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BRIEF REPORT

Brief Report: Sex Differences in ASD Diagnosis—A Brief Report on Restricted Interests and Repetitive Behaviors

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Abstract

Previous research found repetitive and restricted behaviors (RRBs) were less predictive of Autism Spectrum Disorder (ASD) in females, indicating the diagnostic construct may not adequately describe RRB presentations in females. This mixed-methods study investigated the female presentation of RRBs, namely restricted interests, in a clinic sample of 125 participants (n = 40 female; ages 2–83 years; 75 ASD). RRB severity did not differ between sexes, t = 1.69, $p = 0.094$, though male participants scored higher on the Restricted Behavior subscale. Qualitatively, females demonstrated a narrower range of restricted interests and expressed them in a socially oriented manner compared to males. The results suggest unique quantitative and qualitative sex differences in RRB profiles that could shed light on the female ASD phenotype.

Keywords Autism spectrum disorder · Restricted interests · Sex differences · Female · Repetitive behaviors · Assessment

Currently, males are three to eight times more likely than females to receive an Autism Spectrum Disorder (ASD) diagnosis; a discrepancy for which reasons remain unclear (Centers for Disease Control and Prevention 2012; Lai et al. 2015). Recent data suggest that the male to female ratio may be closer to 1:1, with females being under-identified or showing different phenotypic expressions that are not recognized as ASD (e.g., Frazier et al. 2014). Many researchers (i.e. Attwood et al. 2006; Duvekot et al. 2016; Frazier et al. 2014) have suggested that Repetitive & Restricted Behaviors (RRBs) could serve as a potential mechanism for understanding and explaining the sex differences in ASD diagnoses. For example, Duvekot et al. (2016) investigated RRBs as a potential tool for improving differential diagnosis of ASD in female clients and found that parent-reported RRB severity and frequency was less predictive of an ASD diagnosis in females, compared to males. However, Duvekot et al. (2016) were unable to parse apart the subdomains of the RRBs, such as restricted interests (RI) or repetitive behaviors (RB).

RI have recently emerged as an area of focus within the RRB construct as a potential source of sex differences within ASD. In a study of children and adolescents, Hiller et al. (2014) reported that females demonstrated lower clinician- and teacher-reported severity of RRBs compared to males. Within the RI domain specifically, they found substantial sex differences, such that females were more likely to have "seemingly random" RI (i.e. rocks, stickers, pens) while males were more likely to have fixations with screen time (i.e. gaming, iPad, and screen technology). These behavioral reports are consistent with recent neurophysiological data demonstrating sex differences in brain connectivity with respect to Westeinde et al. (2018) reported that females (ages 9–23) demonstrated associations between RI symptoms and the right intraparietal sulcus (associated with intention interpretation) and the right orbital gyrus (typically associated with executive function), but that this connection was not replicated in males. Taken together, such results underscore the question of how females with ASD are expressing their RI and call for further, *descriptive*, investigation into the female presentation of RI.

While the aforementioned research set the stage for further RI exploration, several elements need to be addressed. First, previous studies used the DSM-IV-TR RRB scale, which has since been altered in the DSM-5 (Hiller et al. 2014). Second, previous analyses were only conducted with clinician- and teacher-report, which leaves a significant gap in terms of parent-report. Lastly, these data were collected using children and adolescents with ASD and may not capture the trajectory of RRBs throughout adulthood,

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which are reportedly unique (Esbensen et al. 2009). This paper attempts to address these research gaps by providing *descriptive* RI profiles of males and females diagnosed with ASD across age ranges, based on parent-report measures, and using DSM-5 RRB criteria when available. In addition to addressing shortcomings in previous research, this research serves clinical utility by providing clinicians and researchers with more information on the female presentation of ASD, which can assist in the assessment and treatment process. It was hypothesized that (1) total RRB severity, and specifically RI, would be lower in female than male participants, consistent with prior literature on quantitative teacher and clinician report measures, and (2) the qualitative nature of RI would differ as a function of sex.

