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Pragmatic Language Markers of Autism Diagnosis and Severity

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Abstract

Purpose: Assessment of pragmatic language difficulties is limited with conventional tests but can be performed with informant reports. We evaluated the performance of a parent-completed language scale in differentiating autism from typical development (TD) and another neurodevelopmental disorder. Specifically, we aimed to gauge the respective values of structural and pragmatic language scores for diagnostic discrimination and for predicting severity of social impairment in autistic children.

Method: 174 children aged 7 to 17 (101 with autism, 45 with ADHD, 28 with TD) were evaluated with the ADOS-2 and an abbreviated version of the WISC. Parents completed the Children's Communication Checklist, 2nd Edition (CCC-2) and the Social Responsiveness Scale. CCC-2 mean differences across diagnostic groups were tested with analysis of variance and covariance. Multiple linear regression was used to compare the structural and pragmatic CCC-2 scores in predicting autism symptom severity.

Results: Both structural and pragmatic language scores discriminated between the three diagnostic groups, with stronger effects for the pragmatic scores. Pragmatic scores remained robust predictors of ADHD and ASD diagnoses even after accounting for cognitive and structural linguistic differences. Among autistic children, social impairment severity was associated with pragmatic, but not structural, language profiles.

Conclusions: In order to characterize pragmatic language, easy to administer parent questionnaires such as the CCC-2 may support clinicians who are considering an autism diagnosis and needing to evaluate and monitor social communication.

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Keywords

language assessment; parent report; social communication

Pragmatic language differences are important diagnostic markers for Autism Spectrum Disorder (ASD) (American Psychiatric Association, 2013). Pragmatics have long been considered an important aspect of communication (Tager-Flusberg, 1981; Volkmar et al., 1987) and can be generally described as the way in which context affects use of linguistic constructs including grammar and syntax (Landa, 2000). A person will change their sentence structure, word choice, intonation, and vocabulary depending on a variety of factors including culture, location, familiarity, background knowledge, and shared experiences. Traditional language assessments capture salient details related to the structure of language development; however, challenges remain in assessing social communication and related pragmatic skills. There are few systematic, standardized measures that can quantitatively describe social language based on testing in a formal, clinical environment. Social communication is better observed over time and in natural, familiar environments.

A growing body of research confirms the importance of including parental expertise in evaluations of communication in children across a range of countries (Jensen de López et al., 2021; Väisänen et al., 2014) and notes the benefit of enlisting parent informants who can provide information that is difficult to observe in formal evaluation settings (Ferrara et al., 2020; Norbury et al., 2004). Parent-report measures have been found to capture these differences well (Ghaziuddin et al., 2010; Norbury et al., 2004) and have been shown to be more sensitive to social communication differences than teacher reports (Geurts et al., 2004). Utilizing parent-report, clinicians and diagnostic teams can recognize parents and caregivers as an asset to the assessment process, and further examine aspects of the child's social communication under more realistic conditions.

The Children's Communication Checklist, 2nd Edition (CCC-2) (Bishop, 2003) is a parent-report tool that has been used internationally to measure pragmatic differences well and within a variety of conditions. Validation studies have suggested that CCC-2 results align with formal diagnoses and differentiate between clinical and non-clinical groups (Geurts et al., 2004); Norbury et al. (2004). Additionally, the CCC-2 identifies children with pragmatic impairments among children who have no structural language impairments (Norbury & Bishop, 2005) and can identify pragmatic differences in patients with pediatric traumatic brain injury (Fisher et al., 2020) and autism (Charman et al., 2007). The CCC-2 has also been used in a variety of languages, distinguishing between various neurodevelopmental and communication disorders in Italian (Ferrara et al., 2020), Finnish (Väisänen et al., 2014), Galician (de la Torre Carril & Pérez-Pereira, 2019), Spanish (Andrés-Roqueta et al., 2021), Dutch (Kuijper et al., 2017), English (Staikova et al,. 2013) and Persian (Dadgar et al., 2021) samples.

Previous literature has established a precedent for continued exploration of differentiating clinical groups using the CCC-2. Some studies have found significant differences between groups of children with different neurodevelopmental conditions. Geurts and colleagues (2004) found that CCC-2 scores could differentiate between autism and typical development and also between autism and attention deficit hyperactivity disorder (ADHD), with significant differences across most subscales. De la Torre Carril and colleagues (2021) recently found similar results with CCC-2 scores differentiating between TD and ASD, ADHD, as well as between ASD and ADHD. In both studies, the ADHD group scores were in between those of the children with ASD and TD. Vaïsaïnen and colleagues (2014) examined the CCC-2 to evaluate potential differences between TD and ADHD groups of children. Results indicated that children with ADHD, even with no diagnosed language impairment, performed more poorly than TD children on all subscales.

The CCC-2 provides both a composite score for a child's linguistic structure (syntax, morphology, vocabulary) and a composite score for their social communication (pragmatics). As stated above, pragmatics can influence other areas of linguistic development, so there is a somewhat arbitrary distinction between these linguistic categories. The structural and pragmatic components of the CCC-2 represent one way in which language constructs may be viewed. The Structural Composite include Coherence, for example, which could arguably be a Pragmatic domain. Similarly, the Structural composite includes Semantics, which depends on context for accuracy. Discussions of pragmatic influence on linguistic structure are ongoing ((Norbury, 2014); for the purpose of this investigation, we use the Structural and Pragmatic composites as they were developed for use in the CCC-2 (Bishop, 2003). The Structural composite includes items related to a child's proper use of grammatical constructions, accurate vocabulary, clear speech, and narrative ability. The Pragmatic composite includes items related to initiation, repetitive language, and nonverbal communication.

