

Research Note

Pragmatic Language Assessment in Williams Syndrome: A Comparison of the Test of Pragmatic Language—2 and the Children's Communication Checklist—2

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Purpose: Individuals with Williams syndrome (WS) are recognized as having a strong desire for social relationships, yet many of them have difficulty forming and maintaining peer relationships. One cause may be impairments in pragmatic language. The current study compared the assessment of pragmatic language skills in individuals with WS using the Test of Pragmatic Language—Second Edition (TOPL-2; Phelps-Terasaki & Phelps-Gunn, 2007) and the Children's Communication Checklist—Second Edition (CCC-2; Bishop, 2003).

Method: Twenty children and adolescents diagnosed with WS were given the TOPL-2, and their parents completed the CCC-2.

Results: The TOPL-2 identified 8 of the 14 older children (ages 8–16 years) as having pragmatic language impairment

and all of the 6 younger children (ages 6–7 years) as having such. In comparison, the CCC-2 identified 6 of the 14 older children and 2 of the 6 younger children as having pragmatic language impairment. The older group also had a higher composite score than the younger group on the CCC-2.

Conclusion: The TOPL-2 identified significantly more participants as having pragmatic language impairment than did the CCC-2. The TOPL-2 may be more useful in assessing pragmatic language in older children than younger children. The results offer important preliminary clinical implications of language measures that may be beneficial in the assessment of individuals with WS.

Key Words: pragmatics, assessment, Williams syndrome, developmental disabilities

Williams syndrome (WS) is a neurodevelopmental disorder with an estimated prevalence of 1 in 7,500 (Strømme, Bjørnstad, & Ramstad, 2002). The underlying cause of this disorder is the deletion of ~26 genes on chromosome 7 (Peoples et al., 2000). Individuals with WS are described as having mild to moderate intellectual deficits, heart disease, and distinctive facial features (Bellugi, Klima, & Wang, 1996; Mervis, 2006; Pober & Dykens, 1996). They are also noted as having an extremely friendly and outgoing demeanor that is typically referred to as *hypersocial* behavior (Bellugi, Lichtenberger, Jones, Lai, & St. George, 2000; Jones et al., 2000). Initial research claimed that individuals with WS had intact language in the face of severe cognitive deficits (Jackendoff, 1994; Piatelli-Palmarini, 2001; Pinker, 1999).

However, more recent studies have demonstrated that there is a wide range of cognitive functioning within individuals with WS, and that language is far from intact. Approximately 75% of this population have full-scale IQs that are consistent with intellectual deficits; the remaining 25% have learning disabilities (Mervis & Klein-Tasman, 2000).

Individuals with WS typically have overall language skills that are commensurate with their mental age, although there are some relative strengths and weaknesses. Receptive vocabulary and articulation are considered to be areas of strength, as individuals with WS often perform better than expected given their mental age (Brock, Jarrold, Farran, Laws, & Riby, 2007; Laws & Bishop, 2004). However, expressive vocabulary and certain syntactic structures (e.g., relative clauses) are often relatively weak, with scores on assessments such as the Expressive Vocabulary Test (EVT; Williams, 1997) and the Test for Reception of Grammar (TROG; Bishop, 1989) below that of chronological age-matched peers matched for language (Grant, Valian, & Karmiloff-Smith, 2002; Mervis, Robinson, Rowe, Becerra, & Klein-Tasman, 2003; Stojanovic & Ewijk, 2008). One area of language that is particularly problematic for individuals with WS is pragmatics. *Pragmatics* refers to the use of language in a social context. Successful interactions require individuals to

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understand and follow pragmatic rules. These rules include topic maintenance, appropriate conversational turn-taking, following another person's conversational lead, and understanding metaphorical and figurative language (e.g., non-literal language such as "it's raining cats and dogs"; Berko-Gleason & Ratner, 2009).