Method

Participants

The sample included 125 participants (40 female, 85 male) with and without ASD from a clinic in Southwest Virginia (age range 2–83 years, $M = 14.3$ years, $SD = 14.4$) who sought assessment services for ASD. The racial/ethnic diversity of the sample, although not representative of the entire country, was representative of the geographic region (87% White, 4% African American, 4% Asian, 5% other). SES and parental education metrics were not available for this sample. Of the total sample, 75 participants (60%; 20 females, 55 male) received an ASD diagnosis. The ASD group had a mean age of 12.43 years, ranging from ages 2–57 ($Std = 12.33$ years). Forty-five percent of the sample were ages 2–6 years old at time of testing. Seventy-eight percent of the ASD sample were children and adolescents (< 17 years). Specific age and sex frequencies are available in Table 1. Power analyses indicated a sample size of at least 65 participants required to detect significant effects with power at 0.8 and alpha set at 0.05. This paper analyzed data from the ASD group (N = 75), for which the current sample demonstrates sufficient power.

Table 1 Frequencies of age of assessment (years) as a function of sex

	Frequency	
Age (years)	Male	Female
2	1	1
3	9	1
4	6	0
5	5	3
6	5	2
7	3	0
8	5	1
9	3	0
10	1	0
11	2	2
12	3	0
13	1	0
14	0	1
15	2	0
17	2	0
12	3	0
18	0	2
19	1	2
22	1	0
24	0	1
29	0	1
31	1	0
32	0	1
37	1	0
42	2	0
43	1	0
50	0	1
57	0	1
Total	55	20

Measures

The primary variables of interest included sex, RRB severity, and ASD diagnosis. Because of the wide age range and ability levels in this sample, we also accounted for possible confounds of age and IQ. IQ was recorded as the standard score on a measure of intelligence that was appropriate for the participant's age and developmental level (i.e., Mullen Scales of Early Learning, Wechsler Preschool and Primary Scale of Intelligence, or Wechsler Abbreviated Scale of Intelligence). All of these measures use the same standard scores ($SS = 100$) and standard deviations ($SD = 15$).

An RRB severity composite score was created using the Autism Diagnostic Interview-Revised- RRB subscale (ADI-R), Autism Diagnostic Observation Schedule, 2nd edition—RRB subscale (ADOS-2), Repetitive Behavior Scale-Revised total score (RBS-R), and Social Responsiveness Scale- 2nd edition RRB subscale (SRS-2). All of the scales used in the RRB severity composite were parent-rated forms, parent-informed and clinician rated (ADI-R), or clinician-rated (ADOS). Each score was standardized (z-score) and then summed to create a composite RRB severity score. Due to the limited number of measures that assess RI specifically, only one measure was used as a direct severity measure of RI (i.e., RBS-R restricted interests subscale, discussed below).

Autism Diagnostic Interview-Revised

The Autism Diagnostic Interview-Revised (ADI-R; Lord et al. 1994) is a semi-structured interview for caregivers of children and adults for whom autism is the referral question.

The ADI-R shows good-to-excellent reliability and validity and has been used in practice for over 15 years (Lord et al. 1994). The ADI-R items are rated by the clinician and provide three overall scores, including abnormalities in reciprocal social interaction, impairments in communication and language, and restricted, repetitive behaviors. The RRB subscale score for the ADI-R was used in creating the RRB severity composite score.

Autism Diagnostic Observation Schedule, 2nd Edition

The Autism Diagnostic Observation Schedule, 2nd edition (ADOS-2; Lord et al. 2012) is a semi-structured, standardized assessment of social interaction, communication, and imaginative play for children and adults, ages 12 months through adulthood. The ADOS-2 has varying modules administered based on verbal level and chronological age. All ADOS-2 modules (Todd/1/2/3/4/G) show excellent sensitivity and specificity, with all subdomain scores equating or exceeding 0.85 as indicated by ROC analyses (Gotham et al. 2007). The RRB subscale score of the ADOS-2 was used in creating the RRB severity composite score.

Repetitive Behavior Scale-Revised

The Repetitive Behavior Scale-Revised (RBS-R; Bodfish et al. 1999) was rated by parent to assess the severity and variety of RRBs in individuals with ASD. The RBS-R is grouped conceptually into six subscales, including stereotyped behavior, self-injurious behavior, compulsive behavior, ritualistic behavior, sameness behavior, and restricted behavior/interests. Factor analyses have replicated and substantiated the six subscales, with the total RBS-R demonstrating high levels of internal consistency and interrater reliability for outpatient settings (Lam and Aman 2007). The total RBS-R score was used in computing the composite score and the restricted behavior/interests subscale was also used independently as a measure of RI.

