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Sex/Gender Differences in Screening for Autism Spectrum Disorder: Implications for Evidence-Based Assessment

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Autism spectrum disorder (ASD) is diagnosed more often in boys than in girls; however, little is known about the nature of this sex/gender discrepancy or how it relates to diagnostic assessment practices. This study examined the performance of the Social Communication Questionnaire (SCQ) in screening for ASD among boys and girls. Data were drawn from the South Carolina Children’s Educational Surveillance Study, a population-based study of ASD prevalence among children 8–10 years of age. Analyses were conducted using SCQ data from 3,520 children, with direct assessment data from 272 with elevated SCQ scores. A bifactor model based on the Diagnostic and Statistical Manual of Mental Disorders’s (5th ed.) two ASD symptom domains fit the data well and performed slightly better for girls. In the general population sample, girls exhibited fewer social communication/interaction and restricted-repetitive behavior symptoms than boys. In the direct assessment sample, however, girls with ASD showed greater impairment in social communication/interaction than boys with ASD. Items pertaining to social communication/interaction problems at ages 4–5 were among the most diagnostically efficient overall and particularly for girls. Similarly, receiver operating characteristic analyses suggested that the SCQ performs adequately among boys and well among girls. Results support the use of the SCQ in screening for ASD but do not indicate sex/gender-specific cutoffs. Girls with ASD may exhibit pronounced intraindividual deficits in social communication/interaction compared to male peers with ASD and female peers without ASD. Although more research is needed, careful attention to social communication/interaction deficits around 4–5 years of age may be especially useful for assessing ASD in girls.

Autism spectrum disorder (ASD) is a neurodevelopmental disorder defined by pervasive deficits in social communication and interaction and patterns of restricted, repetitive, stereotyped behaviors and interests (American Psychiatric Association [APA], 2013). Beyond these core criteria, however, there is considerable heterogeneity in the symptom presentations exhibited by children with ASD, including a variety of qualitative and quantitative differences (e.g., severity, language, cognitive ability, co-occurring problems). One critical factor in understanding ASD symptom variability is the role of sex/gender1 (e.g., Goldman, 2013; Lai, Lombardo, Auyeung, Chakrabarti, & Baron-Cohen, 2015).

Sex/gender differences in ASD are both over- and under-acknowledged. On one hand, it has been known for decades that ASD is more common in boys than girls. Estimates suggest that the true sex/gender ratio is about 3.3 to 1 (based on higher quality and population-screening studies; Loomes, Hull, & Mandy, 2017), whereas the ratio among those

1Following Lai et al. (2015), we use both terms (sex and gender) together when referring to differences between boys and girls throughout most of this article to acknowledge that it is not clear whether biological sex at birth or social gender constructs are the key variable; both likely play a role.
clinically diagnosed is about 4.5 to 1 (based on administrative records; Christensen et al., 2016). On the other hand, little is known about the nature of sex/gender differences in symptom presentations among children with ASD. To the extent that ASD presents differently in boys versus girls, it is possible that girls with ASD are underidentified. Indeed, Loomes et al.'s (2017) finding that the magnitude of the sex/gender discrepancy is inversely related to study quality suggests that there is gender-related ascertainment bias in clinical diagnosis. Complicating matters further, the evidence base pertaining to ASD—and, by extension, the diagnostic criteria themselves—historically comes from research among overwhelmingly male samples (e.g., Edwards, Watkins, Lotfizadeh, & Poling, 2012; Watkins, Zimmermann, & Poling, 2014). Thus, although there is clearly a need for more research to better understand sex/gender differences in ASD, it is also important to keep in mind that existing assessment tools and diagnostic criteria may contain sex/gender bias.

The present study seeks to advance the literature on both of these fronts: (a) to elucidate the extent and nature of sex/gender differences in ASD symptom presentations using comprehensive assessment methods and a population-based sample and (b) to help understand the extent to which sex/gender bias may be operating in screening for ASD using the Social Communication Questionnaire (SCQ; Rutter & Bailey, 2003). To investigate these questions, we analyzed data from a large epidemiological sample of school-age children who were screened and assessed for ASD. Thus, the present analysis offers a unique lens for investigating ASD screening and symptom presentation among boys and girls with and without the diagnosis.

**SEX/GENDER DIFFERENCES IN ASD**

Evidence has been mixed with respect to sex/gender differences in core ASD symptoms. Some studies indicate that boys have greater social and communicative problems compared with girls (e.g., Beggio et al., 2017; Head, McGillivray, & Stokes, 2014; Hiller, Young, & Weber, 2016), whereas others show the opposite pattern (e.g., Carter et al., 2007; Frazier, Georgiades, et al., 2014; Hartley & Sikora, 2009), and still others show no particular differences in this domain (e.g., Bölte, Duketis, Pousta, & Holtmann, 2011; Holtmann, Bölte, & Pousta, 2007; Mandy, Chilvers, et al., 2012; May, Cornish, & Rinehart, 2016; Reinhardt, Wetherby, Schatschneider, & Lord, 2015; Szatmari et al., 2012). Similarly, some studies have found boys with ASD to exhibit higher levels of repetitive and stereotyped behaviors than girls (Beggio et al., 2017; Bölte et al., 2011; Hartley & Sikora, 2009; May et al., 2016; Szatmari et al., 2012), whereas others have found no differences in this domain (Carter et al., 2007; Holtmann et al., 2007; Reinhardt et al., 2015). In their meta-analysis, Van Wijngaarden-Cremers et al. (2014) found that among individuals with ASD beyond age 6, males show higher levels of restricted/repetitive behaviors and interests than females; no significant gender differences were found for social interaction or communication (Van Wijngaarden-Cremers et al., 2014). Narrative reviews (e.g., Kirkovski, Enticott, & Fitzgerald, 2013; Lai et al., 2015) have yielded similar conclusions.