Investigators have documented several important differences between structural and pragmatic scores. Geurts & Embrechts (2008) found that autistic children and children with ADHD have more difficulty on the pragmatics scale than on the structural scale. Ferrara and colleagues (2020) similarly evaluated the CCC-2 composites, finding that all clinical groups in their study performed lower than controls on the structural scale, and the pragmatic scale was able to differentiate ASD from dyslexia and developmental language disorder, as well as TD. Volden and Phillips (2010) used the CCC-2 to examine children who had structural scores within normal limits and found that the CCC-2 identified pragmatic impairments better than the Test of Pragmatic Language (Phelps-Terasaki & Phelps-Gunn, 2007). Other researchers found that the CCC-2 differentiated between disorders that are characterized linguistically as primarily structural (e.g., Down Syndrome) and disorders that are primarily pragmatic (e.g., autism and ADHD). Verté and colleagues (2006) found little CCC-2 score differentiation between older autism classifications (e.g., high functioning autism, Asperger's syndrome (DSM-IV, 2000)). They did, however, provide early support for use of the CCC-2 as a potential indicator for level of severity in ASD based on patterns of language profiles observed across the previous subtypes. Further examination of CCC-2 differences in ADHD and ASD comes from Kuijper and colleagues (2016) who found TD children scored differently than children with ADHD and ASD; however, they found only pragmatic and not structural differences between the ADHD and ASD groups. The pragmatic and structural indices are clearly inter-related, and there is no clear understanding yet of the individual contributions of each or how they relate to one another in the context of this instrument.

In this study, we examined a large group of children with ASD that was compared to two control groups, one with TD and one with ADHD. While ADHD in isolation is not considered a communication disorder, pragmatic differences are well documented in children with ADHD (Staikova et al., 2013). ADHD symptoms can interfere with communication and mimic the symptoms of ASD, and ADHD is carefully considered during a differential diagnosis of ASD. Thus, inclusion of a comparison group with ADHD alongside TD controls allowed us to further examine the specificity of pragmatic language differences with respect to ASD.

In this study, we provided replication of the work of Geurts and colleagues (2004) and extend the results by evaluating the respective contributions of various linguistic processes to these differences. Our intention was not to demonstrate clinical validity of the CCC-2 as a screening or diagnostic tool but rather to evaluate language differences broadly between the three groups. The specific objectives of this study were: 1) to evaluate group difference in scores on the CCC-2 in autistic children, children with typical development, and non-autistic children with ADHD; 2) to evaluate the respective contribution of structural language and pragmatic language scores in discriminating across conditions; and 3) to assess within children with ASD the relationship of both pragmatic and structural language scores to autism severity while controlling for background developmental factors.

Methods

Participants

174 children aged 7 to 17 were recruited for a larger study involving functional magnetic resonance imaging by community outreach and referrals from a regional university specialty clinic. Potential participants came in for a screening visit to determine if they qualified for the study. During this initial visit, informed written consent and assent was obtained from all participants, as required, and their parents. All children in the ASD and ADHD groups were directly assessed by experienced child psychiatrists and clinical psychologists who confirmed their diagnosis based on DSM-5 criteria (American Psychiatric Association, 2013). The research diagnostic team reviewed results of the standardized diagnostic assessments (both videos and scored protocols) and used best estimate procedures. ASD was ruled out in the TD and ADHD groups based on the ADOS-2, clinical interview, and parent-completed autism questionnaires (see below). Exclusion criteria for all groups included the presence of seizure disorder, cerebral palsy, pediatric stroke, history of chemotherapy, sensorimotor disabilities, closed head injury, thyroid disorder, schizophrenia, bipolar disorder, current major depressive episode, fetal alcohol syndrome, Tourette’s disorder, severe vision impairments, Rett’s syndrome, current use of psychoactive medications, non-English speaker, or an IQ below 70. The study sample included the following children: 101 with ASD (mean age: 11.3 years; 85 males), 28 with TD (mean age: 11.6 years; 12 males), and 45 with ADHD (mean age 11.5 years; 31 males); see Table 1. When we planned this study, the neuroimaging study was closed to recruitment and we had therefore no opportunity to recruit additional participants with ADHD or ASD or TD controls. Note that TD controls were recruited from normal school settings, had no developmental concern, no

co-occurring psychiatric disorder and were not children who had failed to meet inclusion criteria for the two clinical groups.

Instruments

Autism diagnosis.—The Autism Diagnostic Interview-Revised was administered with caregivers of all potential ASD participants. Developmental history was collected as well as the Social Responsiveness Scale 2nd edition (SRS-2 (see below); Constantino and Gruber, 2012). The Autism Diagnostic Observation Schedule, 2nd Edition (ADOS-2) (Lord et al., 2012) is a semi-structured, standardized assessment in which a trained examiner engages participants in activities that are designed to elicit social and communication behaviors indicative of symptoms of ASD as defined in the DSM-5 (American Psychiatric Association, 2013). In this study, all participants were administered the Module 3 of the ADOS-2, designed for children and adolescents with fluent speech. Module 3 comprises 14 tasks that are generally administered in sequence although the tester has some flexibility to change the task order if clinically indicated. All ADOS-2 interviews were administered by research assistants or a senior clinical psychologist trained to research reliability level. ADOS-2 scoring yields a Social Affect (SA) score (10 items; range 0-20), a Restricted and Repetitive Behavior (RRB) score (4 items; range 0-8), and a Total score that is the sum of SA and RRB (14 items; range: 0-28). In addition, a Comparison Score (CS; range 1-10) can be computed that estimates the overall clinical severity of autism using normative data. Higher values on the ADOS-2 scores indicate more severe ASD symptoms. Autism diagnosis was derived after review of all research and clinical data by two experienced clinicians.