Social Responsiveness Scale- Second Edition

The Social Responsiveness Scale (SRS-2; Constantino and Gruber 2012) is a parent-rated ASD screener used to identify the presence and severity of social impairment in children and adults ages 2.5 through adulthood. The SRS-2 shows excellent internal consistency (0.95), test–retest reliability (0.88–0.95), and interrater-reliability (0.66–0.71), in addition to validity metrics of sensitivity and specificity (0.92; Bruni 2014; Constantino and Gruber 2012). The DSM-5 subscale RRB severity score of the SRS-2 was used in creating the RRB severity composite score.

Procedure

All participants participated in an ASD assessment as part of a larger project in an autism research center. Participants or their parents were mailed all self-report measures and completed them prior to the in-clinic visit. Participants then completed one three-hour assessment session at the clinic during which time all the remaining measures from the list above were administered. Participants and primary caregivers provided verbal and written consent and/or assent to participate in the research study. Participants were accepted on a continuous referral basis and assessed in order of referral. Participants were not required to have a referral from a medical practitioner. Participants received the assessment and final report free of charge as part of an ongoing research study. Minimal attrition was observed except in unique cases of individuals who moved outside of the geographic area before being seen. The clinicians were research reliable in the administration of ASD-specific, gold-standard measures (e.g., ADOS, ADI-R). Approximately 20% of the administrations were co-coded by a second, research-reliable clinician. Consensus meetings were held with a research team to discuss each case and resulting diagnosis based on DSM-5 criteria, supervised by a licensed clinical psychologist. Due to the nature of the research clinic and the multiple projects ongoing, all evaluators were blind to the current hypotheses.

Analytic Strategy

Quantitative Analyses

Data were cleaned to remove any individuals with incomplete assessment data. Listwise deletions were used in cases of missing data. Chi-squared analyses were used to investigate differential probabilities of obtaining an ASD diagnosis based on sex in this clinic sample. Independent samples t-tests were used to investigate sex differences in age, IQ, and RRB severity. Exploratory analyses were conducted to investigate the interaction of age and sex on RRB severity. To further explore sex differences in RRB severity, an ANCOVA was conducted; variables with systematic relationships to both sex and RRBs were used as covariates in this analysis. Next, a one-way ANOVA was conducted to investigate sex differences on the RBS-R Restricted Behavior subscale; the only subscale that assesses RI specifically. Lastly, social communication impairment was investigated using one-way ANOVAs with sex as the grouping factor to investigate differences in social abilities in the ASD sample.

Descriptive Coding

Coding procedures were adopted from Harkness et al. (2011). A data-driven approach was used to preliminarily

record female and male participants' type of RI as reported in the ADI-R and/or ADOS-2. Ten larger RI categories emerged. Each RI category was assigned a number, coded by the lead researcher (TCM), and then co-coded by two coders blind to sex of the participant. If individuals presented with multiple interests, each interest was coded. Interests that spanned categories were dually coded (e.g., "My Little Pony" would be coded as both "technology" (TV show) and "animals").

Results

Quantitative Analyses

Quantitative analyses were conducted using IBM SPSS Statistics software (IBM Corp). The age distribution as a function of sex is available in Table 1. Pearson correlations indicated that age was significantly correlated with RRB ($r = -.264$, $p < 0.05$) and IQ ($r = 0.275$, $p < 0.05$), but no other correlations reached significance. Results indicated that RRB severity was significantly related to age, but not to IQ. One-way ANOVAs with age groups were conducted as exploratory analyses to investigate RRB severity as a function of age. All age groups (0–6, 7–12, 13–17, 18–57) demonstrated no significant sex differences in RRB severity ($ps > 0.2$) or RBS-R Restricted Behavior subscale ($ps > 0.2$); however, these analyses were significantly underpowered (for example, ages 13–17 contained only 6 individuals). Age was positively correlated with IQ. Chi square analyses indicated the female and male participants did not have a significantly different probability of diagnosis. That is, male and female participants in our sample had similar proportions of an ASD diagnosis, $\chi^2 = 3.472$, $p = 0.324$; 65% male, 50% female.

