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Expressive Language Profiles of Verbally Expressive Adolescents and Young Adults with Down Syndrome or Fragile X Syndrome

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Abstract

Purpose: This study examined the expressive language abilities of a subset of highly verbally expressive adolescents and young adults with Down syndrome (DS) and those with fragile X syndrome (FXS) for evidence of syndrome-related differences. FXS gender differences were also examined in an exploratory fashion.

Method: We evaluated 24 adolescents and young adults with DS, 17 of those with FXS, and 21 children with typical development (TD), with the groups matched on nonverbal mental age. Language ability was examined using the Oral and Written Language Scales (OWLS; Carrow-Woolfolk, 1995) and Developmental Sentence Scoring (DSS; Lee, 1974) scores derived from an oral narrative language sample.

Results: Study analyses revealed the following group differences: the FXS group outperformed the DS and TD groups on the OWLS measure; the TD group outperformed both other groups on some of the DSS measures; the FXS group outperformed the DS group on the DSS Sentence Point measure; and females with FXS outperformed males with FXS on several measures.

Conclusions: The study results contribute to the ongoing construction of the language phenotypes of individuals with DS and individuals with FXS and support the conclusion that there are quantitative rather than qualitative differences in their expressive language profiles.

Down syndrome (DS) and fragile X syndrome (FXS) are the two leading genetic causes of intellectual disability. DS affects approximately 1 in 733 infants ("Improved national prevalence estimates for 18 selected major birth defects-United States, 1999-2001," 2006). FXS affects approximately 1 in 4,000 males and 1 in 8,000 females (Crawford, Acuna, & Sherman, 2001). Virtually all individuals with DS, all males with FXS, and many females with FXS experience significant language learning difficulties (Abbeduto, Brady, & Kover, 2007). Despite the large number of individuals with DS or FXS, much remains to be learned about the specific nature of the language difficulties associated with each disorder, especially FXS. Such information is critical to gain a better understanding of the extent, nature, causes, and potential treatments of language difficulties in these populations. Additionally, a greater understanding of the similarities and differences of the behavior phenotypes of DS and FXS may help investigators gain insight into the root cause of language disorders in these populations and develop syndrome-specific interventions (Rice & Warren, 2004). Thus, the purpose of this study was to examine the expressive language abilities of verbally expressive adolescents and young adults (ages 11 through 23 years) with DS or FXS and to compare these abilities across disorders. Because grammatical aspects of language have been found to be particularly difficult for other groups of children with language learning difficulties, such as children with specific language impairment (SLI; Bedore & Leonard, 1998; Leonard, Eyer, Bedore, & Grela, 1997; Rice, Tomblin, Hoffman, Richman, & Marquis, 2004; Rice & Wexler, 1996; Rice, Wexler, & Hershberger, 1998), we chose to pay special attention to the

morphologic and syntactic abilities of individuals with DS or FXS. Given this interest, we focused on individuals who were capable of producing multiword utterances.

Down Syndrome

DS is caused by an extra copy of all or part of chromosome 21. Beginning at early developmental stages, delays in overall cognitive functioning and language development are present for children with DS (Berglund, Eriksson, & Johansson, 2001). Relative to age-matched peers, language delays are apparent across all domains (Chapman & Hesketh, 2000), with expressive delays more severe than receptive language delays (Dykens, Hodapp, & Evans, 1994). These language delays persist well into adolescence and adulthood (Chapman, Hesketh, & Kistler, 2002; Chapman, Seung, Schwartz, & Bird, 1998; Thordardottir, Chapman, & Wagner, 2002).

Compared to children with typical development (TD) with similar mental ages, individuals with DS demonstrate significant deficits in expressive language ability on both syntactic and morphological measures, including mean length of utterance (MLU), number of different words, and total number of words in both conversational (Chapman et al., 1998; Price et al., 2008; Rosin, Swift, Bless, & Kluppel Vetter, 1988) and narrative (Boudreau & Chapman, 2000; Chapman et al., 1998) contexts. In similar comparisons, individuals with DS also show language weaknesses on more fine-grained measures of grammar, including the use of specific tense-related (e.g., past tense, third person singular, and modals) and non-tense-related (e.g., present progressive -ing, plural -s, and possessive -z) inflectional forms (Chapman et al., 1998; Eadie, Fey, Douglas, & Parsons, 2002).

