 ADOS social affect CSS	1.87 (1.28)	1–8	1.81 (1.03)	1–6	5.69 (1.99)	2–10
V24 ADOS RRB CSS	2.44 (2.07)	1–7	2.97 (2.31)	1–8	6.33 (2.14)	1–10

Abbreviations: HR-Neg, high-risk individuals without autism spectrum disorder diagnosis; HR-ASD, high-risk individuals with autism spectrum disorder; LR, low-risk individuals.

with kappa values of 0.86 at 18 months and 0.84 at 24 months. Raters met to consensus code disagreements after final coding was complete.

The *Mullen Scales of Early Learning* (Mullen, 1995) is a measure of cognitive and motor development standardized for children between birth and 68 months of age and was collected at 12 and 24 months of age. The direct behavioral assessment targeted skills and abilities in five domains including gross motor, fine motor, visual reception, receptive language, and expressive language. The measure yielded subdomain *T*-scores and age-equivalent scores for each subdomain. The Mullen early learning composite (ELC) score was of interest in this study and reflected overall developmental level and represents an aggregate score of all subdomains except for gross motor.

The *Autism Diagnostic Observation Schedule* (ADOS; Lord et al., 2000) is a semi-structured direct behavioral assessment of communication, social interaction, play skills, and restricted and repetitive behavior. This procedure was administered and scored at the 24-month assessment by research-reliable examiners. Module 1 was administered to the vast majority of children, while module 2 was administered to a minority of participants when appropriate given a child's language level. Cross-site reliability was initially calibrated through an in-person training meeting and maintained over the course of the study through bi-monthly video-conference calls, where clinicians calibrated to at least 80% agreement on ADOS codes.

The *Vineland Adaptive Behavior Scales-II* (Vineland-II; Sparrow et al., 2005) was administered at 6, 12, 18, and 24 months of age and was designed to capture functional adaptive behavior through a semi-structured parent interview. It was standardized and norm-referenced for use from birth through adulthood. We extracted the Adaptive Behavior Composite (ABC) standard score as part of the characterization of the sample.

The *Repetitive Behavior Scale—Revised* (RBS-R; Bodfish et al., 2000) is a 43-item parent report questionnaire measuring a variety of repetitive behaviors. Items are rated on a four-point Likert-scale (rating frequency of occurrence) and distributed along with six subscales: Stereotyped Behavior, Self-Injurious Behavior, Compulsive Behavior, Ritualistic Behavior, Sameness Behavior, and Restricted Behavior. The RBS-R was administered at the 24-month visit.

Analytic strategy

The intensity and peculiarity rating for any given interest can be conceptualized as a strictly nominal variable (e.g., absolute presence versus absence of an intense or a peculiar interest as determined by the clinical interviewer), in line with DeLoache et al. (2007). Alternatively, the intensity and peculiarity rating for any given interest can also be conceptualized as an ordinal variable that approximates an underlying dimension of clinical relevance. To determine an appropriate statistical approach, we investigated the skew and kurtosis of the intensity and peculiarity of interests in the full sample at both 18- and 24-months (see Table 2 and Figure 1). Variables were treated as normally distributed if skew values fell between -1 and 1 , and kurtosis fell between -3 and 3 . In our full sample, as well as each group separately, the intensity of interests showed acceptable skew and kurtosis values. Thus, we treated this variable as a normally distributed ordinal variable. However, peculiarity of interests demonstrated unacceptably high skew and kurtosis. Visual inspection revealed that most children's interests were rated as having low peculiarity, with few children being rated as having peculiar interests. Thus, we treated peculiarity as dichotomous, coding a value of 1 as non-peculiar, with values 2–5 as peculiar. All analyses examining interest peculiarity

TABLE 2 Summary statistics of intensity and peculiarity of interests by age and diagnostic outcome group

		LR (N ₁₈ = 76; N ₂₄ = 74)		HR-Neg (N ₁₈ = 128; N ₂₄ = 118)		HR-ASD (N ₁₈ = 37; N ₂₄ = 42)		Full sample ^a	
		M (SD)	Range	M (SD)	Range	M (SD)	Range	Skew	Kurtosis
18 months	Interest Intensity	2.22 (1.01)	1–5	2.29 (1.04)	1–5	2.38 (1.11)	1–5	0.23	–0.85
	Interest peculiarity	1.10 (0.35)	1–3	1.29 (0.74)	1–5	1.42 (0.73)	1–4	3.20	11.71
	Age of onset	12.28 (3.31)	1–18	12.80 (3.20)	1–20	12.69 (3.20)	3–18	–0.70	1.03
	Number of interests	3.47 (1.47)	1–7	3.12 (1.43)	1–7	3.18 (1.39)	1–7	0.32	–0.20
24 months	Interest intensity	2.26 (0.93)	1–4	2.67 (1.14)	1–5	2.98 (1.12)	1–5	0.13	–0.72
	Interest peculiarity	1.18 (0.48)	1–3	1.36 (0.76)	1–5	1.90 (1.36)	1–5	2.41	5.51
	Age of onset	15.74 (5.64)	3–24	14.83 (5.62)	1–25	14.97 (5.59)	4–27	–0.21	–0.74
	Number of interests	3.09 (1.47)	1–7	3.17 (1.58)	1–7	3.41 (1.66)	1–7	0.38	–0.41

Abbreviations: HR-Neg, high-risk individuals without autism spectrum disorder diagnosis, HR-ASD, high-risk individuals with autism spectrum disorder; LR, low-risk individuals.