Independent samples t-tests were conducted to evaluate significant sex differences in RRB composite score, age, and IQ amongst participants with an ASD diagnosis. Results indicated significant sex differences for age, F (1, 78) = 6.548, $p = 0.01$, but not for IQ, F (1, 59) = 2.041, $p = 0.16$, or RRB severity, F (1, 78) = 0.054, $p > 0.5$ (for means, see Table 2). An Analysis of Covariance (ANCOVA) was conducted between sex and total RRB severity with age as a covariate. The results suggested that age was a significant covariate in the model, F (1, 1) = 15.82, $p < .01$, but no significant sex differences emerged. Lastly, a one-way ANOVA was conducted to investigate sex differences in the Restricted Behavior subscale of the RBS-R, which is the subscale that includes the variable of restricted interests. Results indicated significant sex differences, F (1, 75) = 4.51, $p = .03$, where male participants scored higher on the Restricted Behavior subscale than female participants ($M_{males} = 4.96$, $M_{females} = 3.23$).

Regarding social communication, males and females did not significantly differ on the ADOS Social Affect subscale, F (1,71) = 0.401, $p = .53$, SRS-2 Cognition subscale, F (1,59) = 0.88, $p = .35$, SRS-2 Communication subscale, F (1,59) = 3.287, $p = .08$, nor the SRS-2 Social Motivation subscale, F (1,59) = 0.008, $p = .90$.

Descriptive Coding

Ten categories of RI were created from the specific interests obtained from the ADOS and ADI-R (See Table 3). The inter-rater reliabilities for RI qualitative coding (K = 0.93) and number of RI (r = .96) were excellent. See Table 4 for RI categories by sex.

Female and male RI demonstrated multiple similarities. Both sexes expressed the same average *number* of interests ($M_{male} = 1.07$, $M_{female} = 1.05$). Also, 31% of female and 34% of male participants did not have any RIs. Males and females also demonstrated similar levels of RI in the areas of electronics (examples: "video games", "iPad", "watching TV"; 12% male and 8% female) and reading (examples: "autobiographies", "history nonfiction books"; 1% male and 4% female). The sexes differed, however, in their presentation of RI in other ways. Although both male and female participants on average had one RI, male participants demonstrated a wider range of interests. Male participants were more likely to report interest in vehicles (examples: "trains", "construction vehicles", "trucks"; 13%) and history (examples: "history nonfiction

Table 2 Autism spectrum disorder sample descriptive results

	IQ		Age**		RRB severity (Z scores)		Restricted behavior severity (RBS-R subscale)*	
	M	SD	M	SD	M	SD	M	SD
Male	91.80	18.01	10.34	10.02	0.95	1.84	4.96	3.41
Female	103.13	15.55	17.91	15.91	0.58	1.81	3.23	2.78

*p < .05

**p < .01

Table 3 Restricted interests categories, operational definitions, and numerical code

Interest	Definition	Code
People	Human individuals or groups currently living	1
Animals	Animals, animal parts, animal representations (i.e. animals print, stuffed animals)	2
Science	Subjects learned about in science curricula (i.e. physics, astronomy, physics, chemistry, geology)	3
History	Any association with a historical event or person	4
Technology	Anything relating to or associated with electronics. (Including interests that need to be accessed via a technological platform, such as TV shows)	5
Vehicles	Any piece of machinery powered by motor or representations of these (i.e. toy cars)	6
Symbols	Defined by the conventions of the Romance languages (letters, numbers, punctuation)	7
Reading	Anything related to reading (verb) or reading materials (nouns)	8
Other	Does not fit in above categories	9
None	No restricted interests	10

Table 4 Restricted interests percentages by sex

	Female	Male
People	7	2
Animals	27	9
Science	8	3
History	4	10
Technology	8	12
Vehicles	0	13
Symbols	0	4
Reading	4	1
Other	11	12
None	31	34

books", "past presidents"; 10%) relative to female participants (0% and 4%, respectively). Female participants were more likely to report interest in animals (examples: "cats", "snakes"; 27%), people (examples: "past presidents", "Oprah Winfrey"; 7%), and science (examples: "physics", "volcanos"; 8%) relative to male participants (9%, 2%, 3%, respectively).

