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Measuring Social-Communication Difficulties in School-Age Siblings of Children with ASD: Standardized versus Naturalistic Assessment

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Abstract

Younger siblings of children with ASD (high-risk siblings) are at elevated risk for developing the broader autism phenotype (BAP), which consists of subclinical features of ASD. We examined conversational skills in a naturalistic context and standardized assessments of pragmatic language and communication skills in high-risk and low-risk school-age children with BAP (n=22) and ASD (n=18) outcomes, as well as comparison children without ASD or BAP (n=135). Children with BAP characteristics exhibited lower conversational skills than comparison children, but did not differ on any of three standardized measures. Only the conversational ratings significantly predicted membership in the BAP versus Comparison group. This suggests that naturalistic tasks are crucial when assessing social-communication difficulties in children with a family history of ASD.

Lay Summary

The broader autism phenotype (BAP) consists of subclinical features of ASD and is more common among family members of those with ASD. School-age children with BAP characteristics exhibited lower conversational skills than comparison children, but did not differ on standardized language measures tapping similar abilities. This suggests that naturalistic tasks may be more sensitive to the social-communication difficulties seen in some children with a family history of ASD than the standardized language tests used in most evaluations.

Keywords

broader autism phenotype; social communication; high-risk siblings; measurement

Younger siblings of children with autism spectrum disorder (ASD; high-risk siblings) who do not develop ASD themselves are at elevated risk for developing the broader autism phenotype (BAP) compared to siblings with no family history of ASD (low-risk siblings). The BAP consists of subclinical characteristics related to the core features of ASD. These characteristics can include language difficulties or delays, social functioning deficits, and

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restricted or repetitive behaviors and interests or rigidity (Bailey, Palferman, Heavey, & Le Couteur, 1998; Pisula & Ziegart-Sadowska, 2015; Sucksmith, Roth, & Hoekstra, 2011). Among high-risk siblings without an ASD diagnosis, estimates of the BAP from previous studies have ranged from 21% to 30% at 36 months (Charman et al., 2016; Messinger et al., 2013) and from 17% to 41% at school age (Ben-Yizhak et al., 2011; Shephard et al., 2017).

The BAP is not a clinical diagnosis, but it does confer risks and challenges. Definitions and methods of measuring the BAP vary, but all have in common a focus on subclinical (e.g., milder, less functionally impairing) versions of ASD traits or symptoms. The BAP has been linked to difficulties in social relationships and poor mental health outcomes in childhood and adulthood. BAP features (measured using the Broad Autism Phenotype Questionnaire) have been associated with more difficulty initiating and maintaining friendships in emerging adulthood (Jamil, Gragg, & DePape, 2017). Elevated characteristics of the BAP (measured using the Social Responsiveness Scale and Autism Spectrum Quotient, respectively) have been associated with higher internalizing and externalizing problems (Walton, 2016) and behavior problems (Petalas et al., 2012) in childhood. Adult siblings of individuals with ASD, who were themselves identified in childhood as having BAP features, exhibited lower levels of social relationships and employment and higher levels of mental health problems than siblings without BAP characteristics (Howlin, Moss, Savage, Bolton, & Rutter, 2015).

At 36 months, high-risk siblings who have not developed ASD have higher rates of developmental delay, elevated ASD symptomatology, and lower adaptive functioning than low-risk siblings, with approximately one-fifth characterized by higher ASD symptomatology and/or lower levels of developmental functioning (Charman et al., 2016; Messinger et al., 2013). A previous study found that high-risk siblings scored lower on parent-rated pragmatic language than low-risk siblings at 36 months, with 35% showing evidence of pragmatic language impairment (Miller et al., 2015). Such features of atypical development may begin to emerge well before 36 months. Children with atypical outcomes at 36 months have shown significant differences from typically developing low-risk children by 12 months in domains including cognition, motor and language, but most prominently social communication (Ozonoff et al., 2014). These findings highlight the early emergence of BAP features and the importance of ongoing monitoring in high-risk siblings, even in the absence of an ASD diagnosis.

While prospective studies of high-risk siblings provide an opportunity to characterize aspects of the BAP in childhood, only a handful of studies have followed children past 36 months into school age to evaluate later outcomes. At 5 years old, a group of high-risk siblings without ASD showed differences from low-risk siblings in social-cognitive skills and levels of restricted interests and repetitive behaviors (Warren et al., 2012). In a study following children to 7 years old, high-risk siblings without ASD had higher repetitive behavior, lower adaptive functioning, and higher anxiety scores than low-risk siblings, and a subgroup exhibited subclinical ASD traits (Shephard et al., 2017). Another study followed up high-risk siblings between 5.5–9 years old, finding that high-risk siblings were more likely to have clinical concerns than low-risk siblings, including BAP, and the group with clinical concerns demonstrated subclinical autism symptoms (Miller et al., 2016).

Studies that have followed high-risk siblings into school age have tended to focus on group differences between high-risk siblings without ASD and low-risk siblings in domains relevant to ASD and the BAP. While such group differences highlight the increased likelihood of BAP features among high-risk siblings, not all siblings show such difficulties. It is likely that only a subset of high-risk siblings will exhibit difficulties consistent with the BAP. Characterizing specific challenges experienced by children with the BAP is crucial to identifying areas that should be monitored. Studies of the BAP in high-risk siblings have included a wide range of behavioral domains evaluated, as well as a variety of instruments and methods used to assess those domains (Pisula & Ziegart-Sadowska, 2015). It is important to evaluate whether different assessment methods elicit comparable profiles of strengths and weaknesses in order to identify children who may benefit from intervention.