^aSee supplementary information for discussion of instances of missing data for the 1-3.

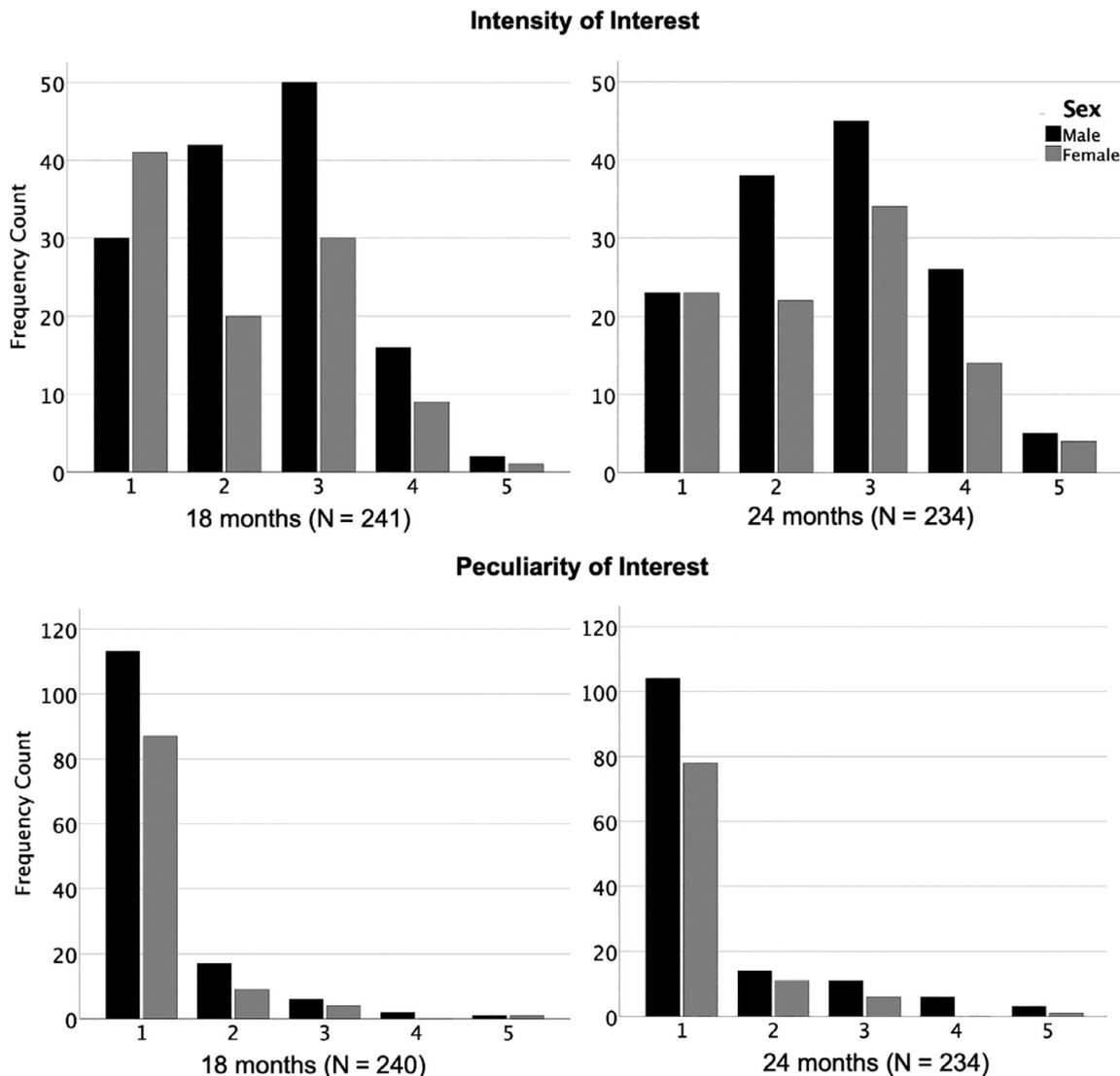


FIGURE 1 Frequency count of intensity and peculiarity of interest levels by sex in the full sample

were also run using non-parametric tests and results remained the same.