Overall, female restricted interests had more of a social quality compared to males. When taken together, 34% of female RI were social, or having to do with living beings and interactions, where these interests only occupied 11% of male interests. Female participants indicated more interests in living constructs (i.e. people, animals), whereas male participants indicated more interests in object-related constructs (i.e. vehicles and symbols). Furthermore, female participants expressed their RI in a more socially-oriented manner compared to male participants. For example, comparing two adolescents with ASD with the same RI in animals, the female expressed her interests in cats by writing letters to animal activists and rescuing feral cats. The male expressed his snake interests by collecting figurines and watching snake videos online.

Discussion

This study aimed to provide more information about sex differences between restricted and repetitive behaviors in ASD, in severity and description. Results demonstrated that males and females had similar composite RRB severity ratings as indicated by parent- and clinician-report, after accounting for age and IQ; however, male participants had higher scores than females on the RBS-R Restricted Behavior subscale, which emphasizes restricted interests. Additional findings include that female participants had a narrower range of interests, which were more socially-oriented than male participants.

With respect to sex differences, our sample demonstrated that females and males were similar in composite RRB severity, accounting for age and IQ, which does not support previous literature claiming females exhibit lower RRB severity than males (e.g., Duvekot et al. 2016; Hartley and Sikora 2009). It is possible that the current sample, a clinical sample from mostly rural areas and a variety of ages, drew from a different population than previous publications that drew from predominantly urban settings with medical practitioner-referral systems. However, when isolating the Restricted Behavior subscale of the RBS-R, sex differences emerged with higher scores in males vs. females. These findings underscore the importance of evaluating and measuring the qualitative nature of female RRBs, specifically with regard to restricted interests. One potential explanation for the results of males having more severe RRBs than females is that the female group was older, trended towards a higher IQ, and potentially were more social than males with ASD. However, as indicated by sex analyses across four measures of social competence in ASD, the male and female groups were equated on social competence and social motivation. Thus, these findings may, in fact, be uniquely related to the female presentation of ASD.

Consistent with the quantitative finding of higher Restricted Behavior scores in our male participants, our qualitative findings demonstrated that male participants reported a wider range of RI, including mostly object-related interests; female participants, however, demonstrated a narrower range of topics, including mostly interests in living beings, and expressed their RI in a more socially oriented manner compared to males. These findings partially replicate those of Hiller et al. (2014), which suggested that female RI were more "seemingly random" or related to TV characters and people, compared to male RI, which tended to be related to wheeled toys and screen time. Our results provide additional data to explain the sex difference in the ASD diagnosis: perhaps females who have RIs simply express them in a more socially appropriate way (e.g., writing letters), and therefore are harder to identify for referral or diagnostic purposes. This information is crucial in aiding the evidence-based assessment and diagnostic protocol for identification of ASD in females.

This study does have several limitations. First, the resulting sample size of female participants diagnosed with ASD was small. Although the initial sample was commensurate with previous publications, a larger female sample would have given us more information regarding RIs. Second, our sample spans all ages and thus age-specific analyses were not able to be computed. While this can be interpreted as a limitation in terms of a broader scope, a lack of age restriction can also be seen as a strength, as these findings can assist in the development of appropriate assessment tools for individuals across the lifespan. Third, it is important to realize that although RRB severity was similar across sexes, this does not mean the RI severity was equal, as there was not enough power to specifically analyze RI-only items of the "Restricted Behavior" subscale of the RSB-R in this sample. Lastly, as this sample was self-referred, it is possible that these results reflect the fact that more boys and men are seeking services due to greater impairment. It is possible that female participants did not seek out services at the same rate as male participants, as indicated by the larger proportion of males in this sample, due to their symptoms (including their RRBs) being less severe. However, if this were the case, the presenting females would have more severe RRBs, making differences between males and females harder to detect. Thus, the significant results concerning sex differences on the RBS-R are still clinically significant and relevant.

Future research should further investigate the nature of RI across sexes to gain a better understanding of the male and female ASD presentation across the broader phenotypes and age ranges. Additionally, it may be helpful for future studies to further examine different dimensions of RI (e.g., nature/content vs. mode of expression) and discrepancies in these between the sexes. Clinically, assessment in the female presentation of ASD should be thorough when assessing for restricted interests and assess for interests that are pervasive, even if socially-appropriate or not as markedly impairing.

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Compliance with Ethical Standards

Conflict of interest All authors declare that they have no conflict of interest.

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