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Pragmatic Language Profiles of School-Age Children With Autism Spectrum Disorders and Williams Syndrome

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Abstract

Purpose—To describe and compare the pragmatic language profiles of school-age children with autism spectrum disorders (ASD) and Williams syndrome (WS) on a standardized measure to determine whether a standard pragmatics tool can differentiate between 2 groups of children with opposing social presentations and pragmatic language difficulties.

Method—Twenty-two parents of school-age children with ASD, 21 parents of school-age children with WS, and 19 parents of school-age typically developing children rated their child on the Children's Communication Checklist—Second Edition (CCC–2; D. Bishop, 2003), a standardized pragmatic language assessment tool.

Results—Both clinical groups demonstrated impairment in overall communication and pragmatic language functioning, but children with WS performed significantly better on overall pragmatic language functioning, and the magnitude of the effect was medium. Profile examination revealed equivalent performances between ASD and WS on most CCC–2 subscales; however, significantly better performances on the Coherence, Stereotyped Language, Nonverbal Communication, and Social Relations subscales were observed in WS.

Conclusions—The CCC–2 appears to provide an effective means to identify and characterize pragmatic language difficulties using a standardized approach in children with ASD and WS.

Keywords

pragmatics; assessment; autism spectrum disorders; Williams syndrome

Pragmatics is the linguistic domain concerned with the appropriate use of language across a variety of social contexts that provides for a listener's accurate interpretation of the speaker's intentions and references (Berko-Gleason, 2005). A broader definition incorporates behaviors that encompass social, emotional, and communicative aspects of

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social interaction (Adams, Baxendale, Lloyd, & Aldredge, 2005; Martin & McDonald, 2003). Pragmatics forms a critical intersection for children's developing language competencies and social interactions. Given the complexity of pragmatic language behaviors, assessment of pragmatics can be difficult, leaving many clinicians to rely on nonstandardized, observational methods that can be challenging for determining service eligibility (American Speech-Language-Hearing Association, 2006; Olswang, Coggins, & Timler, 2001; Young, Diehl, Morris, Hyman, & Bennetto, 2005). Nonetheless, available standardized measures can reliably identify pragmatic language impairment in children with various developmental histories (Bishop & Baird, 2001; Geurts et al., 2004; Gilmour, Place, & Skuse, 2004; Laws & Bishop, 2004; Norbury, Nash, Baird, & Bishop, 2004; Young et al., 2005).

Some developmental syndromes place children at risk for pragmatic language impairment. Autism spectrum disorders (ASD) and Williams syndrome (WS) represent two such developmental disorders (Gillberg & Rasmussen, 1994; Laws & Bishop, 2004; Tager-Flusberg, Paul, & Lord, 2005). ASD are a group of behaviorally diagnosed neurobiological, developmental disorders that occur in roughly 1 of every 150 individuals and involve relative strength in visuospatial skills and deficits in social and communicative functioning, as well as restricted/repetitive interests (American Psychiatric Association, 2000; Centers for Disease Control and Prevention, 2007). By contrast, WS is caused by a Chromosome 7 deletion, occurs in approximately 1 of every 7,500 live births, and results in a distinctive cognitive profile, including strengths in language relative to visuospatial skills, hypersociability, and a cluster of maladaptive behaviors (Mervis & Klein-Tasman, 2000; Semel & Rosner, 2003; Stromme, Bjomstad, & Ramstad, 2002). Although at the surface the disorders appear as phenotypic opposites (e.g., asocial in ASD and hypersocial in WS), pragmatic language difficulties have been described in both. This is intriguing in terms of the potential for similar etiologies between the two disorders, as well as better understanding the possible downstream effects of a splintered cognitive profile and/or altered social trajectories in early development. Standardized assessment of pragmatic language abilities is particularly important for these children with respect to the potential determination for service eligibility and the development and monitoring of appropriate educational modifications, treatments, and interventions.

Pragmatics-related social skills including peer relations, friendships, and variety and range of interests are differentially affected in children with ASD and WS. Social difficulties are a hallmark feature of ASD and accordingly have an impact on social interest in others, social relatedness, and higher level social behaviors, including the development and maintenance of age-appropriate friendships (as reviewed in Carter, Davis, Klin, & Volkmar, 2005). In WS, strength has been consistently noted in terms of a "heightened" interest and empathy for others, despite marked difficulties in the development and maintenance of reciprocal friendships (Jones et al., 2000; Semel & Rosner, 2003; Tager-Flusberg & Sullivan, 2000). Tager-Flusberg and Sullivan (2000) suggested that a dissociation in WS between the preserved ability to feel what another person feels (i.e., emotional perspective taking) and marked difficulties in thinking about why another person might feel or think a certain way (i.e., cognitive perspective taking) helps to explain this otherwise perplexing pattern. Finally, social deficits inherent in both disorders may be related to the repetitive and

restricted range of interests demonstrated by most individuals with ASD and WS across the life span (American Psychiatric Association, 2000; Laws & Bishop, 2004; Semel & Rosner, 2003).

In ASD, pragmatic language difficulties are well documented. Overly literal language comprehension, difficulties understanding gestures and body language, unusual emotional interpretations, and trouble understanding humor characterize receptive pragmatic deficits (Dewey & Everard, 1974; Happé & Frith, 1996; Loveland et al., 1997; Martin & McDonald, 2003; Ozonoff & Miller, 1996). With respect to expressive pragmatics, difficulties related to skill deficits have included limited use of facial expressions and gestures, lack of reciprocity in conversations (particularly with regard to topic establishment and maintenance), limited social initiations with others, impoverished prosody, a lack of cohesion in discourse, and difficulties with the use of pronouns, suggesting referential difficulties (Baltaxe, 1977; Dewey & Everard, 1974; Freeman & Dake, 1996; Happé & Frith, 1996; Lord, Rutter, & LeCouteur, 1994; Martin & McDonald, 2003; Szatmari, Bartolucci, & Bremmer, 1989). Children with ASD may also exhibit expressive pragmatic difficulties of unusual additions such as the use of uninhibited, socially inappropriate comments, an overuse of stereotyped utterances and tangential language, and increased use of idiosyncratic language and neologisms (i.e., novel made-up words for things; Dewey & Everard, 1974; Lord et al., 1994; Volden & Lord, 1991). Conversely, the ability to make conversational repairs may be a relatively preserved pragmatic language skill (Volden, 2004), and research on narrative skills (i.e., storytelling) in ASD supports a possible strength at the local level of storytelling with more difficulties at the global level (Loveland, McEvoy, & Tunali, 1990; Loveland & Tunali, 1993; Norbury & Bishop, 2003; Tager-Flusberg & Sullivan, 1995).