Our first aim validated this new measure by evaluating associations with other assessments of repetitive interests on the RBS-R and ADOS. We also characterized intensity, peculiarity, and category of interests in each diagnostic group. For the second aim focused on sex differences, we examined the frequency/proportion and distribution of interests according to their intensity rating to determine whether the intensity of the interests reported varies according to the sex of the child. We used independent samples *t* tests when examining interest intensity and chi-square tests for interest peculiarity. For our third aim, we examined the clinical utility of the intensity and peculiarity of interests reported within the familial risk design. We used logistic regression analyses to determine whether intensity and/or peculiarity of interests differentiated diagnostic groups (HR-ASD, HR-negative, LR), comparing all combinations of the three groups. We then extended

these analyses to examine correlations between intensity and peculiarity of interests at 18 and 24 months of age and measures of developmental functioning (VABS). We used Pearson correlation values for interest intensity and independent-sample *t* tests for interest peculiarity.

RESULTS

Aim 1

To establish convergent validity of the I-3, we examined whether intensity and peculiarity of interests were associated with other measures of ASD symptoms (Table 3). Intensity and peculiarity of interests at both 18 and 24 months were both associated with the RBS-R total scores across the full sample. Similarly, intensity of interests at 18 and 24 months were associated with RBS-R scores in the restricted behavior domain across the full

sample. Peculiarity of interests at 24, but not 18 months was associated with RBS-R restricted behavior score. ADOS RRB calibrated severity scores (CSS) were also associated with 24-month intensity of interests and 18-month peculiarity of interests. ADOS Social Affect CSS scores were not associated with intensity or peculiarity of interests at either age, $ps > 0.05$.

Means and *SD* of intensity and peculiarity of interests by diagnostic group are presented in Table 2. Interests spanned the range of intensity values, with 45% of the sample showing moderate to severe intensity (values of three or more) at 18 months, and 55% at 24 months. In the HR-ASD sample, 51% showed moderate to the severe intensity at 18 months, and 73.8% at 24 months. A smaller proportion of our sample demonstrated peculiar interests, with 17% endorsing at least mildly unusual interests (values of two or more) at 18 months, and 22% at 24 months. In the HR-ASD

group, 31% at 18 months and 38% at 24 months exhibited peculiar interests. Examples of peculiar interests included vacuums, armpits, cat poop, as well as more mundane interests that had an unusual quality (e.g., an interest of hats with a note that the child insisted he wear a hat when leaving the house). There was the representation of peculiar and non-peculiar interests at each intensity level at both 18 and 24 months, demonstrating the distinguishability of these constructs, see Figure 2. Children's interests varied in content, represented in Figure 3.

Aim 2

Sex differences in mean values of intensity and peculiarity of interests are presented in Table 4, and are visually depicted in Figure 1. In the LR group only, males demonstrated more intense interests than females at

TABLE 3 Convergent validity demonstrating associations between measures of RRBs and intensity and peculiarity of interests

		24 months		
		RBS-R Total Score	RBS-R Restricted Behavior Score	ADOS RRB CSS
18 months	Intensity (correlation)	0.22**	0.19*	0.12
	Peculiarity (<i>t</i> test value)	-2.41*	-1.83	-2.48*
	Number of interests (correlation)	-0.07	-0.09	-0.37
	Age of onset (correlation)	0.01	0.01	-0.03
24 months	Intensity (correlation)	0.30**	0.26**	0.15*
	Peculiarity (<i>t</i> test value)	-4.00**	-3.73**	-1.53
	Number of interests (correlation)	-0.03	-0.07	0.00
	Age of onset (correlation)	-0.17*	-0.17*	0.06

Abbreviations: ADOS RRB CSS, Autism Diagnostic Observation Schedule, restricted and repetitive behavior calibrated severity score; RBS-R, Repetitive Behaviors Scale-Revised.

* $p < 0.05$;