ADHD diagnosis.—Children with ADHD referred to the study participated in a clinical evaluation that was conducted with the standardized, well-normed Conners' Rating Scales-3rd Edition short form (Conners, 2003) and the ADHD Rating Scale (DuPaul et al., 1998) for parent and teacher, the parent semi-structured diagnostic interview Kiddie Schedule for Affective Disorders and Schizophrenia (Puig-Antich and Ryan, 1986), and clinical observation. Best estimate research diagnoses were obtained by two independent experienced assessors (kappa’s>.80). All participants in the ADHD group met DSM-IV-TR ADHD criteria for ADHD, either the inattentive, the hyperactive or combined type. Co-occurrence of ASD and ADHD was allowed. In the ASD group, 60 children (59.4%) also met criteria for ADHD mostly of the combined type (N=44). None of the TD children met ADHD criteria.

Autism symptomatology and severity.—The Social Responsiveness Scale, Second Edition (SRS-2) (Constantino & Gruber, 2012) assesses the presence and severity of social impairment related to ASD. The SRS-2 is an informant-completed and age-dependent scale, with separate forms to evaluate school-aged children (ages 4-18 years), preschoolers (ages 2.5-4.5 years) and adults (ages 19 years and older). Parents rate 65 statements about their child’s social skills as “Not true,” “Sometimes true,” “Often true,” or “Almost always true.” The questions are grouped into multiple subdomains for scoring Social Awareness, Social Cognition, Social Communication, Social Motivation, and Autistic Mannerism; the five subdomain raw scores are summed for the total score, which is then translated to a T-score. Reliability and validity of the SRS-2 have been extensively tested and found to be excellent.

The SRS-2 was normed using five different studies, with 1600 children in the total sample. In this study, we used the total t-score.

Intellectual level.—Intellectual level of participants was estimated with a short form of the Wechsler Intelligence Scale for Children 4th Edition (WISC) (Wechsler, 2003). Three subtests were administered: Information, Block Design, and Vocabulary, allowing a full-scale IQ to be estimated from the sum of scaled scores of the three subtests according to the formula in the WISC supplement (Sattler & Dumont, 2004). This short form the WISC did not allow us to estimate separate verbal and non-verbal IQ.

Language development.—Language characteristics and linguistic pragmatic abilities were assessed using the parent-completed Children's Communication Checklist, second edition (CCC-2) (Bishop, 2003). The CCC-2 is a widely-used, 70-item standardized checklist of pragmatic and social communication behaviors applicable to children ages 4:0 to 16:11. Caregivers are asked to make a frequency judgment about how often behaviors occur on 4-point scale. The CCC-2 is divided into 10 subscales measuring: (A) speech, (B) syntax, (C) semantics, (D) coherence, (E) inappropriate initiation, (F) stereotyped language, (G) the use of context, (H) non-verbal communication, (I) social relationships, and (J) interests. The first four subscales (A–D) evaluate articulation and phonology, language structure, vocabulary, and discourse; four other subscales (E-H) evaluate pragmatic aspects of communication as well as stereotyped language with atypical or unusual expressions and use of nonverbal communication like facial expressions, bodily movements, and gestures. The last two subscales (I and J) measure behaviors characteristic of children with ASD. Each subscale raw score is converted to age-standardized scores (mean=10; SD=3). A General Communication Composite (GCC) is derived by summing scores A to H and has been standardized for the US child (4-16) population (mean=100; SD=15). A Structural Language composite score is derived by averaging scores A to D, and a Pragmatic Language composite score is obtained by averaging scores E to H. Lower scores are indicative of more language difficulties. In our analyses, we include 11 scores: 8 subscales (A-H) and the three composite scores (general, structural, and language). Another score, the Social Interaction Difference Index (SIDI) can be derived from the CCC-2; however, as it relies on the subscales I and J that are direct measures of autistic symptoms and behaviors, we did not use the SIDI or the subscales I and J scores in this study to retain language measures that are independent from autism symptom measurement.

As described in the introduction, there is some degree of overlap within the Structural and Pragmatic domains on the CCC-2. To demonstrate the way the CCC-2 differentiates these two domains, we provide a few examples of items that are included in the Semantics (Structural) domain: making false starts/searching for the right word, mixes up words with similar meaning, mixes up words with similar phonology, uses non-specific language, uses abstract language, and uses categorical language. Some examples of items in the Coherence domain (Pragmatics) include: use of non-referential antecedents, story sequencing, use of background information, and making logical sense. Nevertheless, for the purpose of this study, we did adhere to the original recommendations and scoring rules suggested by the developers of the CCC-2.

Statistical Analyses

Conventional statistical tests (chi-square) were used to compare groups for categorical variables. Internal consistency for CCC-2 subscale scores was computed with Cronbach's alpha coefficient. To account for unequal sample sizes and unequal variances, mean differences across the 3 groups for ADOS-2, SRS-2, IQ and CCC-2 scores were first tested with one-way between-subjects ANOVAs and *p*-values estimated from robust *F* Brown-Forsythe statistics. Follow-up pairwise comparisons were performed with Games-Howell tests to account for unequal sample sizes and unequal variances. Effect sizes were calculated with eta-squared (η^2).