Previous studies have looked at pragmatic language profiles in ASD via standardized measures. Three studies of school-age children with ASD suggested significant difficulties within every pragmatic and social subdomain of the Children's Communication Checklist (CCC), which included Inappropriate Initiation, Coherence, Stereotyped Language, Use of Context, and Rapport (Bishop & Baird, 2001; Geurts et al., 2004; Verté et al., 2006).

Research on pragmatics in WS, conversely, remains patchy and incomplete, with much information based on observational and anecdotal reports (Gillberg & Rasmussen, 1994; Laws & Bishop, 2004; Semel & Rosner, 2003). Furthermore, the presence of hypersociability and heightened empathy in WS has led some to suggest that pragmatics may not be an area of weakness (Jones et al., 2000; Reilly, Klima, & Bellugi, 1990; Rice, Warren, & Betz, 2005). Some social behaviors in young children with WS, including increased interest in others, seeking of face-to-face interaction, and mimicry of facial expressions, indicate potential strength compared with typically developing and developmentally delayed controls (Fidler, Hepburn, Most, Philofsky, & Rogers, 2007; Laing et al., 2002; Mervis et al., 2003). Additionally, Schreiber (2002) observed an intact ability to read others' facial expressions in this population. Other researchers have noted pragmatics issues in this population with specific difficulties related to incessancy such as overtalkativeness; an overuse of learned, mature-sounding phrases; constant, inappropriate requests for attention and greeting behaviors; asking the same question over and over again; difficulties "giving up the floor"; making irrelevant and tangential comments causing

difficulties with topic maintenance; and perseveration or inappropriate and frequent mentioning of the same thoughts and ideas (Gillberg & Rasmussen, 1994; Laws & Bishop, 2004; Schreiber, 2002; Semel & Rosner, 2003; Stojanovik, 2006).

Although empirical analyses of pragmatic behaviors in school-age children with WS have been limited, Stojanovik (2006) analyzed the conversational structure of 5 school-age children with WS, in comparison to children with a history of typical development and children with specific language impairment (SLI). Results suggested that the conversations of children with WS were more immature and inappropriate than both comparison groups. Although children with SLI demonstrated a single area of pragmatic difficulty (i.e., topic drift) with significantly more difficulties in structural language, children with WS displayed predominantly pragmatic language difficulties, tending to provide too little information for the conversational partner, to overrely on the conversational partner's leads and contributions, and to demonstrate overly literal misinterpretations. Comparable performances between children with SLI and WS were evidenced in topic maintenance, language structure, and turn taking. As a function of comparison with SLI, Stojanovik (2006) concluded that the pragmatic difficulties in WS could not be attributed to structural language difficulties but rather to the impaired general cognitive abilities characteristic of the syndrome.

Laws and Bishop (2004) administered the CCC to the parents of 19 individuals with WS between the ages of 6 and 22 and determined significant impairment in pragmatic language functioning relative to typical, Down syndrome, and SLI controls. They cited inappropriate initiation of conversation and use of stereotyped language as two areas of pragmatic competence that were "particularly affected" (p. 59) in children with WS. They concluded that pragmatic impairment in WS is significant enough to warrant interventions in pragmatics and social skills like those offered to children with autism (Laws & Bishop, 2004).

A number of similarities and differences become apparent in comparing the pragmatic language characteristics of individuals with ASD and WS. See Table 1 for a summary of these findings. The similarities and differences reported in Table 1 have not been proved empirically but are rather conclusions drawn from literature reviews of pragmatics in both syndromes, which derive from a combination of both empirical findings and clinical observations. While clear syndrome differences are notable for six pragmatic behaviors (i.e., use of eye contact, reading and interpreting emotion in facial expressions, prosody, social initiations, and social interest), three other social-pragmatic behaviors result in ambiguous interpretation in terms of potential between-syndrome differences (i.e., gesture use, overtalkativeness, and topic maintenance). Finally, another nine pragmatic behaviors present similarly in both syndromes, such that both groups of children either demonstrate or do not demonstrate the particular behavior.

Rice and colleagues (2005) argued that there is a current research need for pairwise comparisons between different developmental conditions to better understand similarities and differences observed between linguistic profiles. While pragmatic language impairments have been documented separately in ASD and WS, the two syndromes have not been

directly compared previously on this dimension using a standardized tool. Such lack of comparison is likely related to difficulties with accessible assessment tools, difficulties matching two syndromes characteristically different with respect to cognitive-linguistic profile, and current limitations in use of this methodology in language studies (Bishop, 1998; Rice et al., 2005; Young et al., 2005). Pragmatic language impairment on a standardized measure of pragmatic assessment has not been replicated in children with WS, and the sample of children used in the Laws and Bishop (2004) study composed a variable age range from children to young adults, spanning several different age cohorts including school-age, adolescence, and young adulthood.

Thus, the purpose of this study was to compare and describe the pragmatic language profiles of school-age children with ASD and WS on a standardized measure for pragmatic language to determine whether pragmatic profile characteristics can differentiate between the two groups of children with opposing social presentations and pragmatic language difficulties. Such research is important for better understanding how differential social trajectories can potentially affect pragmatic language and social development in the school-age years.

We tested the following two hypotheses:

1. Children with ASD and children with WS will demonstrate clinically significant impairment in overall pragmatic language functioning.
2. Profile analysis will indicate a syndrome-specific profile of pragmatic language skills for each group. Specifically, children with WS will show relative strength on the CCC—Second Edition (CCC–2; Bishop, 2003) subscales measuring communicative use of eye contact, reading and understanding of facial expressions, and interest in others (i.e., Nonverbal Communication and Social Relations). Conversely, children with ASD will significantly outperform children with WS on the CCC–2 subscale measuring incessant, inappropriate initiations with others (i.e., Inappropriate Initiation). Finally, while prosody (a strength for children with WS) is measured in the Stereotyped Language subscale of the CCC–2, so is use of overlearned phrases (a weakness for both children with WS and ASD), rendering it a subscale where there will be a smaller significant difference in favor of children with WS over children with ASD.