** $p < 0.01$.

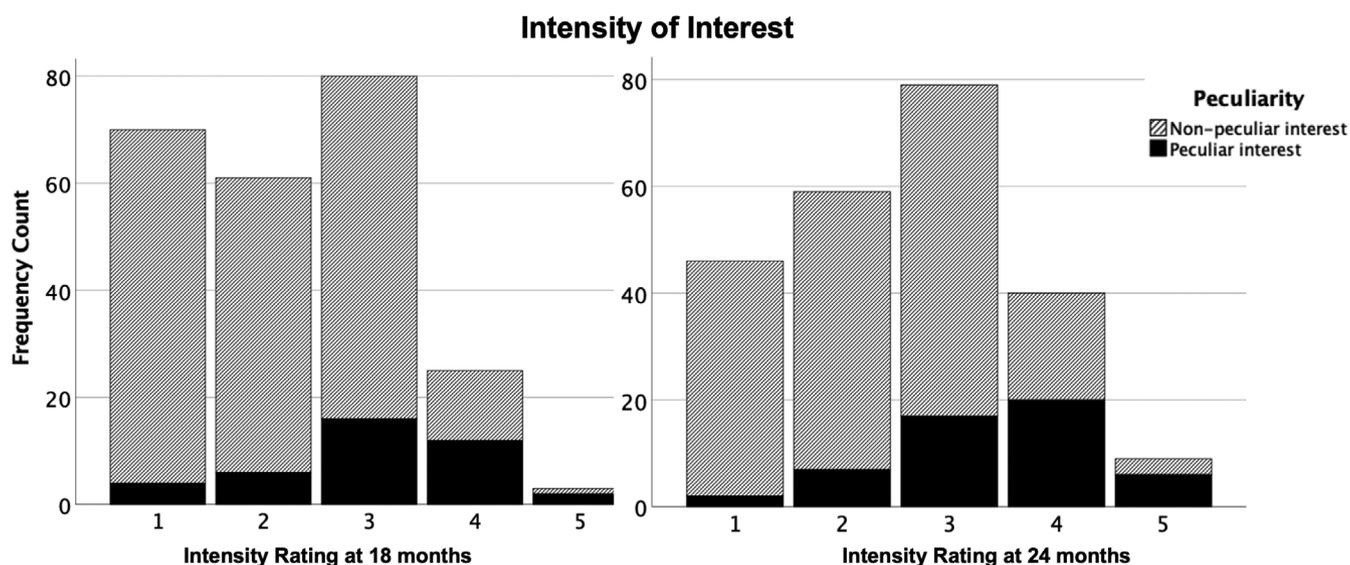
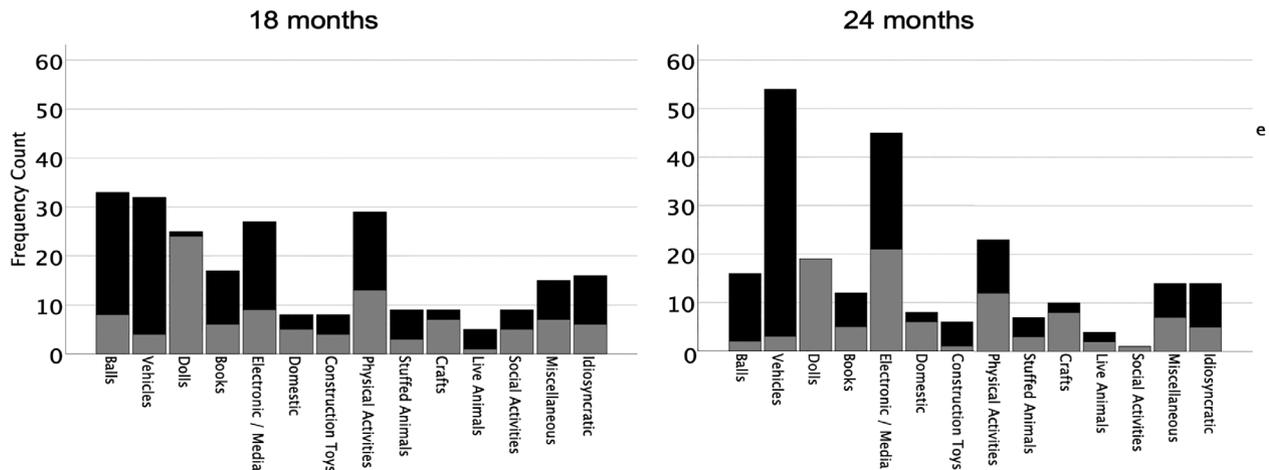


FIGURE 2 Frequency count of the distribution of peculiar interests by intensity of interest at 18 and 24 months