Two sets of analysis of covariance (ANCOVA) were subsequently performed with CCC-2 scores as dependent variables. First, mean differences in CCC-2 scores across groups were examined in ANCOVA models where full-scale IQ was entered as covariate. Effects sizes for the diagnostic independent variable were calculated with partial η^2. Post-hoc pairwise comparisons of estimated marginal means were performed with the least square method. Second, in order to evaluate the ability of Pragmatic CCC-2 scores to differentiate groups controlling for structural language group differences, five ANCOVAs models were estimated with 4 subscale (E to H) and the total Pragmatic CCC-2 scores as dependent variables and IQ and the CCC-2 Structural language composite score as covariates.

Finally, in order to test if CCC-2 scores were associated with autism severity, we used multiple linear regression in two separate models using either the SRS-2 total t-score or the ADOS-2 total score as dependent variables. These analyses were restricted to participants in the ASD group. Age, FSIQ, presence of co-occurring ADHD and the Pragmatic and Structural language composite scores of the CCC-2 were entered as independent variables. Standardized residuals were inspected for normality. Assumptions of homoscedasticity were checked using plots of standardized predicted values against standardized residuals and correlations between standardized predicted values and absolute values of standardized residuals. Absence of multicollinearity was verified for each independent variable, verifying that tolerance and variance inversion factor (VIF) did not cross thresholds set at >.10 and <10, respectively. Potentially influential outlier observations were detected with Mahalanobis distances and Cook's *D*. Throughout, a *p*-value of 0.01 was set a priori as the level for statistical significance.

Ethical Approval

This study was approved by the university Institutional Review Board. Caregiver written consent and participant's assent were obtained prior to participation.

Results

Sample sociodemographic characteristics are summarized in Table 1. There was no group difference for age, race and ethnicity. Not surprisingly, sex was significantly associated with diagnostic group owing to the known male preponderance in the ASD and ADHD groups. CCC-2 scale scores had excellent internal consistency, with α ranging from .75 to .87 for the eight 7-items subscales A to H; the structural and pragmatic composite scores achieved very high consistency (α=.92 and α=.95, respectively). Intercorrelations between subscales were all significant ($p < .001$) and Pearson's r's ranged from .443 to .811 (median r = .693). The correlation between the Structural and Pragmatic composite scores was high (r = .811; $P < .001$). Figure 1 displays the distribution of these 2 CCC-2 composite scores across diagnostic groups.

Clinical features of the 3 groups are summarized in Table 2. On all measures except the Block Design subtest of the WISC, the ASD group obtained significantly lower scores than the other 2 groups. As expected, all ADOS-2 scores were significantly higher in the ASD group than in the 2 comparison groups. Cognitive scores were significantly lower, but still within the average range, for the ASD group compared to TD and ADHD groups. With the exception of the SRS-2, the ADHD and TD groups did not differ significantly from each other in post-hoc tests. The ADHD group had slightly elevated SRS-2 scores compared to TD children but the mean for the ADHD group remained well below the recommended SRS-2 thresholds of 60 and 75 for clinical significance.

As expected, there were significant ($P < .01$) Pearson correlations between the CCC-2 structural language and the WISC Vocabulary standard score (.333 for the Speech subscale; .502 for the Syntax subscale; .495 for the Semantics subscale; .523 for Structural CCC-2 score).

Aim 1: Group Differences in CCC-2 Scores

For all eleven CCC-2 scores, ANOVA models revealed highly significant ($p < .001$) between-groups differences (Table 3; ANOVA column). Games-Howell post-hoc tests revealed that on all CCC-2 scores, children with ASD scored significantly lower than children with ADHD and TD. With the exception of two structural scales (i.e., Speech and Syntax), the ADHD group scored significantly lower than TD, and their scores fell between the ASD and TD groups albeit consistently closer to the TD group.

Aim 2: Structural and Pragmatic Language Score Contribution to Differentiation

Effects sizes were larger for pragmatic scaled scores (η^2: .56 - .65) than structural scaled scores (η^2: .20 - .47). Likewise, the effect size was much larger for the Pragmatic composite than for the Structural composite (.716 vs .444). In fact, from all CCC-2 between-group comparisons, the Pragmatic composite score achieved the strongest differentiation (η^2: .716), even when compared to the GCC total score (η^2: .640).

Covarying FSIQ slightly reduced the effect size of CCC-2 differences across groups (Table 3; ANCOVA 1). The reduction in η^2 was more pronounced for the structural CCC-2 scores as compared to the pragmatic scores. Across all models, FSIQ accounted for relatively small amounts of variance in the CCC-2 scores (partial η^2: .004 - .117). FSIQ was a significant predictor in the 4 ANCOVAs with structural subscales (partial η^2: .048 - .103), the Structural composite score (partial η^2=.117) and the total GCC score of the CCC-2 (partial η^2=.079). By contrast, FSIQ exerted a significant and small effect on one pragmatic subscale only (Context; partial η^2=.040; p = .01) and had no significant effect on the other 3 pragmatic subscale scores (partial η^2: .004 - .014; all NS) or on the Pragmatic composite score (partial η^2=.022; NS).

When structural scaled scores were covaried alongside FSIQ (Table 3; ANCOVA 2), pragmatic scaled scores remained strongly and significantly different across diagnostic groups, with effect sizes ranging from .302 to .359 for the 4 subscale scores; the effect size for the pragmatic composite score was very large and highly significant (partial η^2 = .493; p=.008). In the 5 ANCOVAs, there were statistically significant effects of the structural CCC-2 scores (partial η^2: .136 - .380; all p's<.001) whereas FSIQ remained non-significant in all analyses (partial η^2: .000- .015; all NS).