Investigators have also examined the Index of Productive Syntax (IPSyn; Scarborough, 1990) scores in the evaluation of the use of specific syntactic constructions (Price et al., 2008). With IPSyn, language samples are scored for the presence of 56 different syntactic constructions. For each construction, a score of 0, 1, or 2 is assigned to indicate no uses of the form, a single use of the form, or two or more uses of the target form, respectively. Scores across the 56 constructions are summed to yield an overall IPSyn score. Subsets of scores are also summed to evaluate specific syntactic domains, including noun phrases, verb phrases, question and negation constructions, and sentence structure. Price et al. (2008) used IPSyn scores to evaluate conversational language obtained during the administration of the Autism Diagnostic Observation Schedule (ADOS; Lord, Rutter, DiLavore, & Risi, 2002). In the Price et al. investigation, boys aged 2 through 7 years with TD significantly outperformed boys aged 2 through 14 years with DS on each of the IPSyn measures: IPSyn Total (d = 1.91), Noun Phrases (d = 1.50), Verb Phrases (d = 1.73), Questions and Negations (d = 1.15), and Sentence Structure (d = 1.67), after controlling for differences in nonverbal mental age.

This pattern of individuals with DS demonstrating poorer language skills compared to younger children with TD with similar nonverbal mental ages, however, does not hold across all studies and across all comparison groups. For example, studies examining expressive language abilities within narrative contexts generally yield higher estimates of syntactic competence in DS. Thordardottir et al. (2002) found no differences between adolescents with DS and children with TD with similar MLUs on the proportion of complex sentences in their expressive narratives. Keller-Bell and Abbeduto (2007) also found no differences between adolescents with DS and younger children with TD matched on nonverbal mental age on measures of MLU, number of different words, and clausal density. Each of these measures was derived from a narrative language sample elicited from a wordless picture book. It has been argued (e.g., Abbeduto, Benson, Short, & Dolish, 1995) that a narrative context “pulls” for more complex syntax than does a conversational context; however, it is possible that other conversational and narrative contextual differences (e.g., the visual support provided by a book) may help

individuals with DS appear more capable in the syntactically more demanding narrative context. Thus, although it is evident that individuals with DS have significant language delays, the full extent and nature of these weaknesses require further investigation.

Fragile X Syndrome

FXS is caused by excessive repeats of a trinucleotide (CGG) sequence in the fragile X mental retardation 1 (FMR1) gene on the X chromosome. Cognitive delays, as well as receptive and expressive language learning difficulties, are present at the earliest stages in development for many children with FXS (Roberts, Mirrett, & Burchinal, 2001). Moreover, the language learning difficulties of many individuals with FXS have been found to persist well beyond early development and to affect almost all areas of language performance with expressive language abilities appearing to be more negatively impacted than receptive abilities (Philofsky, Hepburn, Hayes, Hagerman, & Rogers, 2004; Roberts et al., 2001). Most studies of the language abilities of individuals with FXS, however, have focused exclusively on males with FXS, who have been found to be more consistently and severely affected, on average, than females with FXS on virtually all measures of neurocognitive functioning (see Abbeduto, Brady, & Kover, 2007).

The few studies conducted on the morphological and syntactic skills of children and adolescents with FXS have found that individuals with FXS generally have significantly poorer skills than younger children with TD with similar nonverbal mental ages. For example, studies examining conversational language samples acquired during the ADOS found that males with FXS have significantly lower MLUs and mean number of different words than children with TD after controlling for nonverbal mental age and maternal education levels (Price et al., 2008; Roberts et al., 2007).

