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Brief Report: Sex Differences in Parental Concerns for Toddlers with Autism Risk

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Abstract

Research on sex differences in autism spectrum disorder (ASD) suggests both higher prevalence and a more easily-observable presentation of core ASD symptomology in males, which may lead to sex differences in parental concerns. The current study examined whether sex and diagnosis relate to the timing, number, and types of pre-diagnosis concerns for 669 (N_{male} = 468) toddlers who screened at-risk for ASD. No sex differences in parents' concerns emerged for toddlers diagnosed with ASD; however, in the overall at-risk sample, parents of boys endorsed ASD symptoms, including restricted and repetitive behaviors, more than parents of girls. Future research should examine why sex differences in pre-diagnosis concerns emerge and how they might impact early diagnosis for at-risk boys vs. girls.

Keywords

Autism spectrum disorder; early detection; sex differences; toddlers

Most studies regarding autism spectrum disorder (ASD) symptomology are based on predominantly male samples, as ASD is four to five times more prevalent in males than females (Christensen et al. 2016). In addition to sex differences in prevalence rates, early

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Conflict of Interest

D. L. Robins and D. Fein are co-owners of M-CHAT, LLC, which receives royalties from companies that incorporate the M-CHAT(-R) into commercial products. Data in the current study are from the freely available version of the M-CHAT(-R). The remaining authors declare no conflict of interest.

Compliance with Ethical Standards

Ethical Approval

All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Informed consent

Informed consent was obtained from all individual participants included in the study.

research (Wing 1981) suggests girls may present with less severe ASD related language and social impairment than boys. Researchers have since posited that the existence of sex differences in ASD symptoms indicates a distinct female phenotype for autism (Haney 2016), which has implications for early detection and diagnosis. Specifically, applying diagnostic criteria based on the male presentation of ASD to females may result in missed and/or misdiagnosed cases of ASD in females (Ehlers and Gillberg 1993; Gillberg 1993; Gillberg and Coleman 2002; Thompson et al. 2003).

One of the most consistent findings regarding sex differences in ASD symptoms in early childhood is that boys display more restricted and repetitive behaviors (RRBs) compared to girls (Hartley and Sikora 2009; Lord et al. 1982; McLennan et al. 1993). Hartley and Sikora (2009) found that when controlling for nonverbal reasoning skills, girls had more severe communication deficits and more co-existing sleep and internalizing problems than boys. Clinicians identified that girls displayed less severe and fewer RRBs while boys presented with more non-verbal impairments, difficulty maintaining reciprocal conversation, and weaknesses in initiating friendships (Hiller et al. 2014). Overall, these findings suggest that girls with ASD may present with fewer easily observable ASD symptoms and more co-existing challenges (Hartley and Sikora 2009), which may disguise their ASD symptoms and contribute to their later diagnosis (Begeer et al. 2013; Giarelli et al. 2010).

Given research demonstrating early sex differences based on both ASD-specific diagnostic tools and clinical reports of symptoms, it is also important to consider whether sex differences emerge in parents' reports of child behavior. Many studies (Ozonoff et al. 2009; Sacrey et al. 2015), including the study by Richards and colleagues (2016) which the current study extends, demonstrate that parents' pre-diagnosis concerns are good predictors of later ASD diagnoses; however, few studies have examined sex differences in parents' concerns. Given the lower prevalence and more subtle clinical presentation of ASD in girls, there is potential for differences in concerns reported for boys and girls. Horovitz and colleagues (2012) found that the age of first concerns was earlier for females, whereas Hiller and colleagues (2016) found no sex differences in the age of first concerns. Two studies found sex differences in the number and types of pre-diagnosis concerns parents report (Hiller et al. 2016; Little et al. 2017), in which parents of boys reported more concerns for boys with ASD, specifically related to social behavior and RRBs. Based on these studies, the concerns parents voiced about boys may be more easily observable and intuitively related to ASD than the concerns voiced for girls; however, it is important to also consider the limitations of these previous studies. Using a broad age range for children (as in Little et al. 2017) may lead to findings of sex differences that have emerged in later development but that might not be apparent at younger ages. Using retrospective reports of pre-diagnosis concerns (as in Hiller et al. 2016) may be less accurate than contemporaneous reports, in part because parents' recall might be influenced by knowledge of their child's subsequent diagnosis.