Category of Most Intense Interest by Sex



Category of Most Intense Interest by Group

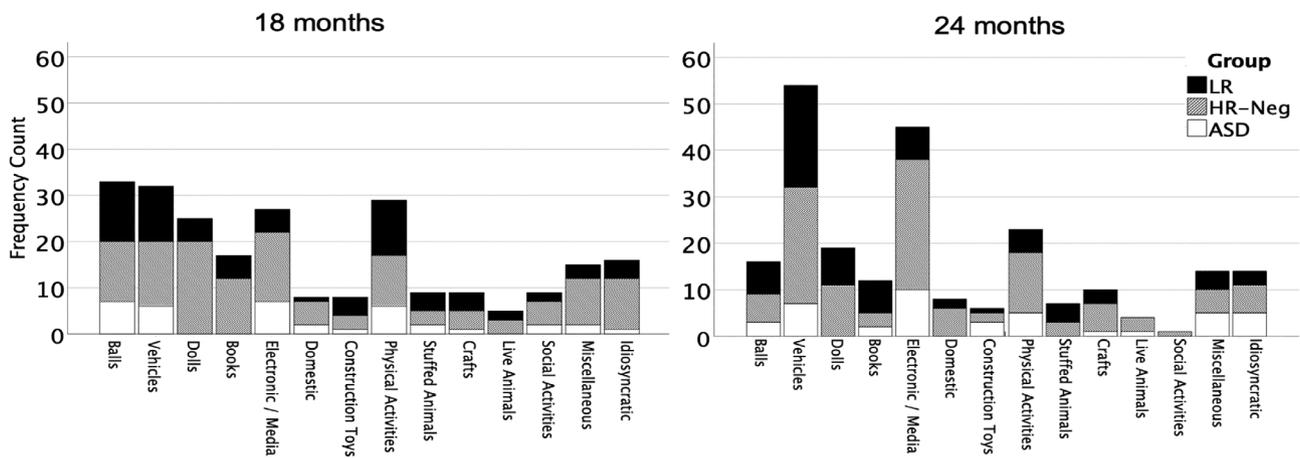


FIGURE 3 Interest categories by sex and diagnostic group at 18 and 24 months of age

18 months of age, but no differences emerged in interest peculiarity, number of interests, or age of onset. At 24 months, there were no sex differences in intensity, peculiarity, or age of onset of interest. In the HR-neg group, there were no sex differences in intensity or peculiarity of interests, number of interests, or age of onset at 18 or 24 months. Finally, we also examined sex differences in the HR-ASD group. Similar to the HR-Negative group, no sex differences emerged in the HR-ASD group in terms of intensity or peculiarity of interests, the number of interests, or age of onset at 18 or 24 months.

Aim 3

Diagnostic group differences

For our third aim, we examined whether intensity and peculiarity of interests at 18 and 24 months were

related to group membership at 24 months of age. Logistic regressions for each predictor on all group comparisons are presented in Table 5. Logistic regression results revealed that 18-month peculiarity of interests as well as 24-month intensity and peculiarity of interests differentiated HR-ASD from LR toddlers. The HR-ASD group also differed from the HR-Neg group in interest peculiarity at 24-months. Lastly, 24-month interest intensity differentiated HR-Neg from LR toddlers.

Correlates of interests

We next evaluated the correlates of intense interests in each group (Table 6). Few associations emerged in the LR group. Intensity of interests at 24 months was positively associated with 12-month Mullen ELC. No other significant correlations emerged.

TABLE 4 Sex differences in interest intensity, peculiarity, number of interests, and age of onset of most intense interest

	LR			HR-Neg			HR-ASD		
	M (SD) male	M (SD) female	Test value	M (SD) male	M (SD) female	Test value	M (SD) male	M (SD) female	Test value
18 months									
Intensity ^a	2.48 (0.99)	1.94 (0.98)	2.34*	2.39 (0.93)	2.17 (1.15)	1.21	2.39 (1.17)	2.33 (0.82)	0.11
Peculiarity ^b	13%	6%	1.09	17%	17%	0.004	33%	31%	0.03
Age of Onset ^a	12.23 (2.68)	12.33 (3.93)	-0.13	12.61 (3.06)	13.02 (3.36)	-0.71	12.66 (3.38)	12.83 (2.32)	-0.12
Number of Interests ^a	3.33 (1.52)	3.18 (1.79)	0.16	3.01 (1.58)	2.81 (1.59)	1.29	2.94 (1.50)	3.5 (1.22)	-0.75
24 months									
Intensity ^a	2.41 (0.89)	2.06 (0.95)	1.63	2.72 (1.15)	2.62 (1.15)	0.48	2.81 (1.15)	3.5 (0.85)	-1.74
Peculiarity ^b	14%	13%	0.05	22%	22%	0.02	44%	20%	1.82
Age of Onset ^a	14.68 (5.57)	17.03 (5.53)	-1.75	14.13 (5.32)	15.62 (5.89)	-1.41	15.1 (5.37)	14.5 (6.68)	0.27
Number of Interests ^a	2.37 (1.52)	3.14 (1.90)	-2.29	2.94 (1.72)	2.82 (1.83)	0.31	3.11 (1.81)	3.2 (2.15)	0.47

Note: For peculiarity of interests, the proportion of children demonstrating a behavior rated as peculiar is reported. For the age of onset of interest, values are reported in months of age.

Abbreviations: HR-Neg, high-risk individuals without autism spectrum disorder diagnosis; HR-ASD, high-risk individuals with autism spectrum disorder; LR, low-risk individuals.

^aResults of *t* test analyses.

^bResults of chi-square analyses.

**p* < 0.05.

In the HR- negative group, intensity of interests at 24 months, as well as peculiarity of interests at 18 and 24 months were negatively associated with 24-month Vineland ABC score, such that higher intensity of interests and having peculiar interests were associated with lower Vineland ABC scores. Peculiarity of interests at 18 months was negatively associated with Mullen ELC at 12 months of age, such that having peculiar interests was associated with lower Mullen ELC scores. No other associations were observed between intensity or peculiarity of interests and adaptive or developmental measures.