Similar to the more detailed examinations of the morphologic and syntactic abilities of individuals with DS, investigators have also used IPSyn measures to describe the language of individuals with FXS. In an early study of the conversational language abilities of males with FXS between the ages of 5 and 36 years, Sudhalter, Scarborough, and Cohen (1991) found the overall IPSyn scores of the males with FXS to be comparable to previously documented scores of younger preschoolers with TD with similar MLUs (Scarborough, 1990). In contrast, more rigorous studies specifically designed to compare the morphologic and syntactic language abilities of boys with FXS (without autism) to younger boys with TD controlling for nonverbal mental age have uncovered significant differences between the groups (Price et al., 2008; Roberts et al., 2007). The Price et al. and Roberts et al. studies included overlapping samples of boys with FXS between the ages of 2 and 15 years and preschool boys with TD between the ages of 2 and 7 years. Based on conversational language samples obtained during the administration of the ADOS, the boys with TD outperformed the boys with FXS on almost all of the IPSyn measures, including IPSyn Total score ($d = 1.17$), Noun Phrases ($d = 1.01$), Verb Phrases ($d = 1.09$), and Sentence Structure ($d = 1.26$). No differences between the TD and FXS groups were found on the Questions and Negations composite.

Although less frequently studied than conversational language, examinations of morphological and syntactic measures based on narrative language samples have not revealed differences between individuals with FXS and developmentally matched children with TD. Keller-Bell and Abbeduto (2007) evaluated the narrative language abilities of adolescent males and females with FXS. Compared to younger children with TD matched on nonverbal mental age, no significant differences were found between the adolescents with FXS and those with TD on microstructural measures such as MLU, percent of grammatical C-units, clause density, and mean number of causal and conditional connectors. Thus, there are inconsistent findings across studies, which may be due to differences in participant age, measures of syntax used, or sampling context.

Group Comparisons

There are only a handful studies that have directly compared the expressive language abilities of individuals with DS and individuals with FXS. In the only known study to compare the conversational language abilities of individuals with DS and individuals with FXS, Price and her colleagues (2008) found that although both groups of individuals performed at significantly lower levels than children with TD with similar nonverbal mental ages on syntactic and morphological measures, individuals with FXS tended to perform at higher levels on these measures than individuals with DS. Specifically, Price et al. found a significant difference between children and adolescents with DS (*M* age = 9.2 years) and those with FXS (*M* age = 9.9 years) on MLU and the IPSyn Total score, with the latter group outperforming the former group (*d*s = .75 and .67, respectively). No group differences were found, however, on the individual IPSyn composites of Noun Phrases, Verb Phrases, Questions and Negations, and Sentence Structure.

Keller-Bell and Abbeduto (2007) analyzed narrative language samples to compare the morphological and syntactic abilities of adolescents and adults with DS to adolescents and adults with FXS. The participants in this study were relatively older than the participants in the Price et al. (2008) study, with the participants with DS having a mean age of 16.83 years and the participants with FXS having a mean age of 16.68 years. The groups were compared on a number of different measures of linguistic complexity in their narratives, including number of communication units (C-units), number of different words, MLU, percent of grammatical C-units, clause density, and mean number of connectors per C-unit. The only significant group difference detected was for the percent of grammatical C-units, with the participants with FXS producing proportionally more grammatical utterances than the participants with DS. Unlike the Price study, no group differences were found on MLU or the other linguistic complexity measures. Again, a different performance pattern across diagnostic groups emerged for conversational and narrative contexts.

Current Study

In light of the discrepant finding across grammatical measures, sampling contexts, and studies, the purpose of the current study was to gain a better understanding of the expressive language abilities, including grammatical abilities, of individuals with DS and FXS. Such information is critical for both scientific and clinical reasons. First, determining the differences and similarities between the language phenotypes of these two syndromes will help to clarify which features of the phenotype are syndrome-specific and which are a reflection of intellectual disability per se. Second, a more complete examination of the language strengths and weaknesses of adolescents and young adults with DS and those with FXS, two groups traditionally under-represented in language studies, is critical for clinical purposes in order to design and implement appropriate, and perhaps, syndrome-specific language intervention programs. Third, comparisons of these syndromes, which result from very different genetic anomalies, will provide insights into the nature of the biological constraints on language development more generally.