In examining sex differences in ASD populations, it is important to note that research findings are not always consistent, such as the discrepancy in findings regarding sex differences in the age of first concerns (Hiller et al. 2016; Horovitz et al. 2012). Additional studies using toddler-aged and community-based samples also found no sex differences in social communication, developmental profiles, or ASD symptoms in those diagnosed with

ASD (Mussey et al. 2017; Reinhardt et al. 2015). It is possible that sex differences in specific areas emerge at different ages and that community-based samples may miss females who are not referred for evaluations because they may present with more subtle symptoms. Furthermore, several studies found that sex differences in ASD are also present in typically developing or non-ASD populations (Hull et al. 2017; Messinger et al. 2015), which may affect evaluation rates for the broader population of boys and girls who screen at-risk for ASD. These inconsistencies indicate the need for additional research to understand when and in what areas sex differences emerge in early development.

This study examined the impact of sex and diagnosis (i.e., ASD vs. non-ASD) on a) the age of first concerns, b) the number of ASD and Non-ASD concerns, and c) the specific types of concerns reported by parents prior to diagnosis in a large sample of toddlers who screened at-risk for ASD on a screening tool. Thus, this study provides additional information about whether sex differences are evident at this young age and whether parents are identifying pre-diagnosis concerns for boys and girls equally when they are at risk and when later diagnosed with ASD. Identifying sex differences in early parental concerns prior to diagnosis is important for understanding how current early detection practices may contribute to differential referral rates for evaluations between males and females. Such knowledge could provide insight into why discrepancies exist in rates and age of diagnosis between males and females (Begeer et al. 2013; Giarelli et al. 2010).

Method

Participants

Participants were recruited from studies screening children at 18- and 24-month well-child visits (Chlebowski et al. 2013; Robins et al. 2014). Children were considered at risk for ASD if they: a) screened positive on the Modified Checklist for Autism in Toddlers (-Revised) with Follow Up (M-CHAT(-R)/F; Robins et al. 1999; Robins et al. 2009), b) screened positive on the Screening Tool for Autism in Toddlers and Young Children (STAT; Stone et al. 2000), or c) physician and/or parent noted concerns for ASD. Inclusion criteria for this study were at-risk classification followed by completion of a clinical evaluation performed by a research team led by a licensed clinical psychologist or developmental-behavioral pediatrician; clinical best estimate integrated data from autism diagnostic tools and cognitive and adaptive functioning measures. Children (n = 669) were evaluated between 15 and 58 months; diagnoses included ASD (45.7%, n = 306) developmental delay (40.5%, n = 271), and no diagnosis (13.8%, n = 92). The current sample added 137 children to Richards et al.'s (2016) sample. Families with another child diagnosed with ASD were excluded from the study. See Table 1 for demographics. [Table 1]

Procedure

Prior to the evaluation, parents completed a detailed history questionnaire reporting demographics, family history, and developmental milestones. Within the history questionnaire were three open-ended prompts regarding parental concerns for the child, including "What were the first things that made you concerned about your child's development?", "How old was your child at that time?", and a section where parents could

list current concerns. Concerns were coded using a scheme adapted from Ozonoff et al. (2009) that categorizes concerns into four ASD-related concerns (speech/communication, restricted/repetitive behaviors, social skills, and named ASD, i.e., parents named autism/ASD specifically) and six Non-ASD concerns (motor, behavioral/temperament, medical/regulatory, eating/feeding, disruptive behavior, and unspecified). See Richards et al. (2016) and Donohue et al. (2017) for complete details regarding the adapted coding scheme utilized in this study. Concerns from a subsample of 30% of parents were coded by two independent research assistants trained to reliability by the senior author; they demonstrated high agreement: ICC = .888 ($p < .001$).