Despite the initial impression of increased sociability, individuals with WS typically struggle with social interactions. Research has noted pragmatic difficulties including poor turn-taking, reduced topic maintenance, and perseveration on personal topics (Laws & Bishop, 2004; Philofsky, Fidler, & Hepburn, 2007). Individuals with WS also have difficulty with nonliteral language as well as with integration in narratives, often introducing irrelevant personal information and social fillers to conversations (Reilly, Harrison, & Klima, 1995; Reilly, Losh, Bellugi, & Wulfeck, 2004; Stojanovik, 2006; Stojanovik, Perkins, & Howard, 2004; Sullivan, Winner, & Tager-Flusberg, 2003). Some of these difficulties may stem from underlying weaknesses in theory of mind (ToM), which refers to a person's ability to understand the perspective of someone else. Several studies have indicated that individuals with WS are extremely delayed in their acquisition of ToM, thereby reducing their ability to have successful conversations (John & Mervis, 2010; John, Rowe, & Mervis 2009; Tager-Flusberg & Sullivan, 2000). Unfortunately, despite these individuals' significant struggles with social language, their relatively strong receptive language skills can put them at risk of being discharged from speech and language services. It is especially important that these areas are targeted aggressively and as early as possible given that recent evidence indicates that early pragmatic delays will lead to pragmatic difficulties later in life (John, Dobson, Thomas, & Mervis, 2012).

Assessing Pragmatic Language

It is likely that these pragmatic deficits (e.g., poor turn-taking, difficulty with topic maintenance, and perseveration) are in part responsible for what is known as the *paradox of WS*. Although these individuals' social drive and empathic qualities are strong, they often have difficulty maintaining relationships with their peers and tend to make inappropriate social judgments (Tager-Flusberg & Sullivan, 2000). In order to effectively target these specific areas of need, it is necessary to accurately assess pragmatic language impairment (PLI) in this population. Pragmatic assessment is challenging due to the complexity of this area of language. Pragmatics has been described as the intersection of social, linguistic, cognitive, and cultural influences (Adams, 2001). An effective tool for assessing pragmatic language skills needs to be able to distinguish between pragmatic differences stemming from normal variations from these influences and true pragmatic disorders. Two types of assessment tools are parental/caregiver checklists and individually administered standardized tests; each has benefits and drawbacks.

Parental/Caregiver checklists. It is common for parental/caregiver checklists to be used when assessing pragmatic language as they allow for the assessment of behaviors that are difficult to elicit in a testing environment,

or that may fluctuate from day to day (Bishop, 1998). One commonly used checklist is the Children's Communication Checklist—2 (CCC—2; Bishop, 2003, 2006) and its predecessor, the Children's Communication Checklist (CCC; Bishop, 1998). These measures have been used in several studies analyzing pragmatic abilities in individuals with WS as well as those with other developmental disabilities (Laws & Bishop, 2004; Philofsky et al., 2007). Laws and Bishop (2004) found that the CCC was effective in distinguishing differences in pragmatic ability between a group of children with WS, a group with Down syndrome (DS), and a group with specific language impairment (SLI).

Philofsky et al. (2007) used the CCC—2 to directly compare pragmatic functioning between children with autism spectrum disorder (ASD) and children with WS. They found that although both groups had difficulty with inappropriate initiations and poor use of context, the WS group performed significantly better than the ASD group. Volden and Phillips (2010) used the Test of Pragmatic Language (TOPL; Phelps-Terasaki & Phelps-Gunn, 1992) and the CCC—2 to compare the identification rates of PLI in children and adolescents with ASD who had structural language skills and nonverbal IQ in the normal range. They found that the CCC—2 identified more children with ASD as having PLI than did the TOPL. Volden and Phillips suggested that the difference in PLI identification rates between the two measures occurs because the CCC—2 gathers information about patterns of behavior seen over an extended period of time, whereas the TOPL offers more of a snapshot of pragmatic functioning at that moment. Their results indicated that the CCC—2 is a valid assessment to use with individuals who have developmental disabilities, particularly those who are at risk of pragmatic difficulties. However, they noted that use of a parent report is not sufficient to determine eligibility for services, and as such, the additional use of a standardized assessment needs to be considered.