Pragmatic communication is the use of both language and nonverbal behaviors to communicate appropriately with others in social contexts. Pragmatic language refers specifically to the appropriate use of verbal information in social contexts to convey messages, including speech acts, discourse management, and skills that reflect understanding of others' perspectives. Nonverbal pragmatic communication includes behaviors such as eye contact, facial expressions, gestures, management of interpersonal space, vocal intonation, and rate and volume of speech, all of which contribute to the social appropriateness and meaning of the speaker's messages (Greenslade, Utter, & Landa, 2019). Impairments in pragmatic communication are a hallmark of ASD, and therefore a domain in which individuals with the BAP may exhibit difficulty, making it particularly relevant to assess such skills appropriately among high-risk siblings (Drumm & Brian, 2013).

Pragmatic communication skills can be particularly difficult to measure, as these abilities are dependent on context and interactive partner (Norbury, 2014). Methods of evaluation include caregiver report (e.g., checklists), formal assessments (e.g., standardized measures), and observation (e.g., structured observation in naturalistic context). While checklists and formal assessments have the benefits of normative data, these approaches may not capture a comprehensive view of pragmatic abilities in the more subtle contexts of real-world settings (Adams, 2002; Norbury, 2014). To evaluate how different assessment methods perform in capturing the social-communication difficulties associated with the BAP, which are more subtle than those associated with ASD, this study compared pragmatic language skills measured via caregiver checklist, standardized assessment, and observation of behavior in a naturalistic context in school-age high- and low-risk children with BAP and ASD outcomes, as well as a comparison group. First, we examined group differences in standardized and more naturalistic measures of pragmatic communication. Then, we examined the relative predictive value of these measures to outcome group.

Method

Participants

Participants were younger siblings of children with ASD (high-risk siblings) or typical development (low-risk siblings), who were participating in a larger longitudinal study. High-risk siblings had at least one older sibling with ASD, confirmed using the ASD criteria on both the Autism Diagnostic Observation Schedule, Second Edition (ADOS-2)

and the Social Communication Questionnaire (SCQ). Low-risk siblings had older sibling(s) who were typically developing and no family history of ASD, confirmed by an Intake Screening Questionnaire and scores below the ASD range on the SCQ. Exclusion criteria for the high-risk group were a genetic disorder in the proband and birth before 32 weeks gestation. Exclusion criteria for the low-risk group were birth before 37 weeks gestation, developmental or learning conditions in an older sibling, and ASD in a first-, second-, or third-degree relative. Parents provided informed consent, children provided assent, and the study was approved by the university's Institutional Review Board.

Participants were assessed at school-age (*M* age = 11.32 years, range = 6.42–16.08, *SD* = 2.53) by examiners unaware of risk status and any prior assessments or outcomes, and they were classified into one of three outcome groups (see Table 1). The ASD group (*n* = 18, 14 male, 17 high-risk) scored above the ASD cutoff on the ADOS-2 at 36 months of age (Lord et al., 2012) and met *DSM-5* criteria for autism spectrum disorder at both 36 months and the school-age visit. The Broader Autism Phenotype (BAP) group (*n* = 22, 15 male, 21 high-risk) did not meet *DSM-5* criteria for ASD at the school-age visit, but demonstrated elevated social-communication difficulties as determined by 1) scores on the Broader Phenotype Autism Symptom Scale (BPASS; Dawson et al., 2007) *and* 2) a clinical best estimate rating of BAP by the examiner (based on all information collected at the visit), confirmed by the supervising clinical psychologist. Within the BAP group, 8 children received no diagnoses and had no other clinical concerns, as rated by the examiner. Of the remaining 14 children in the BAP group, the following diagnoses were made: 1 with Social (Pragmatic) Communication Disorder, 2 with Specific Learning Disorder, 3 with anxiety or mood disorders, and 8 with Attention Deficit/Hyperactivity Disorder. All participants not meeting criteria for inclusion in the ASD or BAP groups were classified into the Comparison group (*n* = 135, 66 male, 74 high-risk). Participants were included in the current study if they completed a conversation task with an examiner and had scores for at least one of the standardized measures.

Measures

Comprehensive Assessment of Spoken Language, Second Edition (CASL-2; Carrow-Woolfolk, 2017).—Participants were administered two subtests of the CASL-2, a measure of oral language skills, by the examiner. The Pragmatic Language (PL) subtest measures knowledge and appropriate application of pragmatic language rules (e.g., appropriate thing to say or do in a given situation), and the Nonliteral Language (NL) subtest measures understanding of meaning when language cannot be interpreted literally (e.g., sarcasm, metaphor, etc.). Standardized scores for each subtest were used for analyses.

Children's Communication Checklist-2 (CCC-2; Bishop, 2006).—The CCC-2 is a widely used 70-item parent questionnaire measuring multiple aspects of language, including syntax, semantics, and pragmatics. It yields an overall standardized score, the General Communication Composite (GCC), that was used for analyses. Though use of subscale scores was considered, all subscales were highly correlated with the GCC (r = .45–.76, ps < .001) and showed the same pattern of results; therefore, the GCC was chosen as the primary CCC-2 variable for analysis.