Results

To analyze the impact of sex and diagnosis (ASD vs. non-ASD) on a) the age of first concerns and b) the number of ASD and Non-ASD concerns reported by parents, three two-way ANOVAs were conducted with sex and diagnosis as fixed factors.

Age of first concerns and the number of concerns reported by parents are summarized in Table 2. [Table 2] There was no significant main effect of sex ($\eta_p^2 = .001$) or interaction of sex and diagnosis on age of first concerns ($\eta_p^2 < .001$); however, there was a significant main effect of diagnosis, $F(1, 391) = 5.79$, $p = .017$, $\eta_p^2 = .015$, such that the age of first concerns was younger for the non-ASD group. Regarding the number of ASD concerns, there was a significant main effect of diagnosis, $F(1, 665) = 36.66$, $p < .001$, $\eta_p^2 = .052$, such that parents reported more ASD concerns for toddlers subsequently diagnosed with ASD. There was no significant main effect of sex ($\eta_p^2 = .005$) or interaction of sex and diagnosis on number of ASD concerns. Regarding the number of non-ASD concerns, no significant findings emerged related to main effects ($\eta_p^2 < .001$) or interaction of sex and diagnosis ($\eta_p^2 = .001$).

To analyze the impact of sex and diagnosis on the specific types of concerns endorsed by parents, chi-square analyses were conducted (See Table 3). [Table 3] For the overall sample of at-risk toddlers, parents expressed at least one concern in the ASD concern category 1.46 times more often for boys than for girls, $\chi^2(1, N = 669) = 3.37$, $p = .043$, regardless of subsequent diagnosis; however no significant sex differences emerged in whether parents reported at least one non-ASD concern ($p = .460$). Analyses examining the specific types of ASD concerns indicated that parents expressed RRB concerns for boys 1.74 times more often than for girls, regardless of subsequent diagnosis, $\chi^2(1, N = 669) = 3.86$, $p = .031$. While not significant, potentially due to a small number of parents endorsing this concern (4 for girls vs. 22 for boys), it is important to note that parents of boys were 2.43 times more likely to name ASD as a concern, $\chi^2(1, N = 669) = 2.77$, $p = .069$, than parents of girls, regardless of subsequent diagnosis.

Discussion

The current study examined whether sex and diagnosis related to the timing, number, and types of parent-reported concerns in a sample of children who screened at-risk for ASD during a pediatric check-up. Regarding the effect of diagnosis, this study replicates previous

findings (Ozonoff et al. 2009; Richards et al. 2016; Sacrey et al. 2015) that parents note more ASD concerns for those toddlers later diagnosed with ASD. Additionally, parents of toddlers with ASD reported first concerns later than parents of children without ASD. One explanation for this surprising finding is that social and language concerns noted in the ASD group might emerge later than the non-ASD related motor or feeding issues reported for toddlers without ASD, which can typically be observed prior to a child's first birthday.

Based on prior research, sex differences in parental concerns within the group of toddlers subsequently diagnosed with ASD were predicted; however no such findings emerged in this study. Instead, parents reported concerns similarly among boys and girls subsequently diagnosed with ASD. The sample in this study was younger than previous studies on sex differences in ASD, suggesting that among children detected around their second birthday, sex differences may not be evident in parents' concerns. Similarly, Hiller and colleagues (2016) also did not find sex differences in the age of first concerns for children later diagnosed with ASD. These findings may indicate that the endorsed symptoms for toddlers with ASD are severe enough for parents to discern in both sexes. When boys and girls are older they may diverge more in social skill acquisition leading to sex differences in symptoms later.