Individually administered standardized tests.

Individually administered standardized tests have several benefits: They allow an individual's performance to be compared to the performances of others, they are typically brief to administer, and they are often considered to be more objective than other forms of assessment (Adams, 2001). However, individually administered standardized tests carry with them the risks of missing certain behaviors that may present rarely, and the testing setup may alter the normal communicative behavior of the individual being assessed (Bishop, 1998; McCauley, 2001).

There are a limited number of individually administered standardized tests for pragmatic language. One that has been used most frequently in studies is the TOPL. Young, Diehl, Morris, Hyman, and Bennetto (2005) used the TOPL in conjunction with the Strong Narrative Assessment Procedure (SNAP; Strong, 1998) to assess pragmatic language in children with ASD and typically developing (TD) children. They found that the TOPL was able to discriminate between the two groups, but the SNAP was not. As mentioned previously, Volden and Phillips (2010) compared the identification rates of PLI using the TOPL

versus the CCC-2 for individuals with ASD. They found that the TOPL did not identify as many participants with ASD as having PLI as did the CCC-2.

Need for Current Study

WS has been compared to ASD in several studies (e.g., Laws & Bishop, 2004; Philofsky et al., 2007), with the overall conclusion being that these groups share some of the same pragmatic difficulties. To our knowledge, there has not been an individually administered standardized test of pragmatic language used with the WS population. However, other clinical groups with similar pragmatic deficits have been assessed using the TOPL, and beneficial information has been obtained. Given that PLI is considered a trademark of an ASD diagnosis, all of the participants with ASD should have had scores indicating PLI on both of the assessments, the TOPL and the CCC-2. In light of the similarities between pragmatic skills in ASD and in WS, a pragmatic assessment that is effective for individuals with ASD might also be useful for individuals with WS.

Whenever possible, it is best to combine parent report with another assessment as this provides a more complete picture of the child (Bishop & McDonald 2009). The CCC-2 is often used to identify social interaction difficulties, but there are aspects of pragmatic language that it does not measure. Namely, it does not provide much information about how an individual understands social situations. The TOPL (and TOPL, Second Edition; TOPL-2; Phelps-Terasaki & Phelps-Gunn, 2007) measures the ability of an individual to view a social situation as an objective bystander and to make judgments from that vantage point. These metapragmatic skills are beyond what the CCC-2 examines and may help evaluate PLI in individuals with WS. Given the difficulty that individuals with WS experience with forming and maintaining social relationships, it is vital to increase the understanding of pragmatic skills in this population.

We designed the current study to examine the use of the TOPL-2 with individuals with WS and to compare the findings with those of the CCC-2. To this end, we asked the following questions:

- Do the scores of children with WS on the TOPL-2 and the CCC-2 differ by age (6-7 vs. 8-16)?
- What is the correlation between the TOPL-2 and the CCC-2 for children with WS? Does age group moderate this relationship for children with WS?
- Are scores of children with WS significantly different between two measures of pragmatic language (TOPL-2 and CCC-2)? Do the different tests identify different rates of children with WS as having a pragmatic language disorder?

Method

Participants

A total of 20 children and adolescents participated in this study. The participants had been genetically confirmed as having WS (per parent report), were between the ages of 6 and

16 years, and were native speakers of English. Children were excluded if they had an additional diagnosis of ASD. The participants were recruited through the Williams Syndrome Association research list. A brief description of the study was e-mailed to families in a regional WS Association, as well as those who had indicated an interest in participating in research at the 2010 Williams Syndrome Association national convention. A total of 25 families responded to these requests. Two children were excluded from the data analysis because they had a diagnosis of ASD, and three were excluded because they were unable to complete the entire assessment. Participant characteristics are provided in Table 1.