Method

Participants

Participants were recruited from two larger studies concerning the developing behavioral phenotypes of ASD and WS (Collaborative Programs for Excellence in Autism [CPEA], principal investigators S. Rogers & S. Hepburn; and Fidler et al., 2007, respectively). Recruitment included advertisements reaching educators, clinicians, and parents of children with disabilities in the community; participation in community workshops; and parent advocacy and support groups, including the Williams Syndrome Association and Autism Society of America.

See Table 2 for study participant characteristics. Sixty-two participants were included in three groups: WS (n = 21), ASD (n = 22), and typically developing (TYP; n = 19). The TYP group was added to assist in establishing the CCC–2's validity in a group of school-age American children. Few children with typical development should show pragmatic language difficulties on the measure. Inclusion criteria for this study were as follows:

1. (a) A diagnosis of WS via genetic testing as endorsed by a parent to be qualified into the WS group, (b) a diagnosis on the autism spectrum (i.e., autistic disorder, pervasive developmental disorder-not otherwise specified [PDD-NOS], or Asperger's syndrome) via clinical opinion and endorsement of clinically significant signs on the Autism Diagnostic Observation Schedule (ADOS; Lord, Rutter, DiLavore, & Risi, 2002) and either the Autism Diagnostic Interview—Revised (ADI–R; Lord et al., 1994) or Social Communication Questionnaire (SCQ; Berument, Rutter, Lord, Pickles, & Bailey, 1999) to be qualified into the ASD group, or (c) absence of cognitive delays and no known current developmental diagnoses to be qualified into the TYP group.
2. Chronological age between 5;11 (years;months) and 12;11.
3. Verbal fluency (i.e., the ability to speak in full sentences) evidenced by an age-equivalent score of 3;11 or older on the Expressive subdomain of the Vineland Adaptive Behavior Scales (VABS; Sparrow, Balla, & Cicchetti, 1984) First or Second Edition (VABS–II; Sparrow, Cicchetti, & Balla, 2005).
4. Parents who were willing to complete and return a checklist concerning their child's pragmatic language abilities, as well as complete a 20–30-min standardized VABS–II interview in person (if local) or over the phone.

Specific participant diagnoses from the *Diagnostic and Statistical Manual of Mental Disorders* (American Psychiatric Association, 2000) were as follows: autistic disorder (n = 13), PDD-NOS (n = 7), and Asperger's syndrome (n = 2). Both the WS and TYP groups demonstrated relatively even gender distributions reflective of their respective larger populations, but the ASD group was overrepresented by males, which is relatively consistent with the larger population of children with ASD (Fombonne, 2005). All children in the TYP group scored above 85 (M = 100 ± 15) in the verbal and nonverbal IQ domains of a standardized measure of cognitive functioning (i.e., the Wechsler Abbreviated Intelligence Scales [WASI] or Wechsler Intelligence Scales for Children—Fourth Edition [WISC–IV]).

Procedures

Families participated in 3–5 sessions either at the laboratory at the University of Colorado at Denver Health Sciences Center or at the family's home. Parents were administered the VABS (original or second edition) by a second clinician in an adjacent room, while the child was participating in autism diagnostic, cognitive, or other measures not reported here. CCC–2 protocols were mailed prior to a child's first lab session. Additional participants were recruited for the WS group, most of whom were not local. Interested parents contacted the study by phone and were screened for eligibility. If a parent agreed to participate in the study, study consents were discussed and a packet was mailed with all forms relevant to the

study. Additionally, an appointment to complete the VABS-II occurred either over the phone or in person.

Measures

Inclusion and outcome measures were used in this study. Inclusion measures comprised two formal autism diagnostic measures (i.e., ADOS and ADI–R or SCQ) for the ASD group only, a form (i.e., Medical Records Checklist) to elicit parent report of a WS diagnosis for the WS group only, and two variables from the VABS (original or second edition) for all three groups: the Adaptive Behavior Composite and the expressive language age equivalent (ELAE). All administrators of the autism diagnostic measures were trained to 80% reliability by a certified trainer and established administration fidelity and interrater reliability from the beginning of the study, which was maintained for at least 20% of assessments. The WASI or WISC–IV was also administered to the TYP participants to ensure typical cognitive functioning. The outcome measure was the CCC–2. All study measures were administered in a standard manner by trained examiners.

Inclusion Measures

ADOS: The ADOS (Lord et al., 2002) is a standardized, semistructured interview that employs developmentally appropriate social and toy-based probes in a 45–60-min interaction between a clinician and the child to elicit possible signs of autism in four areas: social interaction, communication, play, and repetitive behaviors. The Module 3 ADOS was used for this study because that is the module available for “verbally fluent” children, a criterion necessary for study participation. On the Module 3 ADOS, 28 behavioral dimensions (e.g., Conversation, Stereotyped Behaviors, Social Responsiveness) are scored on a 3-point scale, with higher scores indicating more evidence for abnormality of a particular behavior. A total score from the algorithm of the ADOS was used for qualification purposes, with score totals above the “autism spectrum cutoff” necessary for qualifying participants into the ASD group. The reliability and validity of the ADOS are generally strong (Lord et al., 2002).

ADI–R: The ADI–R (Lord et al., 1994) is a structured, standardized parent interview designed to consider the presence and severity of autism behaviors in children across all three main behavioral areas involved in autism: social relatedness, communication, and repetitive behaviors. An algorithm differentiates autism from other developmental disorders. A total score from the algorithm of the ADI–R was used for qualification purposes, with scores above the “autism spectrum cutoff” necessary for qualifying participants into the ASD group. Reliability and validity data for this instrument are generally good to excellent (Lord et al., 1994). Many participants had completed a full ADI–R through their participation in the third author’s longitudinal study of behavior (CPEA). Due to time and cost constraints, some participants received a shortened version of this interview, the SCQ.