Whereas no sex differences were found in the ASD subsample, there were some sex differences in the overall sample of toddlers who screened at-risk for ASD, mostly related to the types of concerns parents endorsed. Specifically, more parents reported ASD-type concerns, including RRBs, for at-risk boys compared to at-risk girls. A study by Messinger and colleagues (2015) found that sex differences in children with ASD are not ASD specific. In other words, sex differences observed in children with ASD are also found in high-risk children without ASD, as well as low-risk children. As such, the sex differences observed in this at-risk group may reflect sex differences that occur in children without ASD. Of note, although it was not statistically significant, parents also named ASD specifically as a concern almost two and a half times more often for boys, despite being aware that their child screened at-risk for a developmental delay.

Although this study found no sex differences specific to children with ASD, it is important to consider the sex differences in parents' concerns present in the overall at-risk group and the impact they might have on parents' subsequent decision to seek an evaluation for their child. Based on prior research (Messinger et al. 2015), it appears that boys with and without ASD are exhibiting more easily observable behaviors, which may be triggering parents' concerns for boys. As such, parents' concerns for boys may cause greater distress to parents and overshadow concerns for girls. This could, in turn, reduce the likelihood of parents pursuing evaluations for girls, even in cases where they may be at-risk for ASD or another developmental delay, simply because they do not present like the at-risk boys. Moreover, if parents are less likely to seek a diagnostic evaluation for girls due to fewer ASD concerns, it may contribute to why girls tend to be diagnosed with ASD later or are less likely to have a documented ASD diagnosis compared to boys (Begeer et al. 2013; Giarelli et al. 2010). A previous study (Dworzynski et al. 2012) found that girls required a greater number of additional problems related to behavior and cognitive ability to meet diagnostic criteria for ASD compared to boys, even when both sexes displayed similar autistic traits. The authors

theorized that a gender bias could have led to these discrepancies in girls meeting diagnostic criteria. Similarly, a gender bias related to pre-diagnosis concerns could influence parents' perceptions of symptom severity and decisions to seek an evaluation when girls screen at-risk for ASD.

Overall, findings from the current study suggest that sex differences in pre-diagnosis concerns may not be ASD specific. Instead, parents appear to have different concerns related to ASD for girls and boys who screen at-risk, regardless of subsequent diagnoses. Such differences in ASD concerns between the sexes could be related to sex differences present in typical and non-ASD populations or gender biases influenced by the circulation of information to parents and pediatricians that ASD is relatively uncommon in females. Future research is needed to explore sex differences and potential gender biases and the role they play in parent and clinician perceptions of ASD symptoms in girls vs. boys. Furthermore, it is important to understand whether these sex differences or gender biases related to parental concerns lead to differences in evaluation participation, pediatrician referrals, and early diagnosis of ASD and developmental delays in girls and boys.

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Table 1

Demographic characteristics of the at-risk sample

Toddler characteristics	Total (n=669)	ASD (n=306)	Non-ASD (n=363)
Mean age months (SD)	25.6 (5.2)	25.9 (5.0)	25.4 (5.4)
Female n (%)	201 (30.0)	74 (24.2)	127 (35.0)
Race n (%)			
Black	145 (21.7)	81 (26.5)	64 (17.6)
White	251 (37.5)	123 (40.2)	128 (35.3)
Other	136 (20.3)	70 (22.9)	66 (18.2)
Not reported	137 (20.5)	32 (10.4)	105 (28.9)

Parent characteristics	Mother (n=553)	Father (n=490)
Mean age years (SD)	31.99 (8.02)	35.09 (7.82)
Education level n (%)		
College degree or more	260 (38.9)	122 (18.2)
Less than college degree	308 (46.0)	191 (28.6)
Not reported	101 (15.1)	356 (53.2)

Table 2

Means for age of first concerns and number of concerns endorsed for boys and girls