Procedure

A licensed speech-language pathologist (SLP) who was certified by the American Speech-Language-Hearing Association administered the TOPL-2 during a single testing session at either the convention or the participant's home. The participants were also administered the Kaufman Brief Intelligence Test, Second Edition (KBIT-2; Kaufman & Kaufman, 2004). The CCC-2 was given to a parent/caregiver of each participant and was scored by the same SLP.

Measures

TOPL-2. The TOPL-2 is a 43-item assessment for ages 8 to 18 years, with a 17-item version for ages 6 and 7 (Phelps-Terasaki & Phelps-Gunn, 2007). It assesses seven underlying areas of pragmatics, including physical context, audience, topic, purpose, visual-gestural cues, abstractions, and pragmatic evaluation, although separate scores are not provided for these specific areas. The test is standardized with a mean of 100 and standard deviation of 15, with higher scores indicating better pragmatic abilities. The test authors advise considering any score <90 as being below normal (Phelps-Terasaki & Phelps-Gunn, 2007). The TOPL is considered to be a well-designed assessment tool for giving an overall picture of an individual's pragmatic language (Wilkinson, 1995).

CCC-2. The CCC-2 is designed to assess pragmatics, syntax, morphology, semantics, and speech in children from 4 to 16 years of age (Bishop, 2006). This test is composed of 70 items that span 10 different subscales. These subscales include speech, syntax, semantics, coherence, inappropriate initiation, stereotyped language, use of context, nonverbal communication, Social relations, and interests. Using this checklist, a person who has known the child for at least 3 months makes a

Table 1. Study participant characteristics.

Characteristic	<i>M</i>	<i>SD</i>	Range
Age	11.7	3.7	6-16.7
IQ	74	16	53-100
Verbal IQ	74	12	56-92
Nonverbal IQ	80	18	55-109
Female, <i>N</i> (%)	15	75	

Note. IQ data are missing for two participants.

judgment as to whether the child displays a certain behavior *less than once a week or never, at least once a week but not every day, once or twice a day, several times a day, or always.*

The scoring of this test results in two overall scores: a general communication composite (GCC), which evaluates the individual's overall communication skill, and a social interaction difference index (SIDI), which specifically assesses the individual's pragmatic language. The GCC is standardized with a mean of 100 and standard deviation of 15, with higher scores indicating better communicative abilities. The SIDI is calculated by subtracting the sum of the scaled scores for the subtests dealing with the structural components of language from the sum of the scaled scores for the subtests focusing on the social components. A negative SIDI indicates that the child has a greater pragmatic impairment than would be expected given his or her structural language skills. SIDI scores ranging from -10 to 10 are considered average. No mean or standard deviation was reported for this score by the test author (Bishop, 2006).

For our study, we used the scoring criteria based on the version of the CCC-2 that is used in the United Kingdom (Bishop, 2003). It is the version that Volden and Phillips (2010) used in their study. With this version, in order to qualify as having a PLI, the participant must earn either (a) a GCC standard score <80 (indicating overall communication impairment) and SIDI <0 (indicating that language pragmatics is more impaired than language structure) or (b) a SIDI of <-15. The decision to use the U.K. scoring protocol as opposed to that of the U.S. version was due to the lack of a clear criterion in the U.S. version for indicating PLI separate from those behaviors associated with ASD (Bishop, 2003, 2006).