For HR-ASD children, intensity of interests at 18 months was negatively associated with an adaptive level on the Vineland ABC at 24 months. Peculiarity of interests was associated with lower adaptive skills at 24 months. No other associations were observed between the intensity of interests or peculiarity of interests with adaptive or developmental measures.

DISCUSSION

This was the first study to comprehensively characterize interests in a large sample of children at the high and low likelihood of developing ASD before the age of diagnosis. We developed a new measure to interrogate interests in young children and characterize both the intensity and peculiarity of interests, which bridges the study of intense interests in typical development and repetitive interests in ASD. We partially replicated previous studies showing early sex differences in intensity of interests (DeLoache et al., 2007), with low-risk males demonstrating more intense interests than females at 18 months. We also found that intensity of interest differed by familial risk at 24 months, while peculiarity of interest differentiated ASD from non-ASD participants at 24 months, highlighting the importance of characterizing interests in young children.

In our LR sample, we replicated DeLoache et al. (2007)'s finding of higher rates of intense interests in males compared with females at 18 months, though the effect size of this difference was rather modest. We did not detect a similar sex difference at 24 months, though males were reported to have an earlier onset of interests. This highlights the importance of considering development in examining intense interests; sex differences in intensity of interest may vary as children age. The research on sex differences in ASD has been mixed (Kaat et al., 2021; Van Wijngaarden-Cremers et al., 2014). Some studies have identified similar sex differences in core ASD symptoms in ASD samples as in comparison samples (Messinger et al., 2015), while others did not observe differences in early childhood (Bölte et al., 2011; Van Wijngaarden-Cremers et al., 2014). We did not find sex differences in our sample at high familial risk for ASD, indicating a different process might be occurring in the HR sample. Given our finding of

TABLE 5 Results of logistic regressions evaluating how well intensity and peculiarity of interests differentiate HR-ASD, HR-negative, and LR groups

	18 months						24 months					
	Intensity			Peculiarity			Intensity			Peculiarity		
	β	p	OR	β	p	OR	β	p	OR	β	p	OR
HR-ASD vs. HR-Neg	0.08	0.65	1.08	-0.75	0.08	2.12	0.24	0.14	1.27	-0.78	0.045	2.18
HR-ASD vs. LR	0.14	0.46	1.15	-1.47	0.006	4.34	0.71	0.001	2.04	-1.37	0.003	3.94
HR-Neg vs. LR	0.07	0.66	1.07	-0.72	0.12	2.05	0.37	0.01	1.44	-0.59	0.15	1.81

Note: Bolded results reflect significant differences.

Abbreviations: HR-ASD, high-risk individuals with autism spectrum disorder; HR-Neg, high-risk individuals without autism spectrum disorder diagnosis; LR, low-risk individuals; OR, odds ratio.

TABLE 6 Associations between intensity and peculiarity of interests in the LR, HR-Neg, and HR-ASD samples

	Intensity of Interest ^a		Peculiarity of Interest ^b	
	18 months	24 months	18 months	24 months
LR				
12 month Mullen ELC	0.13 ($p = 0.26$)	0.31 ($p = 0.01$)	1.44 ($p = 0.15$)	-1.54 ($p = 0.13$)
24 month Mullen ELC	0.05 ($p = 0.65$)	0.07 ($p = 0.58$)	0.18 ($p = 0.86$)	-0.78 ($p = 0.44$)
24 month Vineland ABC	0.12 ($p = 0.34$)	0.02 ($p = 0.88$)	0.74 ($p = 0.46$)	-1.07 ($p = 0.29$)
HR-Neg				
12 month Mullen ELC	-0.19 ($p = 0.18$)	-0.16 ($p = 0.10$)	3.23 ($p = 0.002$)	1.70 ($p = 0.09$)
24 month Mullen ELC	-0.17 ($p = 0.08$)	-0.10 ($p = 0.27$)	1.21 ($p = 0.23$)	1.21 ($p = 0.23$)
24 month Vineland ABC	-0.09 ($p = 0.33$)	-0.21* ($p = 0.02$)	2.20 ($p = 0.03$)	2.73 ($p = 0.007$)
HR-ASD				
12 month Mullen ELC	0.13 ($p = 0.45$)	0.32 ($p = 0.07$)	-0.09 ($p = 0.93$)	-0.30 ($p = 0.76$)
24 month Mullen ELC	-0.12 ($p = 0.47$)	-0.08 ($p = 0.60$)	0.19 ($p = 0.81$)	1.03 ($p = 0.31$)
24 month Vineland ABC	-0.39* ($p = 0.02$)	-0.12 ($p = 0.49$)	-0.60 ($p = 0.55$)	3.78 ($p = 0.001$)

Abbreviations: HR-ASD, high-risk individuals with autism spectrum disorder; HR-Neg, high-risk individuals without autism spectrum disorder diagnosis; LR, low-risk individuals; Mullen ELC, Mullen early learning composite; Vineland ABC, Vineland adaptive behavior composite.