Conversational Skills Rating Scale (CSRS; Spitzberg & Adams, 2007).—During the study visit, examiners engaged in conversation with participants for approximately 7–10 minutes (M = 9.76 min). Examiners began with an open-ended question (i.e., "What do you like to do for fun?") and responded naturally to the participant. As needed, examiners introduced additional conversation topics (e.g., "Have you taken any trips or vacations recently?" or "Do you have any brothers or sisters?"). Participants were then rated using the CSRS by examiners, with 27% double-rated for reliability (ICC = .95). The CSRS was developed to provide a practical, efficient, and psychometrically sound method to assess specific, detailed aspects of conversational competence and interpersonal communication skills. It was designed to be relevant for both research and clinical contexts and applicable to a wide variety of conversational settings (Spitzberg & Adams, 2007). The CSRS measures 25 verbal and nonverbal behaviors—such as maintenance of topics and follow-up comments, initiation of new topics, asking of questions, use of humor and/or stories, involvement of partner as a topic of conversation, etc.—rated on a scale of 1 (inadequate) to 5 (excellent). These items are summed to create a total score for analyses (possible range = 25 to 125). The CSRS also yields four subscale scores–Attentiveness, Composure, Expressiveness, and Coordination–each containing a sum of 7 items (see Table 2 for items by subscale). The CSRS has shown good internal consistency of both the total score and subscales (all over .80) and correlates with other measures of conversation and relational quality (Spitzberg & Adams, 2007).

Broader Phenotype Autism Symptom Scale (BPASS; Dawson et al., 2007).—The BPASS was developed to assess subclinical autism-related traits. It is comprised of 13 ratings, 7 of which are based on a semi-structured parent interview and 6 of which are based on direct observation of the child during interactions with the examiner across the full visit. The 13 items are then aggregated into four composite scores, two based on the parent ratings (the Social Interest and Flexibility/Restricted Interests composites) and two based on direct observation (the Expressiveness and Conversation composites). Composite scores above 2 are indicative of difficulties in that domain (Dawson et al., 2007; Gerdts, Bernier, Dawson, & Estes, 2013); for the present study, at least one domain score above 2 was required for assignment to the BAP group. Among children in the BAP group, 11 had one domain score above 2; 5 had two scores above 2; 3 had three scores above 2; and 2 had four scores above 2. Only 3 of 22 participants were classified as BAP based on the observational items alone, with the other 19 classified based on parent ratings or a combination of both.

Wechsler Abbreviated Scale of Intelligence—Second Edition (WASI-II; Wechsler, 2011).—Participants were administered the WASI to assess cognitive functioning and verbal ability. Standardized scores for the Verbal Comprehension Index were used to characterize Verbal IQ (VIQ) of the sample.

Statistical Analyses

To examine how well the standardized measures versus the naturalistic task captured the phenotype of BAP, we first examined group differences. For each measure (CASL-2 Pragmatic Language and Nonliteral Language standard scores, CCC-2 General Communication Composite standard score, and CSRS Total Score and subscale scores), an

analysis of variance (ANOVA) was conducted to determine if there was a difference between outcome groups on the scores. Risk group was assessed as a potential covariate. ANOVAs were followed by planned comparisons between groups, with Bonferroni adjustment for multiple comparisons. Next, stepwise multinomial logistic regression analysis was employed to examine the relative predictive value of standardized assessments of pragmatics (CASL-2 and CCC-2) and the more naturalistic measure of CSRS ratings to outcome group. ROC analysis was used to determine the optimal cutoff score on the CSRS for discriminating the BAP from the Comparison group.

Results

Summary statistics (mean, *SD*) for CASL-2 subtests, CCC-2 GCC, and CSRS are presented in Table 1. There were no group differences in age. Verbal IQ was lower in the ASD group than the BAP and Comparison groups, but the BAP and Comparison groups did not differ in verbal IQ (see Table 1). Correlations between measures are presented in Table 3.

Group Differences

Patterns of group differences did not change with the inclusion of risk group as a covariate in any of the models, and therefore all models are presented without risk group included. Within the Comparison group, there were no group differences between high-risk and low-risk children on verbal IQ, CSRS Total Score, CASL-2 NL score, CCC-2 GCC, or any BPASS subscales. The only significant difference was found on the CASL-2 PL score, with high-risk children scoring lower ($M = 106.85$, $SD = 15.26$) than low-risk children ($M = 112.39$, $SD = 14.27$), $p = .03$.

Overall, the ASD group scored significantly lower than both the BAP and Comparison groups on all measures. The BAP group tended to score in the intermediate range between the ASD and Comparison groups, but in general, significantly higher than the ASD group in all areas and most similar to the Comparison group, differing significantly on only one measure (CSRS).

ANOVA showed a significant effect of outcome group on the CASL-2 Pragmatic Language (PL) subtest, $F(2, 169) = 49.25$, $p < .001$ (see Figure 1). Post hoc analyses indicated that the ASD group ($M = 71.29$, $SD = 14.51$) scored significantly lower than the BAP ($M = 102.09$, $SD = 14.93$) and Comparison ($M = 109.39$, $SD = 15.02$) groups, ps < .001. The BAP and Comparison groups did not differ from each other on the PL subtest, $p = .106$.

ANOVA also showed a significant effect of outcome group on the CASL-2 Nonliteral Language (NL) subtest, $F(2, 163) = 46.14$, $p < .001$ (see Figure 1). Post hoc analyses indicated that the ASD group ($M = 75.43$, $SD = 16.47$) scored significantly lower than the BAP ($M = 107.50$, $SD = 16.02$) and Comparison ($M = 114.76$, $SD = 14.24$) groups, ps < .001. The BAP and Comparison groups did not differ from each other on the NL subtest, $p = .121$.

ANOVA showed a significant effect of outcome group on the CCC-2 General Communication Composite (GCC) score, $F(2, 160) = 23.46$, $p < .001$ (see Figure 1). Post

hoc analyses indicated that the ASD group (M = 90.13, SD = 17.88) scored significantly lower than the BAP (M = 118.42, SD = 17.89) and Comparison (M = 121.23, SD = 16.93) groups, ps < .001. Again, the BAP and Comparison groups did not differ from each other on the GCC, p > .99.