SCQ: The SCQ (Berument et al., 1999), also sometimes called the Autism Screener Questionnaire, is a 40-item parent interview for autism behaviors developed to be a parent report measure based on the ADI–R. The tool may be used with individuals of any age or IQ level. It has good discriminative validity (.88) for the separation of autism spectrum

diagnoses (autism/Asperger's/PDD-NOS) from other diagnoses and specificity (.75), although the tool may have some difficulty distinguishing between autism diagnoses (i.e., autism vs. PDD-NOS) and is considered conservative for not picking up some high-functioning individuals on the spectrum (Berument et al., 1999).

Medical Records Checklist: The Medical Records Checklist (Fidler & John, 2002) is a one-page form that takes about 5 min for a parent to complete. The form asks if and when the parent's child received a formal diagnosis of WS and includes a yes/no checklist of common, concomitant medical conditions (e.g., heart anomalies, dental abnormalities) that are sometimes associated with WS.

VABS: The VABS (Sparrow et al., 1984) is a standardized 291-item parent interview that determines levels of adaptive behavior in three areas (i.e., communication, daily living, and socialization) for children older than 6 years. A variable within the communication domain, ELAE, was used to qualify every participant into the study to be sure that the child was functioning at an expressive language level suitable for use of the CCC–2 (i.e., speaking in full sentences). Additionally, the Adaptive Behavior Composite (an overall adaptive functioning composite variable) was used as a proxy to describe overall developmental functioning in the clinical groups. The VABS has good to excellent psychometric properties, has been widely used in research contexts, and shows good convergent validity with the VABS-II (Sparrow et al., 2005).

VABS–II: The VABS–II (Sparrow et al., 2005) is a revised and updated version of the VABS, with an expanded age range and updated items to reflect cultural changes and increased research knowledge. The VABS–II comprises the same domains and subdomains as the VABS, and so the same variables were used. The VABS–II shows a high degree of consistency with the VABS and demonstrates good psychometric properties (Sparrow et al., 2005).

WISC–IV: The WISC–IV (Wechsler, 2003) is a standardized test of intelligence for children age 6–16 years. It examines verbal and nonverbal intelligence performance and provides age equivalents and standard scores for each. An abbreviated version of the test can be given using four subscales, and this yields two scores relevant to this study: (a) verbal composite index, also referred to as verbal IQ in this study, and (b) perceptual composite index, also referred to as nonverbal IQ. Children in the TYP group needed to score in the nonimpaired range for both (i.e., above 85). The WISC–IV generally demonstrates good psychometric properties and shows acceptable convergent reliability with the WASI (Wechsler, 2003).

WASI: The WASI (Wechsler, 2002) is a standardized test of intelligence for children age 6–16 years. For the purposes of this study, the verbal IQ and nonverbal IQ were used to qualify participants into the TYP group with scores in the nonimpaired range for both (i.e., above 85). The WASI shows acceptable convergent reliability with the WISC–IV (Wechsler, 2002).

Outcome Measure

CCC–2: The CCC–2 (Bishop, 2003) is a 70-item checklist concerning social and communication behaviors that takes approximately 10–20 min to complete by a rater (typically a parent, teacher, or therapist) who has been familiar with a child for at least 3 months. Raters make a frequency judgment for each item (i.e., 0 = less than once a week or never; 1 = at least once a week, but not every day; 2 = once or twice a day; and 3 = several times [more than twice] a day or always). This measure is appropriate for children who are between the ages of 4 and 16 years and who speak in full sentences.

See Table 3 for a general description and domain of the 10 CCC–2 subscales. Each CCC–2 subscale consists of seven items—five items tap into communication deficits, and two items concern strengths. Four subscales comprise structural language, four pragmatic language, and two are additional autism/social subscales that the tool's author included with the intention of signaling children who may require additional evaluation for ASD (Bishop, 2003). Scaled scores may be derived from each subscale's total raw score. Scaled scores of 4 or lower on a subscale indicate impairment within that area, while a scaled score of 6 or higher indicates typical functioning. Two summary variables of interest emerge from a completed CCC–2: the General Communication Composite (GCC) and the Social Interaction Deviance Composite (SIDC). The GCC was designed to identify children with a communication impairment, such that any number below a cutoff score (i.e., 55) is indicative of some type of communication impairment (i.e., structural or pragmatic or both). Additionally, the SIDC variable (i.e., scaled scores for subscales [A + B + C + D]–[E + F + I + J]) may be used only *in conjunction* with children indicated by a low GCC score (i.e., under 55) and is intended to signal pragmatic language difficulties from structural language difficulties in children with a communication impairment (according to their GCC score). An additional summary variable, PRAG (i.e., the sum of scaled scores for pragmatic language subscales: E + F + G + H), was included as an index of overall pragmatic language functioning. Clinical experience in working with this tool, along with the suggestion that such a variable may be "very effective in discriminating children with clinical diagnoses from controls" (Bishop, 2003, p. 27), led the authors to include PRAG for the purposes of this study. Bishop (1998) had included a similar variable (i.e., Pragmatic Composite) in the earlier CCC version of the tool. The Pragmatic Composite summed most social-pragmatic subscales (not Interests) of the CCC but dropped its use in the CCC–2 as a result of "disappointing levels of validity and inter-rater reliability" (Bishop, 2003, p. 27), when used for the purpose of distinguishing subtypes of children with a communication disorder (as opposed to separating out clinical from control children). The PRAG variable is also slightly different than the former Pragmatic Composite (Bishop, 1998), because the Inappropriate Initiation subscale was included (not its own subscale in the CCC), and neither the Coherence nor Social Relations subscale was included.

Reliability and validity data for the CCC–2 are available in the manual (Bishop, 2003). The tool's standardization procedures are based on three small to medium-sized samples of British English-speaking children. Additional standardization for Australian children in the manual suggested slightly different norms for Australian versus British children. According to the British standardization sample, internal consistency of subscales ranged from .65 to .

80. Interrater reliabilities for each scale were computed between parents and either teachers or speech-language pathologists, with correlations ranging from .16 to .52 (i.e., the Stereotyped Language subscale was the lowest, and the Speech subscale was the highest); however, use of the SIDC significantly boosted the interrater reliability between parents and professionals to .79 (Bishop, 2003; Norbury et al., 2004). Validation data for the CCC–2 suggested good criterion validity in terms of identification of impaired versus typical pragmatic functioning. These data also suggested that the CCC–2 may not be as good at distinguishing between subtypes of communication impairment (Bishop, 2003).