With these overarching purposes in mind, in the current study, the expressive language performance of adolescents and young adults with DS and the performance of adolescents and young adults with FXS were compared to children with TD with similar nonverbal mental ages. The performance of the participants in the DS group was also compared with the performance of the participants in the FXS group. Additionally, because of the scant focus on females with FXS, the expressive language performance of females with FXS was compared to males with FXS; however, these analyses were exploratory because few females were tested. We included both a standardized measure of a broad range of expressive skills and more focused measures of grammar.

Similar to the Price et al. (2008) study, grammatical language ability was evaluated by use of a grammatical coding system. However, because the participants in this study were on average 7 years older than the participants in the Price et al. study, Developmental Sentence Scoring (DSS; Lee, 1974) rather than IPSyn scores was used to evaluate grammatical complexity. Like IPSyn, DSS is a coding system that considers linguistic performance across grammatical categories. DSS consists of eight grammatical categories: (1) Indefinite Pronoun/Noun Modifier, (2) Personal Pronoun, (3) Main Verb, (4) Secondary Verb, (5) Negative, (6) Conjunction, (7) Interrogative Reversal in Questions, and (8) Wh-Questions. In each category, forms are assigned scores ranging from 0 to 8, with higher scores indicating the use of more complex later-developing grammatical forms. DSS also includes a Sentence Point score which evaluates the average number of utterances that meet standard adult grammatical rules. The DSS performance of children with TD has been shown to be significantly and positively correlated with both MLU and IPSyn performance (Rice, Redmond, & Hoffman, 2006). Unlike MLU, however, DSS allows for the analysis of specific grammatical categories. In contrast to IPSyn, DSS entails assigning scores to each occurrence of a grammatical form rather than only the first two occurrences; thus, a wider range of scores are possible. These differences may allow DSS to capture more subtle differences in grammatical complexity when comparing the performance of different diagnostic groups in contrast to MLU or IPSyn.

As the foregoing review indicated, sampling context impacts expressive language performance and the pattern of group differences. In the studies reviewed, group differences on language measures have been more robustly detected when using conversational samples. However, compared with conversational language samples, narrative language samples have been found to elicit more grammatically complex language from individuals with intellectual disability (Abbeduto et al., 1995), but with fewer group differences emerging. Such findings suggest that group differences in conversation reflect performance differences, rather than differences in the upper bound of grammatical capabilities. Because of this study's focus on probing grammatical abilities, it was important that our measures be based on the most complex language that could be obtained. Thus, we decided to utilize narrative language samples instead of conversational language samples. Additionally, we used DSS coding, which we believed would be more sensitive to group differences in narrative samples relative to previous studies.

Study Questions and Predictions

This study was designed to address four questions:

1. How do the expressive language profiles of adolescents and young adults with DS who produce multiword utterances compare to those of children with TD at similar cognitive-developmental levels?
2. How do the expressive language profiles of adolescents and young adults with FXS who produce multiword utterances compare to those of children with TD at similar cognitive-developmental levels?
3. How do the expressive language profiles of adolescents and young adults who produce multiword utterances differ across DS and FXS, controlling for cognitive-developmental level?
4. How do the expressive language profiles of adolescents and young adult females with FXS compare to those of adolescents and young adult males with FXS?

The variables used to evaluate the expressive language profiles for each study question included scores from the Oral Expression Scale of the Oral and Written Language Scales (OWLS; Carrow-Woolfolk, 1995), which is a global measure of expressive language performance and the DSS Sentence Point and Total measures, which are grammar-specific measures of expressive language. Group comparisons were also completed for specific DSS subcategories:

Indefinite Pronoun/Noun Modifier, Personal Pronoun, Main Verb, Conjunction, and Negative. All measures, except the OWLS were based on a narrative language sample. It was predicted that significant group differences would be found based on all expressive language measures with the TD group outperforming the DS and FXS groups. Additionally, it was predicted that the FXS group would outperform the DS group on each of the expressive language measures and in the exploratory gender analyses, that females with FXS would outperform males with FXS.