Statistical Analyses

Standard scores were summarized using means and standard deviations. As the TOPL-2 uses different test forms for younger and older participants, we divided the WS group into two subgroups based on age. The younger group ($n = 6$)

consisted of the 6- and 7-year-old participants; they used the shorter test form. The older group ($n = 14$) consisted of the 8- to 16-year-olds; they used the longer test form. Scores for the two tests (TOPL-2, CCC-2) were computed overall and also by age group. Due to small and uneven group sizes, t tests assuming unequal variances were used to compare average scores between the age groups. Fisher's exact test was used to compare the rates of PLI between the age groups. Pearson product-moment correlations were used to describe the relationship between the two tests (TOPL-2, CCC-2), and paired t tests were used to compare the mean scores for the two tests. Rates of PLI for the two tests were compared using the exact McNemar's test, which is appropriate for paired data arising from administering both tests to each participant and a small sample size. All statistical analyses used SAS version 9.3, and all hypothesis tests were two-sided.

Results

TOPL-2 and CCC-2 Scores by Age Group

Average summary scores for the TOPL-2 and CCC-2 are shown in Table 2, overall and by age group. The older age group scored significantly higher than the younger group on the GCC of the CCC-2 ($p = 0.03$), and their TOPL-2 scores were also higher, though not quite approaching significance ($p = 0.07$). The SIDI scores (reflecting stronger pragmatic language skills) were actually higher for the younger participants, though the difference was not statistically significant. Both the TOPL-2 and CCC-2 scores were more variable in the older group, likely due to the larger sample size in this group ($n = 14$ vs. $n = 6$). The TOPL-2 scores ranged from 67 to 86 in the younger group and from 66 to 104 in the older group. The CCC-2 scores ranged from 60 to 78 in the younger group and from 50 to 113 in the older group.

Correlation Between TOPL-2 and CCC-2

The scores of the two measures for the younger and older participants are plotted in Figure 1. The correlation

Table 2. Study participants' scores on the pragmatic language measures, overall and by age group.

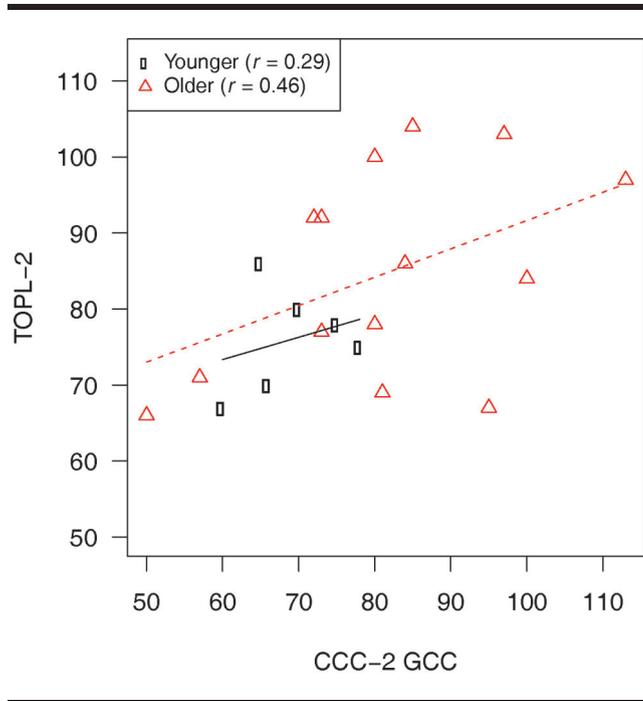
	Overall		6- to 7-year-olds		8- to 16-year-olds		p value*
Number of participants	20		6		14		
Mean scores (SD)							
TOPL-2 language quotient	82	(12)	76	(6.9)	85	(14)	0.07
CCC-2 GCC	78	(15)	69	(6.7)	81	(17)	0.03
CCC-2 SIDI	1.6	(9.0)	5.2	(11)	0.0	(7.9)	0.34
N (%) classified as impaired							
TOPL-2 ^a	14	70	6	100	8	57	0.11
CCC-2 ^b	6	30	2	33	4	29	1.00

Note. TOPL-2 = Test of Pragmatic Language—Second Edition (Phelps-Terasaki & Phelps-Gunn, 2007); CCC-2 = Children's Communication Checklist—Second Edition (Bishop, 2003); GCC = general communication composite; SIDI = social interaction difference index.