In addition, there are clinically important sex/gender differences in ASD that are not related to the core diagnostic symptoms. Compared to boys, girls with ASD more often go undiagnosed or are diagnosed at a later age, particularly girls with less severe ASD symptoms and more intact language and cognitive skills (Begeer et al., 2013; Giarelli et al., 2010; Rutherford et al., 2016). Girls with ASD may also be better able to compensate for symptoms despite having persistent core deficits associated with ASD (Livingston & Happé, 2017), which might contribute to greater social “camouflage” (Hull et al., 2017). For example, some evidence suggests that girls with ASD perform better on measures of nonverbal communication, which may mask their symptoms (Rynkiewicz et al., 2016). Despite this compensation, research examining peer relationships found that boys and girls with ASD exhibit more similarities with one another than with their same-gender, typically developing peers; however girls with ASD appear to face more social, friendship, and language demands than boys with ASD (Dean et al., 2014). More broadly, girls can exhibit patterns of restricted interests and repetitive behaviors and social and communicative problems which might seem more socially acceptable than the patterns seen in boys with ASD (Lai et al., 2015). This could help explain why girls with ASD often have more severe behavioral, emotional, and cognitive problems compared to boys with ASD (e.g., Frazier, Georgiades, et al., 2014; Holtmann et al., 2007; Horiuchi et al., 2014; Stacy et al., 2014), and even compared to girls at risk for ASD who are not ultimately diagnosed (Dworzynski, Ronald, Bolton, & Happé, 2012). That is, perhaps girls must exhibit more severe symptoms, impairment, or co-occurring problems to receive a diagnosis of ASD.

One possible explanation for these sex/gender differences is the “extreme male brain theory” of ASD (Baron-Cohen, 2002). After reviewing the evidence for behavioral sex/gender differences, Baron-Cohen concluded that on average, males exhibited weaknesses at empathizing and strengths at systematizing compared to females. Thus, ASD could be a disorder of the extreme male brain, characterized by low levels of empathizing traits (e.g., social-emotional understanding, pragmatic language, friendship development and maintenance) and high levels of systematizing traits (e.g., attention to detail, preference for rule-based systems and facts, preoccupation with cause-and-effect systems, and islets of ability; Baron-Cohen, 2002; Baron-Cohen, Knickmeyer, & Belmonte, 2005). This theory has garnered some support by way of between-group behavioral differences (e.g., Stauder, Cornet, & Ponds, 2011; Tan et al., 2015) and evidence linking masculinization and ASD traits to fetal testosterone exposure (e.g., Auyeung et al., 2009; Baron-Cohen et al., 2011). However,
this account has also been criticized for being too biologically reductive and neglecting gender socialization processes (e.g., Buchen, 2011; Krahn & Fenton, 2012). Some evidence suggests that ASD is a gender-defiant disorder rather than a disorder of masculinization (Bejerot et al., 2012), and other research suggests that normative sex differences in typically developing populations are absent in children with ASD (Park et al., 2012). Further research is needed to clarify these mixed findings.

RELEVANCE TO SCREENING

Much of the research just summarized has focused on children who have already received the diagnosis, sometimes with a non-ASD comparison group. Although such studies provide insight into clinical populations, they do relatively little to improve the assessment of boys and girls whose ASD diagnostic status is unknown. This is a major gap in the literature. Without addressing the nosological and diagnostic challenges pertaining to sex/gender considerations, any research on ASD based on existing assessment practices is subject to the underlying problem of not knowing how ASD should be defined and diagnosed in males compared to females (Lai et al., 2015). Thus, there is a need for rigorous population-based assessment research with attention to sex/gender. It is possible that systematic sex/gender differences could arise at any step in the assessment pipeline—from eliciting concerns about ASD to the results of diagnostic evaluations. Screening measures are particularly key for understanding sex/gender differences in symptom presentation and for addressing any systematic problems related to which children get referred for ASD evaluations. Improved interpretation of screening measures may lead to earlier identification for children in need of services. The SCQ (Rutter & Bailey, 2003) is one of the most widely researched and recommended parent-report screening measures for ASD in youth (Norris & Lecavalier, 2010; Ozonoff, Goodlin-Jones, & Solomon, 2005). Although previous research has investigated the general diagnostic utility of SCQ and similar measures for screening for ASD (e.g., receiver operating characteristic [ROC] and sensitivity/specificity; Barnard-Brak et al., 2016; Chandler et al., 2007; Duvekot, Van Der Ende, Verhulst, & Greaves-Lord, 2015; Eaves, Wingert, Ho, & Mickelson, 2006; Ung et al., 2016), there has been little attention to sex/gender differences. The notable exception is that some authors have found evidence for little to no measurement invariance in the SCQ (Wei, Chesnut, Barnard-Brak, & Richman, 2015) or similar screening measures (Frazier, Ratliff, et al., 2014; Frazier & Hardan, 2017).

Although the SCQ demonstrates excellent psychometrics among school-age children (Chesnut, Wei, Barnard-Brak, & Richman, 2017; Norris & Lecavalier, 2010), its clinical and research utility is limited by its lack of subscales, yielding only a single total score. In developing the SCQ (Berument, Rutter, Lord, Pickles, & Bailey, 1999; Rutter & Bailey, 2003), the authors pulled items from the three Autism Diagnostic Interview–Revised domains (Lord, Rutter, & Le Couteur, 1994), offering one possible subscale structure. Then they estimated a three-factor exploratory principal components analysis from their clinical sample of 200, offering a different possible structure. Neither of these models has been validated for clinical or research purposes. Others (Wei et al., 2015) have subsequently adopted the SCQ’s exploratory model or developed their own (e.g., Gau et al., 2011). However, the most compelling and copious evidence from a variety of ASD measures (e.g., Frazier & Hardan, 2017; Frazier, Youngstrom, Kubu, Sinclair, & Rezai, 2008; Frazier et al., 2012; Mandy, Charman et al., 2012) supports the two-domain framework that was codified in the Diagnostic and Statistical Manual of Mental Disorders (5th ed.; DSM-5; APA, 2013). For this reason, and to optimize the usefulness of our results, we examine a two-domain bifactor model of the SCQ.