Method

Participants

This study included three groups of participants: adolescents and young adults with DS, adolescents and young adults with FXS, and younger children with typical cognitive development (TD). The participants with DS and the participants with FXS were recruited through newspaper advertisements, postings in newsletters and on internet websites of regional and national advocacy organizations for individuals with developmental disabilities, a university-based registry of families with a son or daughter with a developmental disability, and mailings to special educators and genetic clinics. Because of prevalence differences, the participants with FXS were recruited from a larger geographic area than the participants with DS. The participants with TD were all recruited locally through public postings, a university-based registry of school-aged children, and area preschools.

The participants in the current study came from a pool of 236 individuals (77 DS; 55 FXS; 104 TD) who were enrolled in a study focused on language development of individuals with DS or FXS and thus, the present participant samples overlap to some degree with those in analyses reported elsewhere (Abbeduto et al., 2003; Abbeduto et al., 2006; Keller-Bell & Abbeduto, 2007; Lewis et al., 2006). As part of the larger study, participants completed an extensive battery of cognitive and language tests, only some of which were analyzed in the present study. Because a particular focus of this study was to examine the morphological and syntactic abilities of individuals with DS and FXS, it was important for all of the adolescents and young adults included in the study to provide a sufficient language corpus to analyze and to be using complex language forms. Thus, to be included in the present study participants were required to have produced 50 or more complete and intelligible C-units based on a narrative language sample and to have a mean length of utterance greater than 3.0 based on the number of morphemes per C-unit in a narrative language sample. We chose this C-unit cutoff to increase the homogeneity of the sample. In most previous studies, small sample sizes combined with the wide range of language abilities represented makes it difficult to interpret null findings. In particular, inclusion of participants with low MLUs and thus, limited syntactic skills increases the likelihood of “floor effects.” Additionally, to adequately match across groups it was necessary to set an upper nonverbal mental limit of 10 years based on the Copying, Pattern Analysis, and Bead Memory subtests of the Stanford-Binet, 4th edition (Thorndike, Hagen, & Sattler, 1986) because a few girls with FXS, but none of the participants with DS had mental ages exceeding this limit. Knowing these limits affect generalizability of our findings, we felt this criteria was essential to fairly evaluate the existence of syndrome-specific grammatical profiles.

To be included in the study, it was also necessary that the participant completed each of the study measures, demonstrated normal to no more than a mild hearing loss (i.e., pure-tone average across 500, 1000, and 2000 Hz less than or equal to 40 dB) in at least one ear, and only speak English. Participants were excluded if they met diagnostic criteria for autism (for more details see Lewis et al., 2006). Additionally, for the children with TD, parents had to indicate that their children had no diagnosed disability and that they were not receiving special education services other than speech articulation therapy.

Of the original 236 participants, 50 individuals (15 DS; 3 FXS; 32 TD) were excluded because they did not complete all study measures; 102 individuals (34 DS; 22 FXS; 46 TD) were excluded because their mean length of utterance was less than 3.0 and/or their narrative language sample did not include 50 complete and intelligible utterances; 5 males with FXS were excluded for meeting autism diagnostic criteria, 5 females with FXS were excluded for having a mental age greater than 10 years; and an additional 8 individuals (4 DS; 2 FXS; 2 TD) were excluded for failing to meet other study inclusion criteria. A total of 66 individuals met the participation criteria: 24 individuals with DS, 18 individuals with FXS, and 24 children with TD. However, one male with FXS was excluded from the study because he was considered an outlier. He had over 400 complete and intelligible C-units, which was well over the group's mean of 70 C-units. Additionally, three children reported to have typical development received standard scores on the nonverbal Stanford-Binet composite less than 80 and were also excluded from the study. Thus, a total of 24 individuals with DS (mean age = 16.9 years), 17 individuals with FXS (mean age = 15.79 years), and 21 children with TD (mean age = 4.82 years) were included in this study. It is clear from the large number of participants excluded from the study that this criterion yielded a particular subset of adolescents and young adults with DS or FXS who had relatively high levels of expressive language abilities. Thus, our results should be viewed as generalizable only to higher-functioning individuals with DS or FXS producing multiword utterances, although it should be noted that all study participants with DS or FXS had IQs in the range of intellectual disability.