Statistical Analysis

Data were analyzed using SPSS Version 15.0 for Windows. Descriptive statistics (means and standard deviations) were calculated for all participant characteristic and outcome variables. All between-group comparisons for participant characteristics, CCC–2 outcome variables, and CCC–2 subscale raw scores used either multivariate analysis of variance or multivariate analysis of covariance (MANCOVA). Raw scores were selected (as opposed to standard scores) for CCC–2 scale scores because the raw scores offered a fuller range of values than the standard scores, increasing the like-lihood of detecting more subtle differences. Because ELAE on the VABS (original or second edition) might influence CCC–2 performance, all CCC–2 comparisons were conducted by covarying for effects of ELAE. When differences were detected, post hoc analyses with Bonferroni correction for inflation of alpha were run. When the homogeneity of variance assumption was violated, the Games-Howell procedure was used. Cronbach's alpha coefficients were used for determining the internal validity of the CCC–2. Because the variability of responses was limited for many of the questions within each individual group, use of the entire sample was warranted for this calculation. Acceptable levels of internal consistency were sought for every CCC–2 subscale (i.e., above .7; Cicchetti, 1994). An alpha level of .05 was chosen for reporting significance for all analyses.

Results

Preliminary Analysis

Group characteristic comparisons—There was no significant difference between chronological age for the clinical groups, but the typical participants were significantly chronologically younger (M = 6;11, SD = 1;0) than either clinical group, M(ASD) = 9;6, SD = 1;7, and M(WS) = 9;1, SD = 2;2, $F(2,59) = 15.0$, $p < .001$, $\eta^2 = .99$.[1] The ELAE of the typical group was compared with the chronological age of both clinical groups, revealing no significant between-group differences, $F(2, 59) = 0.21$, $p = .81$, $\eta^2 = .01$, suggesting that the typical group of children were verbally expressing themselves (M = 9;4, SD = 3;4) at the same approximate chronological age level of both clinical groups, M(ASD) = 9;6, SD = 1;7, and M(WS) = 9;1, SD = 2;2. In comparing the chronological age of the typical group with the ELAE of both clinical groups, significant differences were detected as follows: TYP = ASD > WS, $F(2, 59) = 9.3$, $p < .0001$, $\eta^2 = .24$, suggesting that the participants with ASD

[1]There was a significant Levene's test ($F = 11.6$, $p < .001$), suggesting that the homogeneity assumption was violated for chronological age only.

were speaking at approximately the same chronological age level as the typical children, *M*(ASD) = 6;8, *SD* = 1;3, and *M*(TYP) = 6;11, *SD* = 1;0. The participants with WS were speaking at a significantly lower expressive language age level (*M* = 5;7, *SD* = 1;0), supporting a need to control for the effects of ELAE when comparing CCC–2 outcome variables.

Internal reliability of the CCC–2 within the sample: Cronbach's alpha—The alphas for the entire sample combined for all 10 CCC–2 subscales ranged between .71 and .87 and were relatively consistent with the internal consistency values reported in the CCC–2 manual for British English-speaking children. These data suggest that the different CCC–2 subscales are internally consistent in this sample of American English-speaking children. The similar patterns of internal consistency in this sample and the CCC–2 standardization sample suggest that it is appropriate to use the scaled scores derived from the CCC–2 manual to compute summary variables for this study.

Study Analyses

Analysis of the CCC–2 summary variables—See Table 4 for mean CCC–2 summary scores (i.e., GCC, SIDC, and PRAG) by diagnostic group, as well as adjusted means, covarying for ELAE. Recall that the GCC variable is the sum of all 10 scaled scores on the CCC–2 and indexes overall communication functioning, the SIDC variable is a ratio score to signal either a structural or pragmatic language impairment in children with a communication impairment (low GCC score), and PRAG is the sum of the scaled scores for the four purely pragmatic subscales of the CCC–2. One ASD participant was removed for being an extreme outlier on both the GCC and PRAG CCC–2 summary variables such that this particular participant's GCC and PRAG scores were more than 2 *SDs* above the next lowest ASD participant.[2] Also, the TYP group was not included in inferential analyses because a major purpose of the study was to compare ASD and WS on pragmatics, allowing for an accurate interpretation of effect size for each outcome variable between clinical groups (effect sizes would be elevated by including the TYP participants). Visual inspection of CCC–2 summary scores (i.e., GCC and PRAG) in the TYP group suggested both typical functioning (according to the CCC–2 manual) and much stronger performances in GCC and PRAG variables compared to both clinical groups. Recall that the SIDC variable should not be interpreted for individuals receiving GCC scores above 55 (all TYP participants) and were accordingly not reported.

Participants with ASD and WS were not significantly different on either the GCC or SIDC CCC–2 summary variables, covarying for ELAE: GCC, $F(1, 39) = 2.6$, $p = .12$, $\eta^2 = .06$, and SIDC, $F(1, 39) = 1.2$, $p = .29$, $\eta^2 = .03$, indicating indistinguishable performances in overall communication functioning and overall pragmatic language impairment. However, there was a significant between-group difference for PRAG, covarying for ELAE, $F(1, 39) = 11.3$, $p = .002$, $\eta^2 = .22$, such that WS significantly outperformed ASD in overall pragmatic language functioning.

[2]GCC and PRAG scores for the outlier were 79 and 34, respectively, with the next lowest values in the ASD group being 46 and 20, respectively.

Comparison of ASD and WS on raw subscale scores by CCC–2 profile—See Table 5 for mean raw scores, standard deviations, and range by diagnostic group along with MANCOVA results comparing ASD and WS, controlling for ELAE. The adjusted means slightly elevated the WS group means and decreased the ASD group means, in line with the slight advantage given to the WS group to account for between-group differences in ELAE. Significant differences were noted, such that WS outperformed ASD on four CCC–2 subscales: Coherence, Stereotyped Language, Nonverbal Communication, and Social Relations. Conversely, nonsignificant differences were noted in six CCC–2 subscales: Speech, Syntax, Semantics, Inappropriate Initiation, Use of Context, and Interests. Also, although the difference was nonsignificant, the Inappropriate Initiation subscale was the only social-pragmatic subscale where the mean score of the children with WS was higher than the mean score for children with ASD (higher scores indicate more impairment).