ANOVA also revealed a significant effect of outcome group on CSRS Total Score, $F(2, 172) = 78.61$, $p < .001$ (see Figure 1). Post hoc analyses indicated that the ASD group (M = 66.00, SD = 18.45) scored significantly lower than the BAP (M = 85.68, SD = 7.70) and Comparison (M = 93.76, SD = 7.15) groups, ps < .001. In contrast to the other analyses, the BAP group scored significantly lower than the Comparison group, $p < .001$. These patterns remained when age was included as a covariate in the model. As a follow-up, we also examined group differences on the four subscales of the CSRS (see Table 1 for group means). Results were consistent with analysis of the total score (i.e., ASD group scored significantly lower than both the BAP and Comparison groups, and the BAP group scored significantly lower than the Comparison group) on the Attentiveness, Composure, and Expressiveness subscales, all ps ≤ .01. The exception was the Coordination subscale, which distinguished only the ASD group from the other groups, ps < .001, but found no significant difference between the BAP and Comparison groups, $p = .167$.

Prediction of Outcome Group from Standardized and Naturalistic Measures

A forward entry stepwise multinomial logistic regression model was used, in which significant predictor variables were added one at a time (in order of the largest statistically significant change in −2 Log Likelihood) and tested for significance until no remaining variables significantly contributed to the model. The dependent variable was outcome group classification, with the Comparison Group as the referent, and the predictor variables were the three standardized measures (CASL NL and PL subtests and CCC-2 GCC) and the more naturalistic measure (CSRS). The total score of the CSRS was used in regression analyses because of high correlations between the total score and subscale scores (range = .89 to .92) and among subscale scores (range = .72 to .89). We also confirmed that results remained the same if a forced entry method (with all variables included in the model simultaneously) was used, but we present the forward entry methodology in service of finding the most parsimonious model.

Model parameters and model fit statistics are presented in Table 4. The first variable entered into the model was CASL PL, which significantly improved model fit from an intercept-only model, $\chi^2(2) = 67.08$, $p < .001$. The second variable entered into the model was CSRS, which significantly improved model fit from the previous model, $\chi^2(2) = 14.72$, $p = .001$. Both CASL NL and CCC-2 GCC were omitted from the final model, as they did not significantly contribute above and beyond the other predictors. In predicting membership in the ASD group over the Comparison group, the CASL PL score was the strongest predictor, followed by CSRS score. In predicting membership in the BAP group over the Comparison group, the CSRS was the only significant predictor.

An ROC analysis was conducted to determine the optimal cutoff score on the CSRS for discriminating the BAP from the Comparison group. A threshold of 88.5 was the best discriminating cutoff score, with an area under the curve (AUC) of 72.45% (95%

Confidence Interval [CI] = 61.96% to 82.93%). Specificity was 72.55% (95% CI = 65.48% to 79.62%), sensitivity was 68.18% (95% CI = 48.72% to 87.65%), negative predictive value was 94.07% (95% CI = 89.81% to 98.33%), and positive predictive value was 26.32% (95% CI = 14.88% to 37.75%).

Discussion

This study examined the utility of standardized and naturalistic assessments in identifying social-communication difficulties in school-age children with BAP characteristics. School-age children with BAP outcomes exhibited lower conversational skills with an examiner than comparison children, but they did not significantly differ from comparison children on any of the three standardized language measures. Further, only the conversational ratings significantly predicted membership in the BAP group versus the Comparison group. This suggests that assessments measuring skills exhibited in a more naturalistic interactive context may be more sensitive than standardized language assessments in identifying social-communication difficulties in children with the BAP.

Children with ASD exhibited lower skills than both BAP and comparison groups on all measures—conversational skills rated in a naturalistic setting as well as standardized measures of communication skills. It is not surprising that children with ASD perform differently on the standardized measures, as these measures are designed to elicit such differences. The CASL-2 was validated with a clinical sample, demonstrating differentiation between typically developing individuals and those with several diagnoses, including ASD. The CCC-2 is also designed to assist in determining if a child may benefit from further assessment for ASD. In our sample, these measures both differentiated children with ASD from those without ASD—including from children identified as having the BAP but not ASD. However, they did not distinguish children with the BAP from comparison children. While standardized measures may work well for identifying difficulties at a level of impairment consistent with an ASD diagnosis, these measures do not appear to capture more subtle social-communication difficulties.

On standardized measures administered in this study, children with BAP characteristics scored in the average range. When asked, in a structured testing format, how to behave or respond in a hypothetical situation, these children were able to demonstrate knowledge of skills comparable to children in the comparison group. However, when children were participating in a naturalistic conversation with a social partner, differences emerged. This suggests that children with BAP features may not lack knowledge or formal understanding of pragmatic communication rules, but rather have difficulty implementing them within an ongoing interaction. Such difficulties are consistent with a performance or production deficit (Gresham, 2017), in which children can produce competent strategies when asked, but not spontaneously in real-time contexts. This distinction is critical in guiding the selection of appropriate intervention strategies, which may then focus on enhancing a child's *performance* of an existing skill rather than on the *acquisition* of such skills (Bellini, Peters, Benner, & Hopf, 2007). It is therefore crucial to include naturalistic assessments of social-communication skills within evaluations, particularly for children with a family history of

ASD, as more subtle difficulties children are experiencing may not be clearly evident using standardized assessments alone.

Another observational measure rated from naturalistic conversation, the Pragmatic Rating Scale—School Age (PRS-SA), has shown similar group differentiation between high-risk siblings with the BAP and comparison children (Greenslade et al., 2019). Both the PRS-SA and the CSRS include items involving use of nonverbal communication (such as eye contact, facial expressiveness, use of gestures) and verbal skills (such as topic maintenance, sufficiency of information). Additionally, in a study using items from the ADOS, a subgroup of school-age high-risk siblings (9–12 years old) with BAP features had lower pragmatic abilities than high-risk siblings without the BAP and low-risk siblings (Ben-Yizhak et al., 2011). Directly assessing such behaviors—in a real-world context—using measures that capture both verbal and nonverbal aspects of pragmatic communication may best capture pragmatic difficulties in these areas among high-risk siblings.