The abbreviated FSIQ was heavily dependent on verbal ability scores (WISC Vocabulary and Information sub-tests) and was significantly (p <.001) correlated with both the CCC-2 Structural composite (r=.478) and the Pragmatic composite (r=.375). Thus, it was possible that adjusting on the FSIQ had left little or no variance to be explained by the CCC-2 Structural language score in the ANCOVAs. Thus, we repeated Table 3 analyses using the Block Design standard score as a measure of IQ (instead of FSIQ) in the ANCOVAs. Block design had lower correlations with the CCC-2 Structural composite (r=.215) and Pragmatic composite (r=.109). However, results remained identical and full details are presented in Supplemental Table S1.

Aim 3: Relationship of Structural and Pragmatic Scores to Autism Severity

In the autism subsample, the multiple regression analysis using SRS-2 Total t-score to index autism severity showed a significant regression model with 55.2% of the SRS-2 variance accounted for by the 5 explanatory variables (Table 4; Model 1, top). Age, IQ, presence of co-occurring ADHD and the CCC-2 Structural composite score had small standardized β coefficients that were not significantly different from zero. By contrast, a large, negative and significant coefficient was found for the CCC-2 Pragmatic composite score (β =−.710; t = −7.59; p <.001). Squared semi-partial correlation coefficients indicated that 28.9% of the SRS-2 variance was uniquely accounted by the CCC-2 Pragmatic score; the corresponding value for the CCC-2 Structural score was negligible (.2%). The regression model with the ADOS-2 score as dependent variable was also significant (Table 4; Model 2, bottom) although, compared to the previous model, less variance (32.9%) in the autism severity measure was explained jointly by the 5 explanatory variables. FSIQ significantly contributed to the model (β = −.527; p <.001) and uniquely accounted for 23.3% of the ADOS-2 total variance. The unique contributions of the CCC-2 scores were lower (Structural: 0.26%; Pragmatic: 4.88%), non-significant for the Structural scale score and significant for the Pragmatic score at the set threshold of .01. In either model, the presence or absence or co-occurring ADHD was not significant and did not change the relationship of both CCC-2 scores to the dependent variables. Assumptions of normality of residuals and homoscedasticity were verified in both models; as well, there was no evidence of multicollinearity or of outliers influencing the results. The two models in Table 4 were re-evaluated using Block Design instead of FSIQ as a predictor; the results were unchanged (data not shown).

Discussion

Our findings confirm that a parent-based measure of child's everyday language represents a good supplement to a comprehensive clinical evaluation of communication. The general goal of this investigation was to meaningfully differentiate children, not diagnose, using the CCC-2. Our first objective was to evaluate group difference in scores on the CCC-2 in autistic children, children with typical development, and non-autistic children with ADHD. In our sample, the CCC-2 scores differentiated children with ASD not only from TD controls but also from participants with ADHD, confirming the results of a 2004 study by Geurts and colleagues. A direct comparison with Geurts et al.'s study is hampered by the fact that these authors used a prior version of the CCC that did not have the same scoring instructions and subscale structure. However, like in our study, they reported no differentiation for the two structural language subscales of Speech and Syntax between the ADHD and control groups whereas scores of the ASD participants on these two scales were significantly lower than those of the two comparison groups. By contrast, for other subscales, participants with ADHD could be differentiated from both the TD controls and the ASD participants with their mean scores falling in between with the exception of the Initiation subscale that failed to separate ASD and ADHD (whereas it did in our study). Likewise, in our study, children with ADHD had significantly poorer language scores than TD participants on two of the four structural and all pragmatic subscales; however, compared to TD controls, the magnitude of difficulties in children with ADHD was less pronounced than that for children with ASD. Of note, all CCC-2 scores discriminated the three diagnostic groups with the pragmatic scores yielding the strongest effects. These results are consistent with prior work (Ferrara et al., 2020; Norbury et al., 2004; Volden & Phillips, 2010) and further demonstrate the utility of this caregiver-informed instrument.

Our second objective was to evaluate the respective contribution of structural language and pragmatic language scores in discriminating across conditions. When the effects of IQ were taken into account, the discriminant performance of CCC-2 scores remained; however, it was notable that the attenuation of effect size was much more pronounced for structural than for pragmatic scores. In fact, adjusting on IQ had only a minimal effect on the discriminant capacity of pragmatic scores both at the fine-grained subscale level and at the composite level. Taken together, the results indicate that the language features measured by the structural CCC-2 scores are more dependent upon developmental and cognitive levels of children than upon pragmatic scores, and indeed the pragmatic scores appeared less contingent upon the child's IQ. This characteristic of pragmatic language features as measured by the CCC-2 make it an especially desirable measure to evaluate pragmatics in the context of neurodevelopmental disorders, and particularly among children with ASD who tend to have greater cognitive variability and lower average intellectual levels (Charman et al., 2011).

Our third objective was to assess within children with ASD the relationship of both pragmatic and structural language scores to autism severity while controlling for background developmental factors. As reported previously with the CCC-2, we found that structural and pragmatic language scores were highly inter-correlated (Andrés-Roqueta et al., 2021; Andrés-Roqueta & Katsos, 2017) making it difficult to disentangle their effects in interpreting associations of CCC-2 scores with those from external measurements. To the best of our knowledge, this study was the first to investigate the relative benefits of the structural and pragmatic CCC-2 scores in predicting two external criteria (diagnosis and autism severity). Two findings are worth noting. First, when group differences in structural language and IQ were taken into account in covariance analysis, the performance of CCC-2 pragmatic scores was maintained and robust differentiation between the 3 diagnostic groups was preserved. Second, when examining the association of both structural and pragmatic scores with autism severity in a multiple regression model, pragmatic language difficulties were strongly and positively correlated with higher levels of social impairment whereas no such association was detected for structural scores. The combined results suggest that pragmatic scores of the CCC-2 contribute the most relevant clinical language information for providers involved in differential autism diagnosis and autism severity decisions.