THE PRESENT STUDY

In sum, the literature documents a large sex/gender discrepancy in ASD diagnoses and symptoms, with mixed evidence and explanations as to why. The present study investigates the extent and nature of sex/gender differences in ASD symptoms among a large epidemiological sample of school-age children and how these differences affect the SCQ in screening for ASD. Specifically, we examine (a) the prevalence of ASD markers in school-age children, overall and by sex/gender; (b) differences in SCQ results related to sex/gender and ASD diagnostic status, and their interaction; (c) the diagnostic efficiency of the SCQ in screening for ASD in boys and girls; and (d) whether different clinical cutoffs should be considered for boys and girls. Based on previous research, it was hypothesized that, among those with and without ASD diagnoses, boys would show higher ASD symptoms overall and particularly in restricted/repetitive interests and behaviors. It was expected that this would lead to sex/gender-driven measurement problems, potentially detrimentally affecting the identification of girls’ ASD symptoms. Because diagnostic status was used as our criterion, this study could not examine sex/gender bias in the diagnostic construct but rather focused on the performance of the SCQ. Results may help advance assessment practices and knowledge related to sex/gender differences in ASD or in the performance of the SCQ.

METHODS

Participants

Data were drawn from the South Carolina Children’s Educational Surveillance Study (SUCCESS), a population-
based study of ASD prevalence among school-age children. The study design and methodology has been detailed elsewhere (see Carpenter et al., 2016). The present analyses and descriptive statistics are based on all available data for children whose parent provided consent and fully completed the English version of the SCQ ($n = 3,520$). The target population consisted of all children born in 2004 living in a three-county catchment area in coastal South Carolina. Participants were 8 to 10 years of age at the time of the initial screening. Those who were invited for a direct assessment were slightly older by the time their evaluation occurred ($M = 10.3$ years; $SD = 0.5$; range = 8.8–11.4).

Procedures (detailed next) were designed to obtain as large and representative a sample as possible, and preliminary results suggest a reasonable degree of representativeness was achieved. In the population-screening sample, racial/ethnic backgrounds were as follows (roughly similar to census estimates): 61% non-Hispanic White, 27% non-Hispanic Black, 6% Hispanic, 3% other, and 3% multiracial. In the direct assessment sample, racial/ethnic background proportions were as follows: 44% non-Hispanic White, 37% non-Hispanic Black, 13% Hispanic, 1% other, and 4% multiracial. Compared to girls, greater proportions of boys fell in the clinical range ($SCQ \geq 15$) and in the at-risk range ($8 \leq SCQ < 15$; see Table 1) during the screening, rendering them more likely to be eligible for a direct assessment (35% of boys vs. 24% of girls). Of these, boys (29%) were more likely to be invited to and ultimately complete an assessment compared to girls (22%). Thus, the gender ratio shifted from census-estimated 51% male in the population to 49% male in the screening sample to 65% male in the direct assessment sample. Sociodemographic variables were not used to adjust the population screening procedures, direct assessment sampling, clinical assessment, or analyses.

### Procedures

All procedures were approved by the researchers’ institutional review board. As described by Carpenter et al. (2016), a multiphase sampling design was used. Procedures were designed to maximize participation rates from the entire population, including special education students. Extensive efforts were taken to ensure that the sample was as representative as possible, including steps to boost participation among students from ethnic minority and lower socioeconomic backgrounds. Recruitment and sampling procedures were developed based on the literature and in partnership with schools and organizations in the three-county catchment area. Ultimately, 123 of 127 public and private schools agreed to participate. Within a 2-month period, families of eligible children received via their school an introductory letter; packet with cover letter, waiver, and SCQ; and up to two reminders. Parents were allowed to complete an online or paper version of the SCQ or to decline. Incentives for responding were provided for students, parents, and teachers.

After completing the SCQ, a subset of participants was identified and invited for an in-person ASD assessment.
based on their SCQ scores. Given questions regarding the optimal cutoff value for the SCQ (Eaves et al., 2006; Norris & Lecavalier, 2010), all those in the “at-risk” range (SCQ ≥ 15; 100% invited; 44% completed; n = 112) and a randomly selected portion of those in the “subthreshold” range (8 ≤ SCQ ≤ 14; 69% invited; 20% completed; n = 160) were invited for a direct assessment. This included a separate informed consent and a comprehensive ASD diagnostic assessment (measures described next). These direct assessments were completed by doctoral-level psychologists with appropriate training and expertise in ASD evaluation. Participants’ ASD diagnostic status was determined based on the integration of all assessment data. Examiners were not blinded to SCQ scores, but neither these nor sex/gender status were considerations for diagnostic decision making. All cases were reviewed by the team on a weekly basis, with diagnostic ambiguity resolved by consensus. Examiners’ interrater reliability was 100% for case status.

From the census-estimated population of 8,780 children, 4,185 survey responses were recorded, of which 3,698 (42%) were usable data. The present analyses are based on data with complete responses on the English SCQ (excluding Spanish and partial SCQs), resulting in a final analytic sample of 3,520 (40% of population), including 272 who ultimately completed a direct assessment.