^aTOPL-2 < 90; ^bGCC score <80 and SIDI < 0, or SIDI <-15.

*Two-sample t test for means (unequal variance), Fisher's exact test for proportions.

Figure 1. Test of Pragmatic Language—Second Edition (TOPL–2; Phelps-Terasaki & Phelps-Gunn, 2007) and Children’s Communication Checklist—Second Edition (CCC–2; Bishop, 2003) general communication composite (GCC) scores by age.



between the two scores across all participants was moderate ($r = 0.51, p = 0.02$). Correlations within the two age groups were smaller in magnitude than the overall correlation ($r = 0.29$ for younger group; $r = 0.46$ for older group) but were not significantly different ($p = 0.76$). The reduced correlation in the younger group may be an artifact of the fact that smaller variability in outcomes leads to smaller estimates of correlation.

Rates of Pragmatic Language Disorder Identification

Overall, there was no difference in mean scores on the TOPL–2 and CCC–2 GCC (paired t test, Cohen’s $d = -0.31, p = 0.18$). However, the TOPL–2 identified significantly more participants as having PLI than did the CCC–2 ($p = 0.04$); impairment was identified in 14 of 20 participants (70%) using the TOPL–2 and 6 of 20 participants (30%) using the CCC–2 (Figure 2). All of the participants identified

using the CCC–2 had a GCC <80 and a SIDI <0, and none had a SIDI <–15. For the younger age group, the TOPL–2 identified impairment in 6 of the 6 participants, whereas the CCC–2 identified impairment in only 2. For the older age group, the TOPL–2 identified impairment in 8 of the 14 participants, compared to 4 from the CCC–2. Within each age group, the rates were not statistically significantly different between the two tests (younger: $p = 0.12$, older: $p = 0.29$), nor were the mean scores different for the two tests (younger: $p = 0.09$, older: $p = 0.46$), though power was severely limited due to small sample sizes in each group.

Discussion

This study is the first study to compare the TOPL–2 and CCC–2 in terms of their ability to assess pragmatic language skills in children and adolescents with WS. In respect to overall scores, there were no significant differences between scores on the TOPL–2 and those on the CCC–2 for the entire group or in the older/younger subgroups. The older group did score significantly higher on the GCC of the CCC–2 than did the younger group; this overall communicative improvement is likely due to continuing language development in the older children and adolescents. Interestingly, the two groups did not score significantly differently on the SIDI, which is the score of the CCC–2 that is directly focused on pragmatic language skills. The two assessments were moderately correlated with each other, but there were significant differences between the two assessments in the number of participants each classified as having PLI. Overall, the TOPL–2 identified significantly more participants as having PLI than did the CCC–2 (14/20 vs. 6/20).

The TOPL–2 identified every participant in the younger group as having PLI, but the responses of these participants suggest that this test may not have been appropriate for these children. Of the six children in the younger group, one had a raw score of 0, two had a raw score of 1, two had a raw score of 2, and one had a raw score of 4. The missed test items were not necessarily due to faulty pragmatic reasoning; rather, the children may not have understood the questions being asked or may have found the story that accompanied the picture overly confusing. This tendency was evidenced by all of the younger children asking for multiple repetitions of the story, saying “I don’t know,” or attempting to change the activity to something else. Thus, although each of the children in the younger group completed the entire TOPL–2, it is likely that this assessment

Figure 2. Association between classification of impairment by TOPL–2 and CCC–2.

		TOPL–2		
		Impaired	Not impaired	
CCC–2	Impaired	4	2	6 (30%)
	Not impaired	10	4	14 (70%)
		14 (70%)	6 (30%)	

Note. Exact McNemar’s test p value = 0.04.

may not accurately reflect the number of participants who actually have PLI. It is possible that the TOPL-2 could have overidentified the number of younger participants with PLI secondary to the test being too complicated for these children. Finally, given the extremely small group size of these younger participants, any conclusions should be interpreted cautiously.