An additional finding of our study was to document the strong psychometric properties of the CCC-2 as an informant measure. As discussed above, its validity was demonstrated in the very large effect sizes associated with all its scores in between-group comparisons. Moreover, the reliability of the subscale scores as measured by the internal consistency was consistently high for all eight component subscales. Given that each subscale comprised 7 items only, this result is noteworthy. Moreover, the two composite language scores achieved much higher levels of internal consistency than could be predicted by the higher number of items (N=28) included in each composite. The careful construction of the CCC-2 (with 2 of the 7 items of each subscale being positively phrased, and subscale-specific items being spread across the instrument to avoid halo effects) is likely responsible for the remarkable psychometric qualities. These properties are of interest in two applications. Researchers can expect to have improved power to detect association between CCC-2 language scores and independent measures since attenuation of true association should be minimal with its high internal consistency. Clinicians could also be confident that this relatively brief and inexpensive measurement can provide them with reliable and quantitative indices of language that have meaningful value for both diagnosis and management purposes.

Limitations

Due to the exclusionary criteria of the larger study in which our participants were enrolled, we did not have access to children with intellectual disability in our sample. It is an important goal in autism research to be inclusive in studies, so future work will strive to better represent the range of ability in autism spectrum conditions. Nevertheless, we did have a range of intellectual ability in our sample, and the scores were only minimally affected when the IQ was covaried. This provides a reason to suspect there may be similar findings across a wider range of intellectual ability. Due to sample size constraints, we could not conduct subgroup analyses to examine separately effects due to gender, race or ethnicity. Additionally, the study design was observational and cross-sectional precluding a causal interpretation of observed associations. Due to the nature of parent report, there may be some degree of correlated error due to some measures being reported by the same informants. Finally, the CCC-2 was developed prior to the introduction of the DSM-5 and may not be fully in congruence with the ideas of social communication in the current version.

Conclusions

We note that the CCC-2 is an easy to use, affordable measure that has the ability to quantify the subtleties of language use in context. The Pragmatic composite can discriminate effectively between neurodevelopmental diagnostic categories and predict the severity of autism. For our population, these differences were largely independent of the child's IQ, indicating that the CCC-2 may be a valuable tool for clinical use and could be an effective outcome measure in intervention research. Our results also indicate that even in a sample of "verbally fluent" autistic children, there are low scores in both structural and pragmatic domains, indicating a need for communication support across domains.

Importantly, our results offer continued support for enlisting parent-informants to supplement our evaluations of communication and social interaction in children, confirming that parents and caregivers provide valid insights for assessment which are tightly linked to diagnostic conclusions. An increased focus on patient-centered care in healthcare settings is reflected in the literature emphasizing the importance of therapeutic relationships for achieving favorable treatment outcomes (Plexico et al., 2010). Utilization of parent-report measures offers a tangible validation for families, indicating they are valued and essential in their child's assessment procedures (Jensen de López et al., 2021). Furthermore, building respect and responsiveness to family input allows greater opportunity for considering cultural and linguistic diversity. This inclusion encourages families to further explore their concerns, share additional information, or identify cultural variants which may inform and validate diagnostic impressions.

Supplementary Material

Refer to Web version on PubMed Central for supplementary material.

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Highlights

- An easy to use, affordable measure has the ability to quantify the subtleties of language use in context.
- Pragmatic scores remained robust predictors of ADHD and ASD diagnoses even after accounting for cognitive and structural linguistic differences.
- Pragmatic scores of a parent-report measure contribute the most relevant clinical language information for providers involved in differential autism diagnosis and autism severity decisions.
- Our results offer continued support for enlisting parent-informants to supplement our evaluations of communication and social interaction in children.