Measures

Screening

The SCQ Lifetime Form was used to screen for ASD in the full sample. The SCQ is a brief, standardized checklist of 40 items pertaining to symptoms of ASD, including problems with communication and reciprocal social interaction, and restricted, repetitive, and stereotyped behaviors (Rutter & Bailey, 2003). All items are in a yes/no format; some ask if the child has ever exhibited the behavior, whereas others focus specifically on the period of 4–5 years of age when symptoms of ASD may become more apparent. Items assess both atypical and typical behaviors, the latter being reverse coded. Possible scores range from 0 to 39, with higher scores indicating greater likelihood of ASD. To minimize the possibility of parents recognizing questions as pertaining to ASD symptoms, the project was promoted as a study of child social development and the SCQ was licensed by the publisher to be presented as a “SUCCESS Questionnaire”; no changes were made to the SCQ instructions or items. The English Lifetime SCQ has demonstrated ample evidence of validity and reliability, including good specificity and sensitivity (Chandler et al., 2007; Chesnut et al., 2017). The SCQ had good internal consistency in the present study (Cronbach’s α = .82). As previously noted, the SCQ does not have validated subscales. For the present analyses, three of the coauthors (two doctoral-level psychologists and one predoctoral psychology intern) with expertise in ASD evaluation divided the SCQ items into two subdomains mapping onto DSM-5 ASD criteria: (a) social communication and interaction (SCI) deficits (25 items; e.g., spontaneously used gestures, smiles back, talks to be friendly) and (b) restricted and repetitive behavior (RRB; 12 items, e.g., unusual special interests, odd mannerisms, repetitive language). Two items pertaining to self-injurious behavior (SCQ Item 17) and solitary make-believe play (SCQ Item 35), which are included in the total score, were not included in subdomain scores because there was no direct correspondence with DSM-5. This bifactor model was tested and supported via confirmatory factor analysis (see Results).

Direct Assessment

Consistent with recommendations (e.g., Ozonoff et al., 2005), multiple instruments and methods were used in the diagnostic evaluation. First, a structured ASD diagnostic interview was administered to a primary caregiver. This interview was developed for the SUCCESS study to assess current and lifetime symptoms of ASD using an integrated set of criteria that is compatible with DSM-IV (APA, 1994) and DSM-5. Second, the Autism Diagnostic Observation Schedule, second edition (ADOS-2; Lord, Luyster, Gotham, & Guthrie, 2012) was administered. The ADOS-2 is a semistructured, standardized test, commonly considered a “gold standard” instrument in ASD assessment. The ADOS-2 facilitates direct observation of ASD-related behaviors across several developmentally appropriate tasks and items, yielding a total score representing the likelihood of ASD and the severity of symptoms. The ADOS-2 and its predecessors have substantial evidence for validity, reliability, and utility in assessing ASD (Gotham et al., 2008; Lord et al., 2012, 2000; Molloy, Murray, Akers, Mitchell, & Manning-Courtney, 2011). Standard ADOS-2 procedures were followed, with modules determined by the child’s expressive language abilities (96% were Module 3). Finally, a variety of additional measures were administered assessing broadband (e.g., CBCL/TRF) and narrowband (e.g., Social Responsiveness Scale, 2nd ed.) symptoms, adaptive (Vineland-2) and cognitive functioning (e.g., Kaufman Brief Intelligence Test, 2nd ed.), language (Children’s Communication Checklist, 2nd ed.), medical and educational history, and demographics (see Carpenter et al., 2016). The DSM-5 ASD diagnoses were determined using clinical best-estimate procedures incorporating all available data, with primary consideration to the diagnostic interview and ADOS-2 results (Carpenter et al., 2016).

Analytic Plan

Descriptive statistics of SCQ scores and items were inspected overall and by sex/gender and diagnostic subgroups. Group differences in SCQ scores and item endorsements were

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1The other 487 screeners were excluded due to duplicate submissions, not living in the surveillance area in 2012, declined participation, or insufficient number of SCQ item responses for scoring.
estimated using $t$ tests (Cohen’s $d$ effect sizes), analyses of variance (ANOVA; partial $\eta^2$ effect sizes), and chi-square tests (Cramer’s $V$ effect sizes). Hypothesized SCQ factor structures were assessed via confirmatory factor analysis (CFA), with model fit evaluated through collective consideration of the root mean square error of approximation (RMSEA), and confirmatory fit index (CFI) and Tucker–Lewis fit index (TLI). Following recent recommendations (Kline, 2016; Little, 2013), fit indices were interpreted collectively as continuous measures with approximate thresholds (rather than strict cutoffs) for adequate model fit as follows: CFI/TLI $\geq .90$ and RMSEA $\leq .08$. CFAs were estimated in Mplus Version 7 (Muthén & Muthén, 2012) using weighted least squares. All other analyses were conducted in SPSS Version 24 (IBM, 2016).

The diagnostic utility of SCQ scores and items were examined through ROC analyses and diagnostic efficiency statistics as follows: sensitivity (proportion of those with ASD with positive test result$^3$ out of all those with positive result), specificity (proportion of those without ASD with negative test result out of all those with negative result), positive predictive value (PPV; likelihood of ASD diagnosis given a positive test result), negative predictive values (NPV; likelihood of no ASD diagnosis given negative test result), and diagnostic likelihood ratios (DLRs; calculated as [sensitivity]/[1 – specificity]). Clinically, DLR values represent the most concise estimate of diagnostic probability. DLRs around 1 indicate no change in the probability of the diagnosis, whereas higher DLRs represent increases in the probability of the diagnosis (e.g., DLRs 2, 5, 10 correspond to 15%, 30%, and 45% increases, respectively), and DLRs below 1 represent decreasing probability. These estimates should be interpreted according to the pretest and posttest probabilities of the population being considered (e.g., the probability of ASD diagnosis in a clinical setting vs. in the general population) (Youngstrom, 2013). Finally, ROC analyses were also utilized to consider diagnostic efficiency of the SCQ among boys and girls. Complex sampling weights were considered but were not used because they had little influence on other analyses, suggesting that the data are sufficiently representative for non-epidemiological analyses.