The fact that the TOPL-2 identified significantly more participants as having PLI than the CCC-2 is in contrast with the findings of Volden and Phillips (2010), who studied individuals with ASD. In their study, the CCC-2 identified significantly more participants with ASD as having PLI than did the TOPL. This difference could be attributed to the use of the TOPL-2 versus the TOPL, as the TOPL used a lower cutoff score for pragmatic impairment (79), whereas the TOPL-2 scoring guidelines state that any score <90 is considered below normal (Phelps-Terasaki & Phelps-Gunn, 1992; Phelps-Terasaki & Phelps-Gunn, 2007). The change in scoring guidelines could also explain why the rate of identification was so different between the two groups despite there not being a significant difference between the overall scores on the two assessments.

Another explanation might lie in the difficulties that individuals with WS have with ToM. ToM requires that a person be able to take the perspective of someone else, which is essentially what the TOPL-2 requires of the test taker. Although it would be expected that individuals with ASD would also struggle with tasks requiring ToM (this is considered a central weakness of ASD), it is possible that the static test format allowed those individuals with age-appropriate structural language skills and nonverbal IQ to perform at a higher level. This might then be a matter of performance versus competence (i.e., the individuals with ASD in the Volden and Phillips, 2010, study were able to discern the correct response in the structured format of the TOPL-2 testing, whereas their real-time performance of those skills is more problematic, as evidenced by the lower scores on the CCC-2). Furthermore, perhaps the parents of the two groups have varying perceptions regarding their child's pragmatic language abilities, as measured by the CCC-2. Further research is needed to more closely examine the similarities and differences in pragmatic language skills between individuals with WS and those with ASD so that appropriate treatments are used.

Study Limitations

A limitation of this study is the number of participants, particularly in the younger age group. This small group size limited the conclusions that can be made as well as the ability to generalize the results. The different scoring practices of the two assessments are also something that should be considered when drawing conclusions, as they may have also contributed to the discrepancy between the similar scores and disparate diagnosis rates. Additionally, we were unable to administer comprehensive language assessments due to time constraints. Given the general language delay that is common in individuals with WS, as well as the effect that expressive/receptive language disorders could have on pragmatic

language, a full language assessment is recommended for future studies. This is especially true for those participants who struggled with the TOPL-2, as their difficulty with the TOPL-2 could be related to their language skills.

Conclusion

Based on the preliminary nature of this study, it is not yet possible to state which of these measures is more effective than the other for assessing pragmatic language in children and adolescents with WS. The TOPL-2 was problematic for some of the participants, but it does appear that use of the TOPL-2 with children and adolescents who have sufficiently high language skills to process the information within the questions could provide beneficial information to clinicians working with those individuals. The CCC-2 was appropriate for all of the participants but did not provide information regarding the participants' understanding of pragmatic rules. Furthermore, the scoring system of the U.S. version of the CCC-2 does not allow for practitioners to identify PLI separately from behaviors that are indicative of ASD, which necessitates use of the U.K. scoring system (the scoring used in this study).

The results of this study provide an important starting point for future research of children and adolescents with WS in terms of assessing pragmatic language. Given that all of the younger children seemed to experience difficulty with the TOPL-2, it would be beneficial to see if one of the pragmatic subtests of a different language assessment (e.g., the Clinical Evaluation of Language Fundamentals—Fourth Edition; Semel, Wiig, & Secord, 2003, or Comprehensive Assessment of Spoken Language; Carrow-Woolfolk, 1999) might yield results that could supplement the information from the CCC-2 for these younger individuals. In light of the discrepancy between the strong desire for interaction and the ability to form meaningful relationships that is so common for individuals with WS, it is extremely important that pragmatic language be understood more thoroughly and targeted more effectively in therapy. This becomes even more vital as recent research indicates that early pragmatic delays are linked to later pragmatic struggles (John et al., 2012).

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