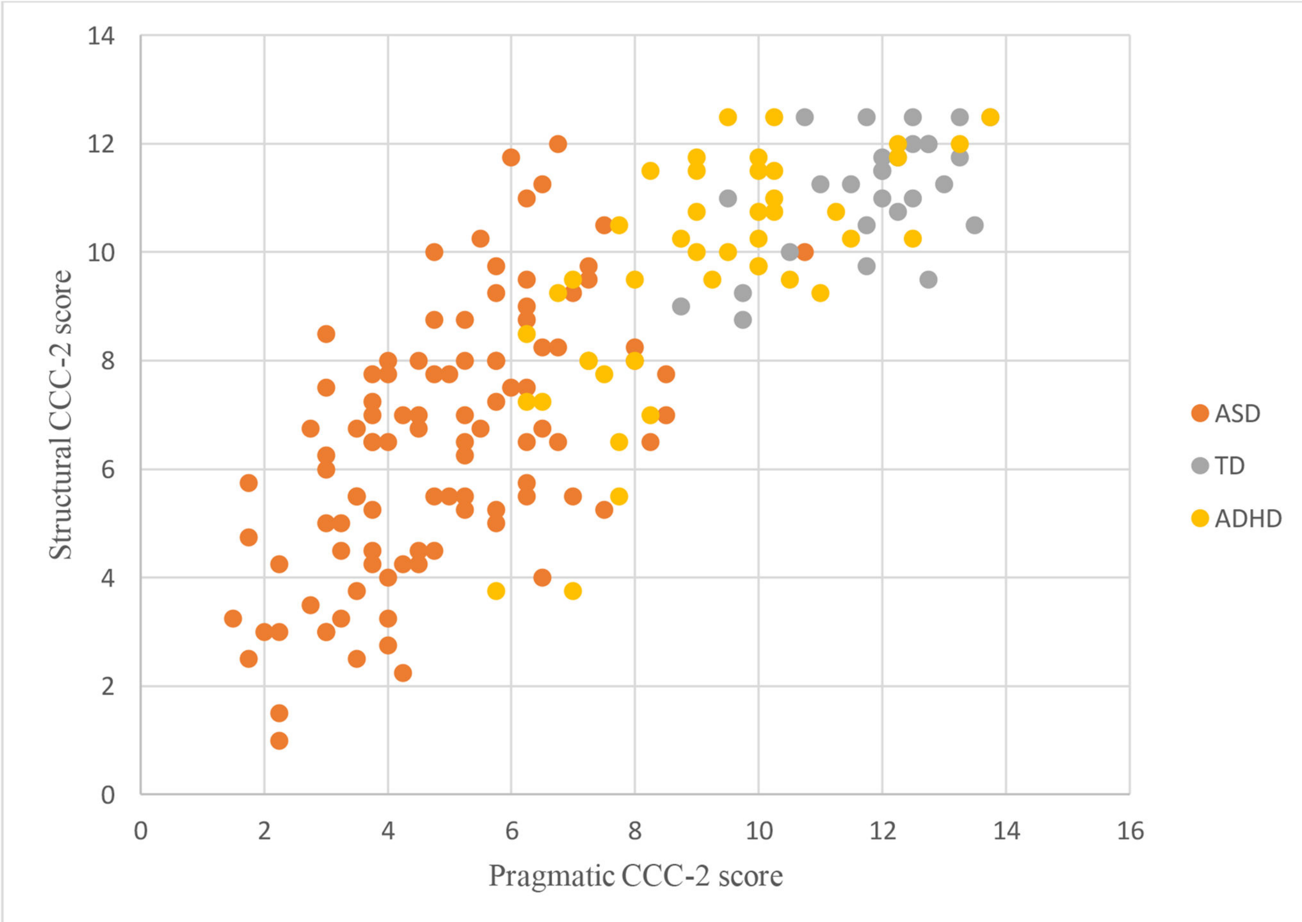

Figure 1.
Correlation between Structural and Pragmatic CCC-2 Scores, by Diagnostic Group

Table 1

Participants' Sociodemographic Characteristics

	ASD	TD	ADHD	Test	*p*
	***n* = 101**	***n* = 28**	***n* = 45**		
Sex, *n* (%)					
Male	85 (84.2)	12 (42.9)	31 (68.9)	χ^2=19.9	<.001
Female	16 (15.8)	16 (57.1)	14 (31.1)		
Age *X (SD)*	11.29 (2.27)	11.61 (1.73)	11.46 (1.61)	*F* = .319	.73
Race and Ethnicity					
American Indian/Alaska Native	3 (3.0)	0			
Asian	0	0		*white vs. non-white*	
Black	0	0		χ^2=0.16	.92
Hawaiian/Pacific Islander	1 (1.0)	0			
White	81 (80.2)	24 (85.7)	37 (82.2)		
More than one race	13 (12.9)	3 (10.7)	5 (11.1)		
Hispanic	16 (15.8)	5 (17.9)	3 (6.7)	χ^2=3.44	.49

Table 2

Participants' Clinical Characteristics

	ASD *n* = 101	TD *n* = 28	ADHD *n* = 45	F^{a}	*p*	ANOVA Pairwise comparisons[b]
ADOS-2 scores, $\bar{X}$ (SD)						
Social affect (SA)	9.79 (3.8)	1.04 (1.9)	1.29 (1.4)	293.3	< 0.001	ASD>TD, ADHD
RRB score	3.46 (1.7)	0.52 (0.7)	0.42 (0.6)	207.1	< 0.001	ASD>TD, ADHD
Total ADOS-2	13.25 (3.9)	1.56 (2.3)	1.71 (1.7)	440.1	< 0.001	ASD>TD, ADHD
Comparison score	7.62 (1.5)	1.22 (1.0)	1.27 (0.6)	803.9	< 0.001	ASD>TD, ADHD
SRS-2 scores, $\bar{X}$ (SD)						
Total T-score	77.7 (10.7)	44.0 (4.1)	53.9 (8.6)	285.9	< 0.001	ASD>ADHD>TD
WISC, $\bar{X}$ (SD)						
Full scale IQ	98.8 (19.5)	113.4 (12.3)	111.6 (13.8)	17.6	< 0.001	ASD<TD, ADHD
Block design scaled	10.8 (3.3)	11.3 (3.4)	11.9 (3.0)	1.9	.151	
Information scaled	9.5 (3.7)	12.4 (2.8)	11.6 (2.7)	13.9	< 0.001	ASD<TD, ADHD
Vocabulary scaled	9.7 (3.9)	13.2 (2.7)	12.3 (2.8)	19.5	< 0.001	ASD<TD, ADHD

Note:

[a:] Brown-Forsythe F

[b:] Games-Howell tests; '< 'or '>' denotes a significant (<0.01) mean difference between groups, a ',' denotes no significant difference.

Table 3

Language Profiles on the CCC-2, by Diagnostic Group.