This study is the first to our knowledge to use the CSRS in children with ASD or the BAP. There is some overlap between the CSRS and the BPASS, in that both measures of social communication are rated by an examiner based on interactions with the child. Since BPASS scores are one aspect of the BAP group definition, it is therefore not entirely surprising that group differences on the CSRS were evident. However, it is worth noting that the CSRS rates many more behaviors (25 observational items) than the BPASS (6 observational items), so it is much broader in the scope of social-communication features it measures. Additionally, most children in the BAP group (19 of 22) were assigned to that group based on parent interview items as well, so it is unlikely that the finding is due simply to rater covariance.

ROC analysis of the CSRS determined an optimal cutoff score for discriminating the BAP from the Comparison group, but we urge caution in application of any threshold due to the small size of the BAP sample in this study and the low positive predictive value of the cutoff. Nevertheless, our study highlights the utility of this kind of measure to identify subtle difficulties in pragmatic communication in children with BAP features. Optimally, to identify children who would benefit from support with pragmatic communication, clinicians should include a measure of behavior observed in a naturalistic setting, such as the CSRS rated from a conversation task used in this study. Relying solely on standardized language assessments or parent report may not capture the more subtle difficulties associated with the BAP that may be flagged by lower scores on a measure such as the CSRS.

Our findings of the relative utility of observational measures are also consistent with work examining identification of BAP features in parents of children with ASD compared to parents of children with Down syndrome (de Jonge et al., 2015). Researchers conducted interviews with parents (about themselves and about their partner), as well as an observation measure, regarding characteristics of the BAP. While all measures differentiated the groups, differences were greatest for the observation measure. Additionally, a pragmatic rating scale did not improve group prediction. As in our study, findings highlight the importance of observational measures of social communication behavior in evaluating the BAP.

The parent-report measure of communication skills, the CCC-2, was least able to identify difficulties in children with the BAP. Previous studies of school-age high-risk siblings have also found either little evidence of impairment or inconsistencies using the CCC-2 when looking for differences between high-risk siblings as a whole and comparison groups. Bishop et al. (2006), for example, found group differences on only one subscale of the CCC-2, Syntax, though 24% of high-risk siblings scored more than 2 standard deviations below the control mean on a total score comprising all 10 subscales. Drumm and colleagues (2015) found no impairments in pragmatic language on the CCC-2 in school-age high-risk siblings compared to test norms; in fact, siblings were rated significantly better than the 50th percentile on the pragmatic scales. Our study, in which children already identified by clinicians as having BAP characteristics were compared to children without ASD or the BAP, found no significant impairment reported for the BAP group. The CCC-2 is rated based on observed behavior, as is the CSRS, but by a caregiver. The lack of differentiation between the BAP and Comparison groups on this measure may suggest the importance of obtaining expert ratings by an examiner trained in ASD and/or using naturalistic interactive contexts in identifying more subtle social-communication difficulties. The CCC-2 may not be particularly sensitive to difficulties exhibited by children with the BAP, and perhaps should not be used in isolation as a screener for BAP-related difficulties.

This study suggests that naturalistic conversation—and measures incorporating subtle verbal and nonverbal aspects of communication—may be more sensitive than standardized language assessments in identifying social-communication difficulties in children with the BAP. Despite scoring in the average range on pragmatic tests, children with the BAP appear to struggle implementing such skills moment-to-moment during interaction, making naturalistic settings crucial when assessing social-communication difficulties in high-risk children.

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References

Adams C (2002). Practitioner Review: The assessment of language pragmatics. Journal of Child Psychology and Psychiatry, 43(8), 973–987. doi:10.1111/1469-7610.00226 [PubMed: 12455920]

Bailey A, Palferman S, Heavey L, & Le Couteur A (1998). Autism: The phenotype in relatives. Journal of Autism and Developmental Disorders, 28(5), 369–392. doi:10.1023/A:1026048320785 [PubMed: 9813774]

Bellini S, Peters JK, Benner L, & Hopf A (2007). A meta-analysis of school-based social skills interventions for children with autism spectrum disorders. Remedial and Special Education, 28(3), 153–162. doi:10.1177/07419325070280030401

Ben-Yizhak N, Yirmiya N, Seidman I, Alon R, Lord C, & Sigman M (2011). Pragmatic language and school related linguistic abilities in siblings of children with autism. Journal of Autism and Developmental Disorders, 41(6), 750–760. doi:10.1007/s10803-010-1096-6 [PubMed: 20844942]

Bishop DVM (2006). The Children's Communication Checklist, version 2 (CCC-2) US Edition. New Jersey: Pearson.

Bishop DVM, Maybery M, Wong D, Maley A, & Hallmayer J (2006). Characteristics of the broader phenotype in autism: A study of siblings using the children's communication checklist-2. American

Journal of Medical Genetics Part B: Neuropsychiatric Genetics, 141B(2), 117–122. doi:10.1002/ajmg.b.30267

Carrow-Woolfolk E (2017). Comprehensive Assessment of Spoken Language, Second Edition (CASL-2) [Manual]. Torrance, CA: Western Psychological Services.