	Total		ASD		Non-ASD	
Mean (*SD*)	**Male (*n*=468)**	**Female (*n*=201)**	**Male (*n*=232)**	**Female (*n*=74)**	**Male (n=236)**	**Female (n=127)**
Age first concerns (months)	13.45 (7.18)	12.69 (6.05)	14.40 (7.60)	13.92 (5.42)	12.46 (6.60)	11.99 (6.32)
Number of ASD concerns	1.19 (.80)	1.01 (.74)	1.36 (.80)	1.30 (.75)	1.02 (.76)	.84 (.672)
Number of Non-ASD concerns	.88 (.92)	.81 (.83)	.84 (.91)	.82 (.85)	.93 (.92)	.80 (.83)

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Table 3

Frequency of parental concerns for boys and girls in the at-risk sample

	Total				ASD				Non-ASD			
[*n* (%)]	**Male (*n*=468)**	**Female (*n*=201)**	**χ^2 Value**	**Odds Ratio**	**Male (*n*=232)**	**Female (*n*=74)**	**χ^2 Value**	**Odds Ratio**	**Male (*n*=236)**	**Female (*n*=127)**	**χ^2 Value**	**Odds Ratio**
ASD Concerns	385 (82.3)	153 (76.1)	3.372*	1.46	206 (88.8)	64 (86.5)	0.288	1.24	179 (75.8)	89 (70.1)	1.422	1.34
Speech/Communication	361 (77.1)	143 (71.1)	2.717	1.37	196 (84.5)	62 (83.8)	0.021	1.05	165 (69.9)	81 (63.8)	1.423	1.32
RRBs[a]	65 (13.9)	17 (8.5)	3.857*	1.74	42 (18.1)	10 (13.5)	0.838	1.41	23 (9.7)	7 (5.5)	1.952	1.85
Social	107 (22.9)	39 (19.4)	0.987	1.23	62 (26.7)	22 (29.7)	0.254	0.86	45 (19.1)	17 (13.4)	1.882	1.52
Named ASD	22 (4.7)	4 (2.0)	2.766	2.43	15 (6.5)	2 (2.7)	1.514	2.49	7 (3.0)	2 (1.6)	0.661	1.91
Non-ASD Concerns	276 (59.0)	117 (58.2)	0.034	1.03	130 (56.0)	43 (58.1)	0.098	0.92	146 (61.9)	74 (58.3)	0.447	1.16
Motor	135 (28.8)	53 (26.4)	0.427	1.13	60 (25.9)	21 (28.4)	0.183	0.88	75 (31.8)	32 (25.2)	1.721	1.38
Behavior/Temperament	46 (9.8)	17 (8.5)	0.310	1.18	30 (12.9)	8 (10.8)	0.232	1.23	16 (6.8)	9 (7.1)	0.012	0.95
Medical/Regulatory	66 (14.1)	33 (16.4)	0.598	0.84	26 (11.2)	9 (12.2)	0.051	0.91	40 (16.9)	24 (18.9)	0.216	0.88
Feeding/Eating	37 (7.9)	14 (7.0)	0.177	1.15	24 (10.3)	8 (10.8)	0.013	0.95	13 (5.5)	6 (4.7)	0.102	1.18
Disruptive Behavior	67 (14.3)	21 (10.4)	1.842	1.43	25 (10.8)	6 (8.1)	0.439	1.37	42 (17.8)	15 (11.8)	2.235	1.62
Unspecified/General	63 (13.5)	25 (12.4)	0.129	1.10	29 (12.5)	9 (12.2)	0.006	1.03	34 (14.4)	16 (12.6)	0.227	1.17
Any Concerns	429 (91.7)	182 (90.5)	0.223	1.15	220 (94.8)	68 (91.9)	0.873	1.62	209 (88.6)	112 (88.2)	0.011	1.04

[a]RRBs = restricted/repetitive behaviors

*p<.05

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