CCC-2 scaled scores, X (SD)	**ASD** ***n* = 101**	**TD** ***n* = 28**	**ADHD** ***N*= 45**	**ANOVA** F^{a}, *P*, η^2 **post-hoc tests**[b]	**ANCOVA 1**[§] F^{c}, *P*, **partial** η^2 **post-hoc tests**[d]	**ANCOVA 2**[§§] F^{c}, *P*, **partial** η^2 **post-hoc tests**[d]
GCC	45.21 (14.9)	91.89 (8.5)	75.22 (15.1)	187.6; < 0.001;0.640 ASD<ADHD<TD	116.8; < 0.001;0.586 ASD<ADHD<TD	–
A: Speech	7.16 (3.6)	10.68 (2.0)	10.20 (2.8)	30.4; < 0.001;0.201 ASD<ADHD, TD	11.2; < 0.001;0.119 ASD<ADHD, TD	–
B: Syntax	7.06 (3.0)	11.11 (1.9)	10.11 (2.5)	45.6; < 0.001;0.290 ASD<ADHD, TD	21.9; < 0.001;0.210 ASD<ADHD, TD	–
C: Semantics	6.38 (2.6)	11.43 (1.7)	9.40 (2.7)	63.6; < 0.001;0.387 ASD<ADHD<TD	37.5; < 0.001;0.313 ASD<ADHD<TD	–
D: Coherence	5.16 (2.8)	11.29 (1.7)	8.82 (2.6)	92.5; < 0.001;0.467 ASD<ADHD<TD	55.9; < 0.001;0.404 ASD<ADHD<TD	–
E: Initiation	5.14 (2.4)	12.50 (2.3)	8.62 (2.7)	107.6; < 0.001;0.559 ASD<ADHD<TD	85.4; < 0.001;0.509 ASD<ADHD<TD	35.4; < 0.001;0.302 ASD<ADHD<TD
F: Scripted Language	5.41 (2.3)	11.96 (1.5)	9.40 (2.0)	164.7; < 0.001;0.608 ASD<ADHD<TD	106.9; < 0.001;0.564 ASD<ADHD<TD	41.3; < 0.001;0.335 ASD<ADHD<TD
G: Context	4.97 (2.4)	11.79 (1.6)	9.98 (2.0)	196.9; < 0.001;0.645 ASD<ADHD<TD	118.1; < 0.001;0.589 ASD<ADHD<TD	45.9; < 0.001;0.359 ASD<ADHD<TD
H: Nonverbal Communication	3.93 (2.3)	11.14 (1.6)	8.69 (2.8)	145.3; < 0.001;0.611 ASD<ADHD<TD	108.1; < 0.001;0.567 ASD<ADHD<TD	41.1; < 0.001;0.334 ASD<ADHD<TD
Structural composite score (A, B, C, D)	6.44 (2.4)	11.12 (1.1)	9.63 (2.1)	93.0; < 0.001;0.444 ASD<ADHD<TD	46.9; < 0.001;0.363 ASD<ADHD<TD	–
Pragmatic composite score (E, F, G, H)	4.86 (1.8)	11.85 (1.3)	9.17 (2.0)	240.4; < 0.001;0.716 ASD<ADHD<TD	170.2; < 0.001;0.674 ASD<ADHD<TD	79.8; < 0.001;0.493 ASD<ADHD<TD

Notes:

[§:] with IQ as covariate

[§§:] with IQ and the CCC-2 Structural composite score as covariates

[a:] Brown-Forsythe *F*

[b:] Games-Howell tests; ‘<‘or ‘>’ denotes a significant (<0.01) mean difference between groups, a ‘,’ denotes no significant difference

[c:] *F* associated with Diagnostic group in ANCOVA

[d:] pairwise comparisons of estimated marginal means with Least Significant Difference.

Table 4

Autism Severity and CCC-2 Structural and Pragmatic Composite Scores.

	Unstandardized	Standardized			Correlations			Collinearity	
	B (SE)	***β***	***t***	***p***	**Zero-order**	**Partial**	**Semi-Partial**	**Tolerance**	**VIF**
Model 1: SRS-2 total T-score									
$R^2 = .552$, $F(5,89) = 21.967$, $p < .001$									
Constant	92.39 (5.97)	–	15.48	< 0.001	–	–	–	–	–
FSIQ	0.06 (0.42)	.108	1.39	.17	−0.001	.146	.099	.839	1.192
Age in years	0.11 (0.35)	.023	.324	.75	−0.025	.034	.023	.967	1.034
Co-occurring ADHD	.32 (1.70)	.015	.189	.85	.281	.020	.013	.846	1.181
CCC-2 Structural Score	−0.27 (0.43)	−0.059	−0.61	.54	−0.442	−0.065	−0.044	.545	1.836
CCC-2 Pragmatic Score	−4.19 (0.55)	−0.710	−7.59	< 0.001	−0.736	−0.627	−0.538	.574	1.743
Model 2: ADOS-2 total score									
$R^2 = .329$, $F(5,90) = 8.81$, $p < .001$									
Constant	25.62 (2.46)	–	10.41	< 0.001	–	–	–	–	–
FSIQ	−0.097 (0.02)	−0.527	−5.59	< 0.001	−0.520	−0.507	−0.483	.837	1.194
Age in years	−0.019 (0.14)	−0.012	−0.13	.89	−0.063	−0.014	−0.012	.967	1.035
Co-occurring ADHD	−0.952 (0.70)	−0.129	−1.37	.17	.047	−0.143	−0.118	.841	1.188
CCC-2 Structural score	.105 (0.18)	.069	0.59	.56	−0.270	.062	.051	.540	1.851
CCC-2 Pragmatic score	−0.582 (0.23)	−0.293	−2.56	.01	−0.269	−0.260	−0.221	.571	1.752