RESULTS

Confirmatory Factor Analysis

The single-factor CFA model (i.e., original SCQ scoring, with all 39 items loading onto a single construct) fit the data poorly, $\chi^2(702) = 17064.53$, $p < .001$, RMSEA $= 0.081$, 90% confidence interval (CI) [0.080, 0.082], CFI $= 0.711$, TLI $= .695$. By contrast, a bifactor model (i.e., with the single-factor plus subdomain factors of SCI and RRB) showed acceptable fit, $\chi^2(665) = 6056.07$, $p < .001$, RMSEA $= 0.048$, 90% CI [0.047, 0.049], CFI $= 0.905$, TLI $= 0.894$. The weighted least squares–adjusted $\Delta \chi^2$ test was significant, $\Delta \chi^2(37) = 4305.26$, $p < .001$, confirming that the bifactor model fit the data better than the single-factor model, and supporting the use of the two subdomain scores and the total score in subsequent analyses. Factorial invariance by sex/gender could not be examined due to nonconvergence of multiple-group models. Thus, the bifactor model was estimated separately by sex/gender, showing adequate fit for both boys, $\chi^2(665) = 3442.52$, $p < .001$, RMSEA $= 0.049$, 90% CI [0.048, 0.051], CFI $= 0.905$, TLI $= 0.894$, and girls, $\chi^2(665) = 2595.03$, $p < .001$, RMSEA $= 0.040$, 90% CI [0.039, 0.042], CFI $= 0.924$, TLI $= 0.916$. This model appears to show better fit for girls than for boys (e.g., nonoverlapping RMSEA CIs).

SCQ Results: Population Screening Sample

Descriptive statistics for SCQ scores are presented in Table 1 and item endorsement frequencies in Figure 1. As shown, markers of ASD symptoms were commonly endorsed in the general population. More than 50% of boys and girls had four or more SCQ items endorsed, and about one fourth of the items were endorsed by at least 20% of the sample. Boys had higher SCQ scores than girls, $t(3518) = 8.77$, $p < .001$, Cohen’s $d = 0.29$, and greater variability in the distribution of scores. A similar pattern was observed for the SCQ risk subgroups: 35.1% of boys fell in the elevated ranges (‘at-risk’ or ‘subthreshold’), compared to only 24.1% of girls, $\chi^2(2, N = 3,520) = 56.48$, $p < .001$, Cramer’s $V = 0.13$. Boys also had higher scores than girls for both the RRB domain, $t(3518) = 24.75$, $p < .001$ (boys $M = 2.52$, $SD = 2.83$; girls $M = 2.00$, $SD = 2.58$; Cohen’s $d = 0.19$) and for the SCI domain, $t(3518) = 57.23$, $p < .001$ (boys $M = 3.97$, $SD = 4.01$; girls $M = 3.04$, $SD = 3.20$; Cohen’s $d = 0.26$).

Figure 1 presents the frequency of item endorsement by sex/gender. The most frequently endorsed items overall included not using gestures at 4–5 years of age, using odd or repetitive speech, not spontaneously copying others at 4–5, getting pronouns mixed up, and making socially inappropriate questions or statements. The least frequently endorsed items included not responding positively to other children at 4–5 years of age, not showing things to parents at 4–5, not wanting parents to join in her or his enjoyment at 4–5, showing limited range of facial expressions at 4–5, and not able to have a to-and-fro conversation. After adjusting for multiple comparisons (family-wise Bonferroni approach; all $ps < .0013$), about half of the items showed significantly pronounced gender differences; in all cases, boys were rated as having higher symptom counts than girls. As shown in
Figure 1, items with the greatest sex/gender differences include interest in parts of toys, unusually intense interests, not having a best friend, odd mannerisms such as hand flapping, odd/repetitive language, and a variety of social deficits at 4–5 years of age (e.g., make-believe/imaginative games, spontaneously copying/joining others).

SCQ Results: Direct Assessment Sample

Among those who completed an in-person diagnostic evaluation, there were significant differences in the frequencies with which boys and girls were diagnosed with ASD, $\chi^2(1, N = 272) = 12.41, p < .001$, Cramér’s V = 0.21, with one fourth (24.9%) of boys assessed receiving the diagnosis compared to only 7.4% of girls assessed. A $2 \times 2$ ANOVA revealed a significant difference in SCQ scores between those with and without a diagnosis of ASD, $F(1, 268) = 72.46$, $p < .001$, partial $\eta^2 = .213$. Although there was no main effect for sex/gender ($p = .160$) after ASD diagnosis was included in the model, there was a significant interaction between sex/gender and diagnostic status, $F(1, 268) = 4.32$, $p = .039$, partial $\eta^2 = .016$. On average, girls diagnosed with ASD had
SCQ scores approximately 4 points higher than boys diagnosed with ASD, whereas boys and girls without the diagnosis did not differ in their SCQ scores. (See Table 1 for all descriptive statistics concerning total SCQ scores.)