Charman T, Young GS, Brian J, Carter A, Carver LJ, Chawarska K, … Zwaigenbaum L (2016). Non-ASD outcomes at 36 months in siblings at familial risk for autism spectrum disorder (ASD): A baby siblings research consortium (BSRC) study. Autism Research, n/a-n/a. doi:10.1002/aur.1669

Dawson G, Estes A, Munson J, Schellenberg G, Bernier R, & Abbott R (2007). Quantitative assessment of autism symptom-related traits in probands and parents: Broader Phenotype Autism Symptom Scale. Journal of Autism and Developmental Disorders, 37(3), 523–536. doi:10.1007/s10803-006-0182-2 [PubMed: 16868845]

de Jonge M, Parr J, Rutter M, Wallace S, Kemner C, Bailey A, … Pickles A (2015). New interview and observation measures of the broader autism phenotype: Group differentiation. Journal of Autism and Developmental Disorders, 45(4), 893–901. [PubMed: 25245786]

Drumm E, & Brian J (2013). The developing language abilities and increased risks of 'unaffected' siblings of children with autism spectrum disorder. Neuropsychiatry, 3(5), 513–524.

Drumm E, Bryson S, Zwaigenbaum L, & Brian J (2015). Language-related abilities in 'unaffected' school-aged siblings of children with ASD. Research in Autism Spectrum Disorders, 18, 83–96. doi:10.1016/j.rasd.2015.07.007

Gerdts JA, Bernier R, Dawson G, & Estes A (2013). The broader autism phenotype in simplex and multiplex families. Journal of Autism and Developmental Disorders, 43(7), 1597–1605. doi:10.1007/s10803-012-1706-6 [PubMed: 23117424]

Greenslade KJ, Utter EA, & Landa RJ (2019). Predictors of pragmatic communication in school-age siblings of children with ASD and low-risk controls. Journal of Autism and Developmental Disorders, 49(4), 1352–1365. doi:10.1007/s10803-018-3837-x [PubMed: 30488153]

Gresham FM (2017). Evidence-based interventions for social skill deficits in children and adolescents. Handbook of evidence-based interventions for children and adolescents, 365–376.

Howlin P, Moss P, Savage S, Bolton P, & Rutter M (2015). Outcomes in adult life among siblings of individuals with autism. Journal of Autism and Developmental Disorders, 45(3), 707–718. doi:10.1007/s10803-014-2224-5 [PubMed: 25189825]

Jamil R, Gragg MN, & DePape A-M (2017). The broad autism phenotype: Implications for empathy and friendships in emerging adults. Personality and Individual Differences, 111, 199–204. doi:10.1016/j.paid.2017.02.020

Lord C, Rutter M, DiLavore P, Risi S, Gotham K, & Bishop S (2012). Autism Diagnostic Observation Schedule–2nd edition (ADOS-2). Los Angeles, CA: Western Psychological Corporation.

Messinger D, Young GS, Ozonoff S, Dobkins K, Carter A, Zwaigenbaum L, … Sigman M (2013). Beyond autism: A Baby Siblings Research Consortium study of high-risk children at three years of age. Journal of the American Academy of Child and Adolescent Psychiatry, 52(3), 300–308.e301. [PubMed: 23452686]

Miller M, Iosif A-M, Young GS, Hill M, Phelps Hanzel E, Hutman T, … Ozonoff S (2016). School-age outcomes of infants at risk for autism spectrum disorder. Autism Research, 9(6), 632–642. doi:doi:10.1002/aur.1572 [PubMed: 26451968]

Miller M, Young GS, Hutman T, Johnson S, Schwichtenberg AJ, & Ozonoff S (2015). Early pragmatic language difficulties in siblings of children with autism: implications for DSM-5 social communication disorder? Journal of Child Psychology and Psychiatry, 56(7), 774–781. doi:10.1111/jcpp.12342 [PubMed: 25315782]

Norbury CF (2014). Practitioner Review: Social (pragmatic) communication disorder conceptualization, evidence and clinical implications. Journal of Child Psychology and Psychiatry, 55(3), 204–216. doi:10.1111/jcpp.12154 [PubMed: 24117874]

Ozonoff S, Young GS, Belding A, Hill M, Hill A, Hutman T, … Iosif A-M (2014). The broader autism phenotype in infancy: When does it emerge? Journal of the American Academy of Child & Adolescent Psychiatry, 53(4), 398–407.e392. doi:10.1016/j.jaac.2013.12.020 [PubMed: 24655649]

Petalas MA, Hastings RP, Nash S, Hall LM, Joannidi H, & Dowey A (2012). Psychological adjustment and sibling relationships in siblings of children with Autism Spectrum Disorders: Environmental

stressors and the Broad Autism Phenotype. Research in Autism Spectrum Disorders, 6(1), 546–555. doi:10.1016/j.rasd.2011.07.015

Pisula E, & Ziegart-Sadowska K (2015). Broader autism phenotype in siblings of children with ASD—A review. International Journal of Molecular Sciences, 16(6), 13217–13258. [PubMed: 26068453]

Shephard E, Milosavljevic B, Pasco G, Jones EJH, Gliga T, Happé F, … Team TB (2017). Mid-childhood outcomes of infant siblings at familial high-risk of autism spectrum disorder. Autism Research, 10(3), 546–557. doi:10.1002/aur.1733 [PubMed: 27896942]

Spitzberg BH, & Adams TW (2007). CSRS, the Conversational Skills Rating Scale: An instructional assessment of interpersonal competence: NCA, National Communication Association.

Sucksmith E, Roth I, & Hoekstra RA (2011). Autistic traits below the clinical threshold: Re-examining the broader autism phenotype in the 21st century. Neuropsychology Review, 21(4), 360–389. doi:10.1007/s11065-011-9183-9 [PubMed: 21989834]

Walton KM (2016). Risk factors for behavioral and emotional difficulties in siblings of children with autism spectrum disorder. American Journal on Intellectual and Developmental Disabilities, 121(6), 533–549. doi:10.1352/1944-7558-121.6.533 [PubMed: 27802099]

Warren ZE, Foss-Feig JH, Malesa EE, Lee EB, Taylor JL, Newsom CR, … Stone WL (2012). Neurocognitive and behavioral outcomes of younger siblings of children with autism spectrum disorder at age five. Journal of Autism and Developmental Disorders, 42(3), 409–418. doi:10.1007/s10803-011-1263-4 [PubMed: 21519952]

Wechsler D (2011). Wechsler abbreviated scale of intelligence-Second edition (WASI-II): PsychCorp.