^aResults of correlation analyses, Pearson's r , p -values presented in parentheses.

^bResults of t test analyses, p -values presented in parentheses.

* $p < 0.05$.

increased intensity of interests in HR children, it may be that intensity of interests is related to familial risk for ASD, as we did not find sex differences in our high-risk sample. Future research may benefit from extending examinations of sex differences to later ages as well as other clinical groups that have genetic associations with ASD (e.g., attention-deficit/hyperactivity disorder; Ghirardi et al., 2018).

Previous studies have speculated that intense interests may serve as conduits for exploration of specific topics/themes/activities and thus would be associated with developmental level (Chi & Koeske, 1983; Johnson et al., 2004; Johnson & Mervis, 1994). Our sample spanned a wide range of topics of interest, both across sex and diagnostic group. In the LR sample, the developmental level was positively related to later intensity of interests. However, in the HR-Neg group we found that

having intense and peculiar interests was associated with lower developmental and adaptive functioning. It may be that intense focus on a specific interest during early childhood has the potential for narrowing a child's diversity of social and emotional experiences, which in turn can have cascading effects on experience-dependent adaptive behavioral development. Alternatively, having lower developmental level may constrain a child's repertoire of interest but not limit the intensity of interests. It could also be that both lower developmental functioning and peculiar interests are both markers of ASD, and we are simply identifying features related to the diagnosis. This does highlight the possibility that measures like the I-3 may be useful for identifying the subset of HR toddlers who have a degree of intensity or peculiarity associated with their interests that may constrain the development of a wider range of play and social-communication skills.

Further, this subset of HR toddlers may be more likely to benefit from intervention approaches that incorporate and expand a child's interests in a way that promotes broader developmental gains (Boyd et al., 2007; Koegel et al., 2012).

Consistent with our hypotheses, intensity of interests differentiated familial risk for ASD (HR-ASD = HR-Neg > LR), while peculiarity of interests differentiated clinical outcomes in a disorder specific manner (HR-ASD > HR-Neg = LR). Having highly intense interests may be an early-emerging feature of the broader autism phenotype, as both high-risk sibling groups demonstrated more intense interests than the low-risk group. This is consistent with the monotropic view of ASD (Murray et al., 2005), where intensity of interest is hypothesized to vary across individuals, with relatives of individuals with ASD potentially showing higher levels of attentional focus. Having early *unusual* or *peculiar* interests seemed to differentiate which high-risk siblings would go on to develop ASD. Initial conceptualizations of intense interests proposed that intense interests would be more applicable to older, verbally-fluent children (Lord et al., 1994). In our sample, many young children who did and did not go onto develop ASD were described as having intense interests. Contrary to the perception that intense interests are not relevant in young children with ASD, we found that many young children exhibited these interests. The peculiarity of these interests, even mildly peculiar interests, seemed to best differentiate ASD from both ASD-negative groups, consistent with prior ADI-R studies in toddlers which found that unusual preoccupations are better indicators of ASD than overly intense interests (Kim et al., 2013).

Many terms have been applied to the multifaceted interests in ASD—circumscribed, restricted, repetitive are often used interchangeably which diminishes their specificity. Our open-ended questions surrounding interests resulted in parents reporting a range of topics (i.e., what a child tends to talk about), types of play items and activities (i.e., how a child plays, what items they tend to play with) that constituted the form or content of their child's specific interest (e.g., Legos, dinosaurs, babies, holding bouncy balls in his hand). We then systematically operationalized these responses in terms of intensity and peculiarity of interests, which showed familial risk and diagnostic differences. For very young children whose language level may limit their ability to talk about their play, focusing on activities or items of interests may be more clinically relevant than measures that assess specific topics or subjects of interest. Questions about topics of intense interest are asked on the ADI-R, but assessing interests in this way may limit the possibility of identifying developmentally-younger forms of intense interest that do not rely on expressive communication skills (e.g., repetitively manipulating the same item or seeking out one specific ball to play with). It is very difficult to differentiate different features of RRBs in young

children, and distinguish general interests from circumscribed or preoccupations that would be considered a symptom of ASD. For young children who have not yet acquired the skills to communicate about a specific topic, it may be more valid to assess their interests by asking about favorite items and activities. In the present study, the I-3 cued parents to report aspects of the child's interest that related to repetitive play items or activities (e.g., repetitively stacking blocks, repetitively rubbing things on their belly), as opposed to only assessing repetitive topics of conversation. Using more open-ended questions that focus on items, activities, and behaviors related to interests may be critical for generating diagnostically-relevant information related to intense interests and for identifying avenues to promote engagement via intervention in toddlers with developmental delays. By separating the assessment of interests as reported by the parent from the rating of the potential intensity and peculiarity of the reported interests, the I-3 also provides a method for operationalizing "restricted interests" and importantly avoids the pitfalls associated with simply labeling all interests that occur in the context of ASD as being pathological.