Next, ANOVAs were reestimated for the RRB and SCI symptom domains. In the RRB model, there was only a main effect for ASD diagnosis in the expected direction, \( F(1, 268) = 6.22, p = .013 \), partial \( \eta^2 = .023 \), with diagnosed children showing higher levels of RRB (\( M = 7.18, SD = 3.17 \)) compared to nondiagnosed children (\( M = 5.17, SD = 3.11 \)), with no main effect or interaction for sex/gender (\( p > .4 \)). In the SCI model, however, there was a significant interaction between diagnostic status and gender, \( F(1, 268) = 7.67, p = .006 \), partial \( \eta^2 = .028 \), such that girls with ASD had SCI scores that were more than 4 points higher (\( M = 16.71, SD = 3.90 \)) than boys with ASD (\( M = 12.23, SD = 5.41 \)), whereas the pattern ran in the opposite direction for nondiagnosed girls (\( M = 6.93, SD = 4.17 \)) and boys (\( M = 7.74, SD = 4.31 \)). There was also a significant main effect for diagnostic status, \( F(1, 268) = 55.57, p < .001 \), partial \( \eta^2 = .172 \), with those with ASD showing greater SCI scores than those without. There was a marginal main effect for gender, \( F(1, 268) = 3.69, p = .056 \), partial \( \eta^2 = .014 \), with boys showing slightly higher levels of SCI overall (\( M = 8.86, SD = 4.99 \)) compared to girls (\( M = 7.65, SD = 4.86 \)).

Diagnostic Efficiency and Clinical Cutoffs

The sensitivity, specificity, PPVs, NPVs, and DLRs are present overall and by sex/gender at the item level in Table 2 and for the total SCQ scores (various cutoffs) in Table 3. At the item level, DLRs were relatively higher (> 2) for items related to spontaneous showing, sharing, initiation of joint attention, shared enjoyment, cooperative, imaginative, and spontaneous play, positive social response, not smiling back, interest in same-age peers, comforting parents, limited range of facial expressions, and odd mannerisms such as hand flapping. Regarding sex/gender differences, DLRs were higher for girls compared to boys on items relating to seeking shared enjoyment, pointing, nodding and shaking yes and no, sharing and showing, playing make-believe games, talking to be friendly, having a to-and-fro conversation, playing cooperative games with children, not looking when parent spoke, having little interest in same-age peers, and not spontaneously copying others. Only a few items showed greater DLRs for boys compared to girls, including odd mannerisms such as hand flapping, and whole-body movements (e.g., spinning, bouncing). Notably, for both boys and girls the most diagnostically efficient items were those pertaining to social-communication/interaction behaviors at 4–5 years of age.

As shown in Table 3, at the existing clinical cutoff of 15, the SCQ demonstrated good sensitivity and NPV, moderate specificity and DLR, and poor PPV. These lower values were driven by low PPV for both boys and girls (reflecting the high proportion of non-ASD cases above the cutoff) and low specificity particularly for boys (reflecting the high proportion of ASD cases below the cutoff). Similar patterns can be seen when alternative cutoffs are considered, with higher thresholds leading to better specificity, NPVs, and DLRs and poorer sensitivity and PPVs, and vice versa for lower thresholds. Figure 2 presents the ROC curves and the distribution of SCQ scores by sex/gender and diagnostic status. The SCQ showed better sensitivity and specificity for the identification of girls with ASD (area under the curve [AUC] = .977). Still, results showed a good AUC for boys (.791) and for the overall sample (.824). Although it is clear that the SCQ performed differently in boys and girls in this sample, it is difficult to discern specific cutoffs based on the observed data due to the truncated range and qualitative symptom differences as noted above. Strictly speaking, the optimal trade-off between sensitivity and specificity (i.e., maximizing the AUC) falls between 15 (boys) and 19 (girls). However, a visual inspection of Figure 2 indicates that using cutoffs this high would have resulted in 10 boys (but zero girls) with ASD going unidentified in the directly assessed sample. To the extent that screening measures should prioritize sensitivity, these results do not provide compelling evidence for developing gender-specific cutoffs or changing the overall clinical cutoff.

DISCUSSION

This study investigated sex/gender differences in ASD symptoms in a large sample of school-age children assessed for ASD. By applying rigorous assessment methods to a population-based sample, this design helps advance the literature beyond descriptions of sex/gender differences among diagnosed populations, toward useful clinical and research recommendations for diagnostic assessment. Our results coalesce around one interesting conclusion: Unlike their typically developing peers, girls with ASD have higher SCQ scores overall, specifically greater social communication problems, compared to boys with ASD. This pattern is only partially consistent with our hypotheses and may help explain other aspects of our results, as discussed next.

In the population sample, boys received higher SCQ scores than girls both overall and in the SCI and RRB domains, a finding that is roughly consistent with prior research (e.g., Van Wijngaarden-Cremers et al., 2014). Among those with ASD, however, girls showed higher SCI scores overall, specifically greater social communication problems, compared to boys with ASD. This pattern is only partially consistent with our hypotheses and may help explain other aspects of our results, as discussed next.

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### TABLE 2

Diagnostic Efficiency of SCQ Items for Identifying ASD in Boys and Girls Sorted by Full Sample DLR (Highest to Lowest)