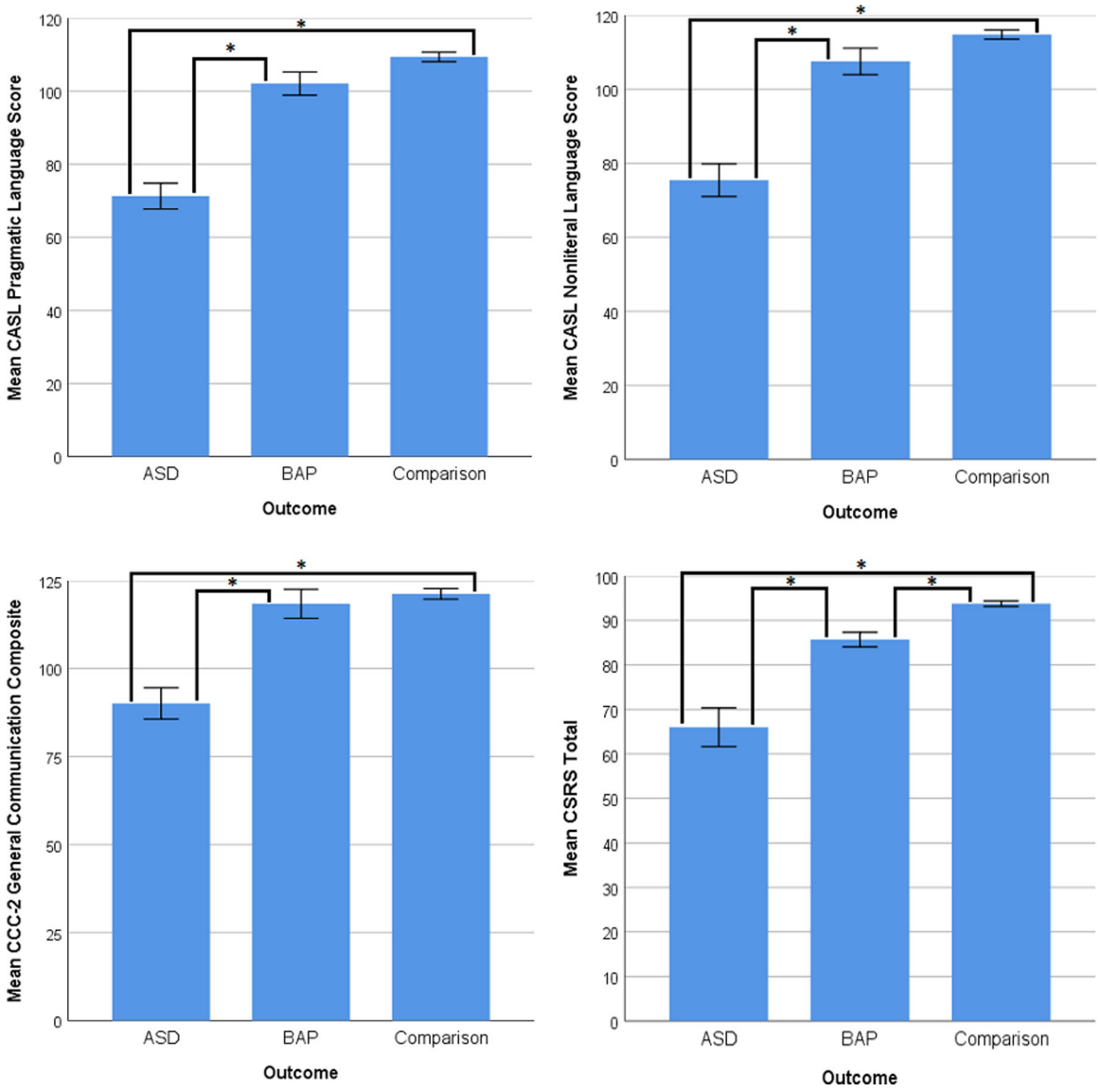

Figure 1.

Mean CASL-2 Standard Scores, CCC-2 General Communication Composite, and CSRS Total Score by outcome group.

Note. CASL-2 = Comprehensive Assessment of Spoken Language, 2nd edition; CCC-2 = Children's Communication Checklist-2; CSRS = Conversational Skills Rating Scale. *$p <$ .001; error bars represent +/− 1 standard error.

Table 1.

Group characteristics and summary statistics for outcome measures.