Limitations and future directions

The results of this study should be considered in light of its limitations. First, as the measure of intense interests was collected via a single assessment at each time point, and the interviews were not recorded, we could not calculate test-retest reliability or inter-rater reliability. While we developed a clear coding scheme and demonstrated convergent and discriminant validity, further investigation is necessary to elucidate the full psychometric profile of the instrument. We are also limited in our assessment of intensity of interest by the behavioral manifestations of intensity, such as difficulty redirecting the child. Difficulty transitioning away from an interest did not necessarily capture the intensity of an interest but was chosen as the operational definition of intensity of interests. Since the children in our sample were quite young, and we could not assess how often they communicated about their interests, this was the best possible way to assess intensity of interests in an interview. Future studies should investigate other ways to capture intensity of interests, including other complementary or alternative assessments of interests (e.g., DeLoache et al., 2007), or eye-tracking that could index duration of fixation. Some caution is warranted in interpreting the age of onset of interests, given the retrospective nature of this instrument, particularly for parents who reported interests manifesting as early as 1 month of age.

We also had limited power to detect sex differences in the HR-ASD group, given the higher proportion of males in that group. This was particularly limited when examining interest peculiarity, which also occurred at a low base

rate. It could be that there are sex differences present in restricted interests but that we were not sufficiently powered to detect an effect. Several large-scale studies have found that girls with ASD aged 3–18 show fewer RRBs than boys with ASD matched for age and IQ, both on the ADI-R, and observed in clinic (Bölte et al., 2011; Kaat et al., 2021; Mandy et al., 2012; Park et al., 2012; Van Wijngaarden-Cremers et al., 2014). In a high-risk sample, sex differences emerged in both HR-ASD and HR-Neg groups, but were less pronounced in the HR-ASD group (Park et al., 2012). At later ages, it could be that girls are better able to camouflage, or mask their difficulties (Lai et al., 2016). Future research should examine whether there are early predictors that contribute to later camouflaging, or lower ASD symptoms in girls.

CONCLUSIONS

The development of a new interview to characterize interests in young children highlights clinical implications for future study. It appears that having peculiar interests was most strongly associated with a later ASD diagnosis, though both intensity and peculiarity of interests were associated with functional adaptive skills, for both HR-ASD and HR-Neg children. Future investigation should determine the added value of specifically interrogating the children's interests to improve early identification efforts.

ACKNOWLEDGMENTS

This study was made possible by an NIH Autism Center for Excellence (ACE) Network grant (R01 HD055741) to J. Piven; grants from Autism Speaks (#6020) and the Simons Foundation (#140209) to J. Piven; grants from NIH (R01 MH118362 and MH118362-02S1) to J. Pruett, and C. Burrows (K12-HD055887) as well as U54 Intellectual and Developmental Disabilities Research Centers HD079124 to UNC (J. Piven); HD86984 to CHOP (R. Shultz). The funders had no role in study design, data collection, analysis, data interpretation, or the writing of the report. All data reported herein are available to qualified researchers through the National Data Archive.

ETHICS STATEMENT

The research was prospectively reviewed and approved by institutional review boards (IRBs) at each site and parents provided informed consent for their children to participate in this research study.

AUTHOR CONTRIBUTIONS

Catherine A. Burrows, Jed T. Elison and Joseph Piven contributed to study design, data processing and analysis, data interpretation, and manuscript preparation. James W. Bodfish, Jason J. Wolff, Elayne P. Vollman, Melody R. Altschuler, and Lonnie Zwaigenbaum participated in

data interpretation and manuscript preparation. Kelly N. Botteron, Stephen R. Dager, Annette M. Estes, Heather C. Hazlett, and Robert T. Schultz participated in data collection and manuscript preparation. All authors read and approved the final manuscript.

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SUPPORTING INFORMATION

Additional supporting information may be found online in the Supporting Information section at the end of this article.

How to cite this article: Burrows, C. A., Bodfish, J. W., Wolff, J. J., Vollman, E. P., Altschuler, M. R., Botteron, K. N., Dager, S. R., Estes, A. M., Hazlett, H. C., Pruetz, J. R. Jr, Schultz, R. T., Zwaigenbaum, L., Piven, J., Elison, J. T., & the IBIS Network (2021). Cataloguing and characterizing interests in typically developing toddlers and toddlers who develop ASD. *Autism Research*, 14(8), 1710–1723. <https://doi.org/10.1002/aur.2543>