<table>
<thead>
<tr>
<th>Item</th>
<th>Full Sample</th>
<th>Boys</th>
<th>Girls</th>
</tr>
</thead>
<tbody>
<tr>
<td>Symptom</td>
<td>Domain</td>
<td>Sens</td>
<td>Spec</td>
</tr>
<tr>
<td>33 At 4–5, Range of Facial Expression</td>
<td>SCI</td>
<td>.45</td>
<td>.85</td>
</tr>
<tr>
<td>40 At 4–5, Group Play</td>
<td>SCI</td>
<td>.76</td>
<td>.73</td>
</tr>
<tr>
<td>37 At 4–5, Response to Peers</td>
<td>SCI</td>
<td>.59</td>
<td>.79</td>
</tr>
<tr>
<td>30 At 4–5, Seeking Shared Enjoyment</td>
<td>SCI</td>
<td>.41</td>
<td>.85</td>
</tr>
<tr>
<td>31 At 4–5, Offering Comfort</td>
<td>SCI</td>
<td>.57</td>
<td>.78</td>
</tr>
<tr>
<td>27 At 4–5, Social Smiling</td>
<td>SCI</td>
<td>.53</td>
<td>.78</td>
</tr>
<tr>
<td>15 Hand or Finger Mannerisms</td>
<td>RRB</td>
<td>.55</td>
<td>.76</td>
</tr>
<tr>
<td>29 At 4–5, Offering to Share</td>
<td>SCI</td>
<td>.61</td>
<td>.73</td>
</tr>
<tr>
<td>36 At 4–5, Interest in Peers</td>
<td>SCI</td>
<td>.69</td>
<td>.67</td>
</tr>
<tr>
<td>34 At 4–5, Imitative Social Play</td>
<td>SCI</td>
<td>.75</td>
<td>.64</td>
</tr>
<tr>
<td>28 At 4–5, Showing and Directing</td>
<td>SCI</td>
<td>.35</td>
<td>.82</td>
</tr>
</tbody>
</table>

**Note:** SCQ = Social Communication Questionnaire; ASD = autism spectrum disorder; DLR = diagnostic likelihood ratio; Sens = sensitivity; Spec = specificity; PPV = positive predictive value; NPV = negative predictive value; 4–5 = 4–5 years of age; SCI = social communication and interaction; RRB = restricted and repetitive behavior.

that, among those likely to be referred for assessment, there might be little or no sex/gender differences in levels of RRB, whether overall or in terms of an interaction with diagnostic status. Thus, despite the robust sex/gender differences in RRB in the population, these particular behaviors do not appear to be associated with sex/gender differences or differentially contribute to an ASD diagnosis for boys more than girls or vice versa. Overall, these results are in line with Park et al.’s (2012) finding that normative sex/gender differences may be absent in children with ASD.

Item-level analyses offer further insight into these findings, with some of the least frequently endorsed items tending to be most useful for screening. Specifically, items pertaining to children’s spontaneous interaction and socially

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orientated behaviors at ages 4–5 appear to be among the most diagnostically efficient (showing good sensitivity and specificity) for informing a diagnosis of ASD in all children, and particularly for girls. Persistent SCI deficits appear to be a relatively sensitive and specific marker for differentiating girls with ASD from their typically developing female peers. Of interest, hallmark RRB features of ASD were generally not among the most diagnostically efficient items; only odd mannerisms such as hand flapping showed good diagnostic efficiency for boys and girls.

The bifactor SCQ model fit the data better than the single-factor model, and slightly better for girls than for boys. Although research examining measurement invariance is limited, studies have found similar item functioning and measurement invariance in the SCQ and other screening measures (Frazier & Hardan, 2017; Wei et al., 2015). Direct comparisons of the SCQ and SRS-2 might be particularly useful. Previous research suggests that age is a key factor affecting the performance of ASD symptom scales and screening measures; different patterns of results have been found, and other measures might perform better among children roughly 6 years of age and younger (Barnard-Brak et al., 2016; Van Wijngaarden-Cremers et al., 2014). However, in the present sample of 8- to 10-year-old children, results supported the SCQ’s bifactor structure and two-domain conceptualization of ASD put forth in DSM-5 (APA, 2013; Frazier et al., 2012; Mandy, Charman, et al., 2012). This suggests that there may be utility in further validation and clinical/research use of these two subscales derived from the SCQ. Although the SCQ items have previously been subdivided according to a variety of different exploratory (Berument et al., 1999; Gau et al., 2011) and conceptual/confirmatory (Rutter & Bailey, 2003; Wei et al., 2015) approaches, these results add to a body of evidence suggesting that ASD symptoms can be differentiated into a general domain with two subdomains, with implications for research and clinical assessment. For example, the SCQ could be refined not only as a screening tool but also as secondary instrument to help support or rule out ASD symptom domains for diagnosis. Due to the different number of items, however (25 for SCI vs. 12 for RRB), results should be interpreted with caution, and future work may be needed to render these scales more comparable.

ROC results show that the SCQ performs adequately as a diagnostic instrument for boys and girls, especially for girls. These findings are consistent with the factor analysis, subdomain, and item-level sex/gender differences just described. These results do not provide a compelling reason to alter the existing cutoff for boys or girls. However, caution in both directions is warranted: For boys and girls alike, scores falling in the “at-risk” range (≥15) are more likely to be false positives than true positives (probability of true positive = 43% and 21%, respectively); and among boys, scores falling below this cutoff (in the “subthreshold” range) still had a 10% probability of diagnosis. Thus, the SCQ should be interpreted cautiously and probabilistically. Clinically, an SCQ score of 15 or higher is associated with a small but clinically significant increase in the probability of ASD. As scores increase beyond 15, the probability of ASD increases proportionately, particularly for girls. Scores between 11 and 15 may increase the probability enough to warrant careful clinical judgment (e.g., Corsello et al., 2007;
Eaves et al., 2006). Overall, at the recommended threshold of 15, the sensitivity and NPV were good, the specificity and DLR were moderate, and the PPV poor. Thus, for both boys and girls, a positive value (above the cutoff) does not necessarily predict a diagnosis of ASD; indeed, a majority of cases above this cutoff were false-positives. For boys in particular, ASD cases were common among those in the subthreshold range. Again, if the full range of possible SCQ scores were represented in the data, these values might differ. As a screening instrument within the broader assessment context, false negatives might be more clinically detrimental than false positives given that the latter only indicates the need for further assessment.