	ASD Group		BAP Group		Comparison Group	
Group Characteristics	***n***	***M* (*SD*)**	***n***	***M* (*SD*)**	***n***	***M* (*SD*)**
Age (years)	18	10.32 (2.88)	22	11.76 (2.68)	135	11.38 (2.45)
BPASS Social Interest	18	3.83 (0.57)[a,b]	22	2.45 (0.69)[a,c]	131	1.89 (0.69)[b,c]
BPASS Expressiveness	18	2.88 (0.51)[a,b]	21	1.96 (0.52)[a,c]	133	1.19 (0.29)[b,c]
BPASS Conversation	18	2.72 (0.94)[a,b]	21	1.90 (0.54)[a,c]	133	1.17 (0.30)[b,c]
BPASS Flexibility/Restricted Interests	18	2.96 (0.62)[a,b]	22	2.29 (0.35)[c]	132	2.06 (0.44)[c]
Verbal IQ	18	82.33 (19.86)[a,b]	22	112.05 (20.71)[c]	135	109.92 (14.31)[c]
Outcome Measures	***n***	***M* (*SD*)**	***N***	***M* (*SD*)**	***n***	***M* (*SD*)**
CASL-2 Pragmatic Language	17	71.29 (14.51)[a,b]	22	102.09 (14.93)[c]	133	109.39 (15.02)[c]
CASL-2 Nonliteral Language	14	75.43 (16.47)[a,b]	20	107.50 (16.02)[c]	132	114.76 (14.24)[c]
CCC-2 GCC	16	90.13 (17.88)[a,b]	19	118.42 (17.89)[c]	128	121.23 (16.93)[c]
CSRS Total Score	18	66.00 (18.45)[a,b]	22	85.68 (7.70)[a,c]	135	93.76 (7.15)[b,c]
CSRS Attentiveness Subscale	18	18.28 (6.76)[a,b]	22	22.82 (3.02)[a,c]	135	25.94 (2.53)[b,c]
CSRS Composure Subscale	18	20.44 (4.48)[a,b]	22	24.82 (2.79)[a,c]	135	26.43 (1.88)[b,c]
CSRS Expressiveness Subscale	18	16.44 (4.66)[a,b]	22	23.50 (2.99)[a,c]	135	26.39 (2.97)[b,c]
CSRS Coordination Subscale	18	18.50 (6.21)[a,b]	22	24.32 (2.63)[c]	135	25.73 (2.64)[c]

Note. ASD = Autism Spectrum Disorder, BAP = Broader Autism Phenotype; BPASS = Broader Phenotype Autism Symptom Scale; CASL-2 = Comprehensive Assessment of Spoken Language, 2nd edition; CCC-2 = Children's Communication Checklist-2; CSRS = Conversational Skills Rating Scale. CASL-2 and CCC-2 GCC scores reflect standard scores.

[a]Indicates significant difference ($p < .05$) from Comparison group.

[b]Indicates significant difference ($p < .05$) from BAP group.

[c]Indicates significant difference ($p < .05$) from ASD group.

Table 2.

Conversational Skills Rating Scale items by subscale.

Subscale	Item
Attentiveness	Lean toward partner (neither too forward nor too far back) Nodding of head in response to partner statements Speaking about partner (involvement of partner as a topic of conversation) Speaking about self (neither too much nor too little) Encouragements or agreements (encouragement of partner to talk) Personal opinion expression (neither too passive nor aggressive) Asking of questions
Composure	Speaking fluency (pauses, silences, “uh”, etc.) Vocal confidence (neither too tense/nervous nor overly confident sounding) Volume (neither too loud nor too soft) Posture (neither too closed/formal nor too open/informal) Shaking or nervous twitches (aren’t noticeable or distracting) Unmotivated movements (tapping feet, fingers, hair-twirling, etc.) Use of eye contact
Expressiveness	Articulation (clarify of pronunciation and linguistic expression) Vocal variety (neither overly monotone nor dramatic voice) Facial expressiveness (neither blank nor exaggerated) Use of gestures to emphasize what is being said Use of humor and/or stories Smiling and/or laughing Use of eye contact
Coordination	Speaking rate (neither too slow nor too fast) Asking of questions Initiation of new topics Maintenance of topics and follow-up comments Interruption of partner speaking turns Use of time speaking relative to partner Speaking fluency (pauses, silences, “uh”, etc.)

Table 3.

Correlations between measures.

	CASL-2 NL	CCC-2 GCC	CSRS Total Score	BPASS Social Interest	BPASS Expressiveness	BPASS Conversation	BPASS Flexibility/Restricted Interests
CASL-2 PL	.71*	.67*	.52*	−.51*	−.58*	−.47*	−.35*
CASL-2 NL		.45*	.52*	−.46*	−.53*	−.48*	−.37*
CCC-2 GCC			.46*	−.45*	−.43*	−.40*	−.38*
CSRS Total Score				−.58*	−.78*	−.74*	−.42*
BPASS Social Interest					.66*	.64*	.53*
BPASS Expressiveness						.75*	.47*
BPASS Conversation							.55*

Note. CASL-2 = Comprehensive Assessment of Spoken Language, 2nd edition, PL = Pragmatic Language, NL = Nonliteral Language; CCC-2 = Children's Communication Checklist-2, GCC = General Communication Composite; CSRS = Conversational Skills Rating Scale; BPASS = Broader Phenotype Autism Symptom Scale.

*$p < .001$.

Table 4.

Multinomial logistic regression model examining the prediction of outcome group.

Parameter Estimates					
				95% CI for Odds Ratio	
	***B* (SE)**	***p***	**Exp (*β*)**	**Lower**	**Upper**
ASD vs. Comparison					
CSRS	−0.19 (0.08)	.021	0.82	0.70	0.97
CASL-2 PL	−0.32 (0.12)	.009	0.72	0.57	0.92
BAP vs. Comparison					
CSRS	−0.10 (0.03)	.004	0.91	0.85	0.97
CASL-2 PL	−0.03 (0.02)	.116	0.97	0.93	1.01
Model Fit Indices					
			Likelihood Ratio Tests		
	−2 Log Likelihood		**Chi-Square**	**df**	***p***
Effect					
CASL-2 PL	134.29		27.35	2	< .001
CSRS	121.67		14.72	2	.001
Model					
Final	106.94		81.81	4	< .001

Note. ASD = Autism Spectrum Disorder, BAP = Broader Autism Phenotype; CSRS = Conversational Skills Rating Scale; CASL-2 = Comprehensive Assessment of Spoken Language, 2nd edition, PL = Pragmatic Language; CI = confidence interval; SE = standard error.