Discussion

This study compared school-age children with ASD and WS on a standardized, parent-rated assessment for pragmatic language functioning, the CCC–2 (Bishop, 2003). A typically functioning school-age control group was included to assess the instrument's validity for use with American English-speaking children. Consistent with the study's hypotheses, the CCC–2 detected similar levels of pragmatic impairment in both clinical groups, although the children with WS performed significantly better in overall pragmatic language functioning than the children with ASD. It should be noted that the magnitude of this difference was medium (according to Morgan, Griego, & Gloekner, 2001). No significant differences were found on the Inappropriate Initiation, Use of Context, and Interests autism/social-pragmatic subscales, and the children with WS demonstrated better performance than the children with ASD on the Coherence, Stereotyped Language, Nonverbal Communication, and Social Relations subscales.

Contrary to the hypotheses, children with ASD did not outperform children with WS on the Inappropriate Initiation subscale. This null finding may be due to low observed power (.17) or small sample size and/or low effect size (.03), because the mean scores of the children with WS were slightly higher in the predicted direction. It is alternately possible that this particular subscale of the CCC–2 is equally sensitive at detecting the unusual initiations characteristic of both groups, thus resulting in similar performances. Also, not predicted in the hypotheses, children with WS also out-performed children with ASD on the Coherence subscale, a structural language subscale of the CCC–2 (formerly a pragmatic language subscale on the CCC; Bishop, 1998), which considers a child's ability to take the listener's perspective into account and provide the right amount of information to understand references to other people and sequence of events. While school-age children with ASD and WS have not been directly compared on these behaviors to date, children with ASD have demonstrated significant difficulties with this subscale on the CCC compared with TYP, attention deficit/hyperactivity disorder, and learning-disabled controls, and children with WS have demonstrated significant difficulties compared with TYP controls (but not Down syndrome or SLI controls) on this subscale of the CCC–2 (Bishop & Baird, 2001; Geurts et al., 2004; Laws & Bishop, 2004; Verté et al., 2006).

This study also served to further validate the use of the CCC–2—available for clinical and research use in the United States and standardized on British English-speaking children—with American English-speaking school-age children. Support for the tool's construct validity was generated by the typical overall communication and pragmatic language functioning of the TYP group, along with impaired overall communication and pragmatic language functioning for both clinical groups. The tool also demonstrated acceptable levels of internal consistency, similar to the levels reported for the tool's standardization sample (Bishop, 2003).

While children with ASD and WS have been previously likened to one another in terms of pragmatics (Gillberg & Rasmussen, 1994; Laws & Bishop, 2004), this is the first empirical comparison of these two groups on this dimension in school-age children. The results of this study suggested that children with WS may perform better in some areas of pragmatics (including use of stereotyped language) and similarly in other pragmatic language abilities compared to children with ASD. Laws and Bishop (2004) cited inappropriate initiation of conversation and the use of stereotyped language as areas of pragmatic competence that are "particularly affected" (p. 59) in children with WS, but comparisons to children with ASD paints a slightly richer clinical picture. It is also important to take into account that according to the CCC–2 behavior groupings, using appropriate prosody contributes to the superior performance of children with WS compared to children with ASD within Stereotyped Language behaviors. Additionally, while children with WS did demonstrate difficulties on the Inappropriate Initiation subscale of the CCC–2, their performance was not significantly different than the children with ASD, though power levels to detect differences were limited in this study.

Consistent with their behavioral phenotypes, children with ASD demonstrated impairment across all domains of social-pragmatic functioning, while children with WS demonstrated a few areas of relative strength compared to ASD. Essentially, parents gave their children with WS significantly higher ratings than children with ASD on items regarding the use and understanding of affective expressions; prosody; overuse of rote, learned phrases; social responsiveness; and empathy and social relatedness with others, reporting differences of large clinical magnitude between the two groups of children. This finding is consistent with previous research on toddler and preschool-age children with WS who demonstrate relative strength in emotional responsivity, including interest in others, dyadic face-to-face interaction, and increased affect imitation, compared to children with other developmental delays and typical development (Fidler et al., 2007; Laing et al., 2002; Mervis et al., 2003). Further, the significant differences noted in favor of children with WS (over ASD) on questions relating to the ability to appropriately sequence and reference events for a listener were of a smaller magnitude (medium effect size).

Children with ASD and WS both have difficulties with friendships (Carter et al., 2005; Tager-Flusberg & Sullivan, 2000). Both clinical groups demonstrated impairment in their quality of initiations with others, use and understanding of abstract language concepts like humor, and in overall quality and variety of interests. Difficulties in these areas could account for the reason that children with WS have difficulty with friendships. Despite a clear interest in friendships (including relative strengths of large magnitude in relation to ASD in

Social Relations), children with WS lack some of the pragmatic tools necessary for developing and nurturing them.

Clinical Implications

This study has implications for speech-language pathologists, educators, and interventionists working with children with these disorders. Given the significant and sharp rise in prevalence of ASD over the past decade (Centers for Disease Control and Prevention, 2007), children with the disorder are likely to be increasing members on therapist caseloads. Therefore, information about relevant clinical tools is critical to the speech-language pathologists working with these children. Additionally, a closer look at the pragmatic functioning of these two syndrome groups allows for a richer understanding of what constitutes atypical pragmatic language functioning in school-age children. Because pragmatic language behaviors have historically been clinically elusive, with norms lacking even for typical development in the school-age years, delineating the parameters of atypicality further assists in our understanding of pragmatic language development in school-age children (Norris, 1995).

With respect to assessment, this study provided data to support the use of a relatively new tool for understanding the pragmatic language functioning of a child. The CCC–2 identified every child with WS and ASD as demonstrating communication impairment (per the GCC variable) and many children with ASD and WS as demonstrating a pragmatic language impairment (per the SIDC variable). There were still some children with both WS and ASD who were not identified as demonstrating pragmatic language impairment, according to the SIDC variable, but were still demonstrating pragmatic language difficulties, according to the PRAG variable. Additional use of the PRAG variable (i.e., the sum of the scaled scores of the four pragmatic language scales) is accordingly recommended for use in clinical populations as a more sensitive index of potential pragmatic language difficulties. Although the PRAG variable was not developed to function as a summary variable in the CCC–2 because it was not good at discriminating between children with structural and pragmatic language impairments (children with SLI obtained unexpectedly poor scores on the variable; Bishop, 2003), this variable could potentially provide additional useful information to clinicians about an individual child's pragmatic language functioning, particularly for those children with communication impairments (GCC scores below 55) who do not evidence SIDC scores within the range of pragmatic language impairment. Use of a standardized measure like the CCC–2, along with other methods for looking at pragmatic language (i.e., direct assessment, observation across a variety of contexts, etc.), is currently considered best practice for assessing pragmatic language functioning in these populations (American Speech-Language-Hearing Association, 2006; Olswang et al., 2001).