We interpret our sex/gender-discrepant findings as evidence for qualitative rather than quantitative differences in ASD symptom presentations between boys and girls. The pattern of sex/gender symptom differences observed among those with ASD (SCI, boys < girls; RRB, boys = girls) is qualitatively distinct from the pattern observed in the general population (SCI, boys > girls; RRB, boys > girls). These results differ from the conclusions of a recent meta-analysis by Hull, Mandy, and Petrides (2016), which found no difference in RRB, and equivocal evidence for social impairment (Hull et al., 2016). In general, our findings are broadly consistent with the well-established sex/gender differences in ASD prevalence (e.g., Christensen et al., 2016; Loomes et al., 2017) but do not align neatly with existing theories in the literature that attempt to explain this discrepancy. For example, if the extreme male brain theory (Baron-Cohen, 2002) were supported, we might expect similar patterns of male–female levels of SCI (related to empathizing) and RRB (related to systematizing) in those with ASD as in those in the full population sample; this was not the case. Similarly, we did not find evidence for a female camouflage effect (e.g., Hull et al., 2017; Livingston & Happé, 2017; Rynkiewicz et al., 2016) insofar as parents did not rate girls diagnosed with ASD as possessing compensatory social skills (i.e., lower SCI scores), which might obfuscate their symptoms. It is possible that camouflage may still exist in settings that parents typically do not actively observe, such as educational
settings. Notably, the pattern of results shown in Figure 2 suggests it may be more difficult to differentiate ASD versus non-ASD status in boys than in girls.

**Strengths, Limitations, and Implications**

One strength of the present study is that comprehensive, multimethod assessment practices were used, which minimizes the possibility of results being influenced by bias or error. In other words, if the present gender-discrepant results reflect assessment error, then it is likely an underlying problem in ASD diagnostic criteria and assessment tools in general rather than the particulars of the present study. Herein lies the dilemma articulated by Lai et al. (2015): Because our existing conceptualizations and instruments (including SCQ, ADOS-2, and DSM-5 criteria) are derived from predominately male ASD samples, there remains a challenging problem of “the chicken and the egg.” That is, to the extent that ASD symptoms truly manifest differently in boys compared to girls, studies such as this one are not able to ascertain this difference. Broader research is needed to understand the qualitative nature of SCI and RRB among typically and atypically developing girls.

This does not, however, rule out the possibility of informant bias affecting SCQ scores, and this should be considered in interpreting these results. Parents’ perceptions of SCI deficits in boys and girls are likely influenced by sex/gender expectations relative to typically developing same-sex peers. This is consistent with previous research suggesting that the social relationships between boys and girls with ASD are more similar than relationships with their same-gendered peers (Dean et al., 2014). Similarly, bias may be operating in items pertaining to 4–5 years of age, as these rely on parents’ recall of behaviors occurring several years ago. The possibility of response bias might be illustrated in the rates at which different items were endorsed. Approximately half of children were rated as not using gestures at 4–5 years of age, suggesting that parents may be misinterpreting this item. This may be a limitation of the yes/no format of the SCQ, which some parents might struggle with. For example, Frazier et al. (2010) found that 5.1% of unaffected siblings were rated by their caregivers with SCQ score of 15 or higher. Thus, results of single items should be interpreted cautiously.

Additional limitations should be noted. First, using SCQ scores as the basis for direct assessment sampling results in an artificially truncated range and distribution of SCQ scores, which would not be seen if all participants received all measures. Thus, there may be children with ASD with scores in the low-risk range (SCQ < 8) who were missed, whereas those in the at-risk range (SCQ ≥ 15) were more likely to be invited and assessed than those in the subthreshold range (8 ≤ SCQ ≤ 14). Second, despite our large overall sample size, our direct assessment sample was relatively small in terms of gender-by-diagnosis subgroups, with 177 boys (only 25% of whom had ASD) and 95 girls (only 7% of whom had ASD). A related consequence is that the completion of in-person assessments may have been higher due to greater levels of parental concern; indeed, the response rate was higher among the at-risk group (44%) compared to the subthreshold group (29%). Although these data are considered to be a representative sample from a weighted epidemiological study, the present results should not be generalized to the entire population in an epidemiological manner (e.g., complex survey weights were not used in analyses). Rather, these findings can be interpreted simply as results of screening and assessment analyses conducted among the observed data with its limitations as just noted. However, these limitations are also reflective of a larger strength of this study—representative sampling of a population of more than 8,000 children, with nearly 50% participation and inclusion of subthreshold children so as to not miss more mildly affected cases.

Third, this study did not use a well-validated, published diagnostic interview. Rather, the evaluations used an unpublished structured diagnostic interview designed to map onto both DSM-IV and DSM-5 criteria, assessing lifetime and present symptoms within a reasonable administration time. Lastly, a larger and more pernicious problem is the possibility of sex/gender bias in the diagnosis of ASD itself. The present study (and much of ASD research) relies upon diagnostic criteria that have developed over the years from research largely among boys with ASD. The present study utilized a rigorous assessment protocol to ascertain the diagnosis; however, to the extent that the ASD construct is gender biased, these results cannot shed light on the nature of that bias and only highlight the need for broader research.

These findings have several implications for clinical and research assessment practices. First, elevated scores on the SCQ should be taken seriously regardless of sex/gender. It may be the case that clinically some girls with ASD are overlooked due to their perceived strengths in certain domains. These are important questions for a diagnostic evaluation. During the screening phase, however, a high score should not be overridden based on other perceived strengths. In addition, responses to specific items should be interpreted according to their developmental and social context. Careful attention might be given to items addressing social-communication and interaction behaviors at 4–5 years of age. In particular, girls with ASD may exhibit pronounced intraindividual SCI deficits compared to both their male peers with ASD and their female peers without ASD. Finally, positive results on screening measures should not be interpreted as indicating a diagnosis but only a need for a more comprehensive evaluation.
REFERENCES