In terms of intervention, a better understanding of the pragmatic language characteristics of individuals with ASD and WS may be useful to educators and interventionists for implementing successful treatment approaches (Hodapp & Fidler, 1999). Because most pragmatics interventions have been designed for children with ASD, increased understanding of the relative strengths and weaknesses of children with WS compared to ASD could allow for appropriate and sensitive alterations in pragmatics intervention

techniques designed for children with ASD. Whereas targeted social skills/pragmatics training in autism tends to be cognitive and rule-based, techniques using feelings and how different behaviors affect others may be more effective for teaching children with WS about appropriate social behavior, given their relative strength in Nonverbal Communication and Social Relations. Thus, knowledge of the behavioral tendencies of a child with WS can inform the therapist as to possible techniques for altering an existing intervention protocol. Finally, valid tools for measuring pragmatics may aid therapists in developing and monitoring treatment plans for these populations (Young et al., 2005). For example, individual examination of a child's CCC–2 pragmatic profile might alert a therapist to address inappropriate initiating and abstract language understanding/use but not nonverbal gesture use.

Limitations and Future Work

Some limitations and directions for future work were noted. WS is a rare genetic disorder, limiting the sample size for this study and potentially contributing to some of the null effects. The age variability of the children introduced another potential confound. Not matching the groups on a third control variable posed another possible limitation; however, the children were not significantly different in chronological age, and all were talking in full sentences. This has been the precedent for research comparing different diagnostic groups on pragmatics using the CCC and CCC–2 (Bishop & Baird, 2001; Botting, 2004; Botting & Conti-Ramsden, 1999; Geurts et al., 2004; Gilmour et al., 2004; Laws & Bishop, 2004; Norbury et al., 2004; Verté et al., 2006). Additional implications for future work include assessment of interrater reliability on the CCC–2 between American professionals and parents, establishment of norms in typical school-age populations (Norris, 1995), consideration of the variability of pragmatic language functioning in children with both disorders and the efficacy of targeted treatments on social outcomes in these populations (Adams et al., 2005), and examination of the relations between the CCC–2 and other measures of pragmatic language functioning, such as the Test of Pragmatic Language (Phelps-Teraski & Phelps-Gunn, 1992).

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TABLE 1

Comparative summary of pragmatic language behaviors in autism spectrum disorders (ASD) and Williams syndrome (WS).

Pragmatic behavior	ASD	WS
Unusual use of eye contact	+	−
Difficulty reading emotion in facial expressions	+	−
Difficulties expressing emotion in facial expressions	+	−
Difficulty understanding communicative, nonverbal gestures	+	+
A lack of use of communicative, nonverbal gestures	+	−/+
Difficulties with conversational repair	−	−
Lacking prosody (i.e., voice melody)	+	−
Difficulties with topic maintenance	+	−/+
Overtalkativeness	−/+	+
Lacking social initiations with others	+	−
Difficulties with conversational reciprocity	+	+
Perseveration with language	+	+
Difficulties with topic coherence	+	+
Use of tangential language	+	+
Difficulties interpreting abstract language	+	+
Difficulties demonstrating interest in others	+	−
Difficulties with friendships	+	+
Restricted and repetitive interests	+	+

Note. References for these pragmatic behaviors in ASD and WS are available in the text. A plus symbol indicates the behavior was observed in diagnostic group; a minus symbol indicates the behavior was not observed in diagnostic group.

TABLE 2

Participant characteristics.

Variable	ASD (*n* = 22)	WS(*n* = 21)[a]	TYP(*n*=19)
Diagnostic status	100% endorsement of an autism spectrum diagnosis on both ADOS and ADI–R or SCQ and by expert clinical opinion	100% parent endorsement on Medical Records Checklist	—
Gender (% male)	M 19/F 3 (86)	M 11/F 10 (52)	M 8/F 11 (42)
Chronological age in years;months[b]			
M	9;6	9;1	6;11
SD	1;7	2;2	1;0
Range	7;1–12;10	6;2–12;5	5;10–10;0
ELAE on the VABS/II chronological age in years;months[c]			
M	6;8	5;7	9;4
SD	1;3	1;0	3;4
Range	4;5–8;9	3;11–7;5	5;6–11;3
ABC of the VABS/II[d]			
M	60	69	108
SD	15.3	10.3	10.0
Range	33–93	40–90	86–124

Note. Dash indicates not applicable. TYP = typical; ADOS = Autism Diagnostic Observation Schedule; ADI–R = Autism Diagnostic Interview—Revised; SCQ = Social Communication Questionnaire; ELAE = expressive language age equivalent; VABS/II = Vineland Adaptive Behavior Scales, First or Second Edition; ABC = Adaptive Behavior Composite.

[a] Because VABS/II data were not available for 1 participant with WS, the average values for the WS group for those variables were substituted for missing values for that participant.

[b] Indicates TYP < ASD = WS.

[c] Indicates significant between-group differences: TYP > ASD > WS; accordingly, CCC–2 comparisons will covary for ELAE.

[d] Indicates significant between-group differences: TYP > ASD = WS.

TABLE 3

Children's Communication Checklist—Second Edition (CCC–2) subscales.

Subscale	Theme of behaviors making up subscale items	Domain
A: Speech	Articulation and intelligibility	Structural
B: Syntax	Word order, grammatical markings	Structural
C: Semantics	Fluency with word recall, word-finding, and vocabulary access	Structural
D: Coherence[a]	Making sense in conversation through the proper referencing and sequencing of people and events	Structural
E: Inappropriate Initiation	Indiscriminate, talks too much, does not initiate topics about reciprocal interests, repetitive initiating	Pragmatics
F: Stereotyped Language	Overuse of "learned chunks" in conversations, prosody, being overly "precise" in communications	Pragmatics
G: Use of Context	Use and understanding of the social rules governing communication, including politeness, sarcasm, and humor; ability to correctly interpret others, including abstract language concepts	Pragmatics
H: Nonverbal Communication	Understanding and using nonverbal conversational cues including both gestures and facial expressions/affect	Pragmatics
I: Social Relations	Interest in and relations with peers	Autism/social
J: Interests	Restricted and/or repetitive interests, flexibility	Autism/social

[a]While the Coherence subscale was considered pragmatics in the CCC and contains questions about behaviors that many would consider pragmatic language behaviors (including discourse), validity analyses for the CCC–2 shifted this particular subscale into structural language as a result of its inability to differentiate children with SLI from children with more extreme pragmatic language difficulties and because some findings have argued that narrative ability is more closely related to core language abilities than to pragmatic language abilities (Norbury et al., 2004).

TABLE 4

Mean CCC–2 summary scores by diagnostic group with adjusted means (covarying for ELAE on the VABS/II) presented for the clinical groups.

Study group	WS (*n* = 21)	ASD (*n* = 21)[a]	TYP (*n* = 19)
GCC			
M	32.3	26.8	87.3
SD	12.1	9.5	13.4
Range	8–28	10–46	57–116
Adjusted *M*	32.6	26.5	NA
SIDC			
M	−0.6	−2.7	NA for children with GCC > 54
SD	8.0	7.1	
Range	−17–15	−22–10	
Adjusted *M*	−0.21	−3.03	
PRAG			
M	15.8	11.7	43.6
SD	5.0	4.5	7.9
Range	8–28	3–20	32–60
Adjusted *M*	16.4	11.1	NA

Note. GCC = General Communication Composite, an index of overall communication skills; GCC scores above 54 are considered within normal limits, while scores below 55 are considered indicative of some type of language impairment SIDC = Social Interaction Deviance Composite, an indicator of relative difficulties with pragmatics or structural language; positive SIDC values suggest more structural language impairment, and negative SIDC values indicate more pragmatic language impairment in children with a language impairment (GCC scores below 55). PRAG = pragmatic language composite, the sum of the scaled score values for 4 pragmatic language subscales; PRAG scores above 24 are indicative of children with typical pragmatic language functioning, scores between 17 and 24 indicate borderline typical pragmatic language functioning, and scores below 17 indicate impaired pragmatic language functioning.

[a]One ASD participant's CCC–2 data were removed as a function of being an extreme outlier.

TABLE 5

Mean raw scores (higher scores indicate greater difficulty) and mean adjusted raw scores (controlling for ELAE) for the clinical groups obtained on the CCC-2 subscales by diagnostic group and multivariate analysis of variance results for the ASD and WS groups, covarying for ELAE.

CCC–2 subscale	ASD (*n* = 20)[a]	WS (*n* = 21)	TYP (*n* = 19)	ASD vs. WS
A: Speech				
Raw *M*	4.2	3.0	0.6	
SD	3.1	3.0	1.1	
Range	0–11	0–7	0–4	
Adjusted *M*	4.5	2.7		
F value				3.0
Significance				.09
Effect size (η^2)				.07
Observed power				.39
B: Syntax				
Raw *M*	5.9	5.1	.6	
SD	3.0	3.5	1.0	
Range	1–14	1–14	0–3	
Adjusted *M*	6.0	5.0		
F value				0.8
Significance				.37
Effect size (η^2)				.02
Observed power				.15
C: Semantics				
Raw *M*	9.1	9.4	2.4	
SD	3.1	4.2	2.6	
Range	3–14	3–19	0–10	
Adjusted *M*	9.5	9.1		
F value				0.13
Significance				.72
Effect size (η^2)				.00
Observed power				.06
D: Coherence				
Raw *M*	11.0	8.3	1.2	
SD	3.7	3.1	1.7	
Range	5–18	4–14	0–5	
Adjusted *M*	11.2	8.1		
F value				7.1
Significance				.01
Effect size (η^2)				.15
Observed power				.74
E: Inappropriate Initiation				

CCC–2 subscale	ASD (*n* = 20)[a]	WS (*n* = 21)	TYP (*n* = 19)	ASD vs. WS
Raw *M*	12.6	13.6	3.2	
SD	3.9	2.2	2.6	
Range	4–17	8–19	0–8	
Adjusted *M*	12.8	13.3		
F value				0.2
Significance				.66
Effect size (η^2)				.01
Observed power				.07
F: Stereotyped Language				
Raw *M*	8.8	6.4	1.0	
SD	3.0	2.5	1.1	
Range	3–14	2–12	0–3	
Adjusted *M*	9.3	6.0		
F value				13.5
Significance				.001
Effect size (η^2)				.26
Observed power				.95
G: Use of Context				
Raw *M*	12.2	11.0	2.1	
SD	4.2	3.5	1.7	
Range	6–19	5–17	0–5	
Adjusted *M*	12.7	10.5		
F value				2.5
Significance				.12
Effect size (η^2)				.06
Observed power				.35
H: Nonverbal				
Raw *M*	11.1	6.3	1.3	
SD	3.3	2.5	1.2	
Range	4–16	1–14	0–5	
Adjusted *M*	11.6	5.9		
F value				35.1
Significance				<.001
Effect size (η^2)				.47
Observed power				1.0
I: Social Relations				
Raw *M*	10.2	5.6	1.0	
SD	3.7	2.9	1.1	
Range	2–16	0–11	0–5	
Adjusted *M*	10.4	5.4		
F value				19.3
Significance				<.001

CCC–2 subscale	ASD (*n* = 20)[a]	WS (*n* = 21)	TYP (*n* = 19)	ASD vs. WS
Effect size (η^2)				.33
Observed power				.99
J: Interests				
Raw *M*	11.5	10.5	2.4	
SD	4.0	4.4	1.6	
Range	5–20	0–18	0–6	
Adjusted *M*	11.8	10.2		
F value				1.1
Significance				.31
Effect size (η^2)				.03
Observed power				.17

[a]One ASD participant's CCC–2 data were removed as a function of extreme outlier CCC–2 summary scores.