The participant characteristics for each study group are presented in Table 1. Analyses of variance (ANOVA) comparing the groups on key characteristics and study entry criteria including chronological age, nonverbal mental age, and the number of complete and intelligible C-units in the narrative sample were conducted. No significant differences were found for nonverbal mental age or number of different C-units. Both of these analyses yielded p-values greater than .50, indicating that the diagnostic groups could be considered to be well matched on these measures (Mervis & Robinson, 2003). As was expected, group differences were identified for chronological age and nonverbal intelligence. In terms of chronological age, the TD group was significantly younger than both the DS and FXS groups. For nonverbal intelligence, the TD group's values were significantly higher than both the DS and FXS groups. No significant differences were detected between the DS and FXS groups (ps > .17) for chronological age, nonverbal mental age, nonverbal IQ score, and number of C-units. Additionally, chi-square analyses of group differences based on gender, race, and maternal education yielded no significant group differences.

Based on genetic test results provided by parents, DS was due to trisomy 21 for 17 of the participants with DS. For one participant with DS, testing revealed translocation, and for six participants genetic testing results were unavailable, although each parent indicated that genetic testing had been completed confirming the DS diagnosis. For all of the participants with FXS, molecular genetic testing revealed that they had the full mutation with four individuals identified as being mosiac.

The characteristics of the male and female participants in the FXS group are presented in Table 2. Significant differences based on gender were identified for nonverbal mental age, nonverbal intelligence standard scores, and mean length of utterance. On each measure, the female participants outperformed the male participants. This finding is consistent with reports of males with FXS being more severely affected, on average, than females with FXS (Hagerman, 1999). The gender comparison for chronological age approached a conventional significance level (p = .06), with the females with FXS having a higher mean age than the males. Based on chi-square analyses, the gender groups did not differ based on race and maternal education.

Procedures

Prior to completing any of the study testing, parents of the participants signed consents that were approved by an Institutional Review Board of the University of Wisconsin-Madison. In most cases, participants were tested across two sessions that occurred in a single day with a 1- to 2-hour break between sessions or over two separate days. If the sessions were conducted on separate days, no more than 3 weeks lapsed between test days. A quiet room designated for study testing was used to test participants individually with parents having the option to view through an observation window. For each participant, tests were administered by a single examiner.

Study Measures

Nonverbal Intelligence

Stanford-Binet Intelligence Scale, 4th Edition (Thorndike et al., 1986): Nonverbal cognitive ability was assessed using three subtests of the Stanford-Binet Intelligence Scale: Bead Memory, Pattern Analysis, and Copying. For each of the subtests, few verbal instructions are necessary and examinees respond nonverbally. A nonverbal partial composite IQ score was derived from the standard scores from each of the subtests. Nonverbal mental age was determined by taking the mean age equivalents from each of the three subtests. This composite has been found useful in previous studies of language in DS and FXS (Abbeduto et al., 2003; Abbeduto et al., 2008; Chapman et al., 1998).

Expressive Language

Oral Expression Scale of the Oral and Written Language Scales (OWLS; Carrow-Woolfolk, 1995): The OWLS assesses a wide-range of expressive language ability including lexical, syntactic, pragmatic, and supralinguistic (e.g., figurative language, logic, inference) language. Test items require participants to answer questions, complete sentences, or generate sentences in response to oral or verbal stimuli paired with visual stimuli. Raw scores were obtained for each participant and used for study analyses. In this study, the OWLS was used as a global measure of expressive language ability.

Narrative Language Sample: An oral narrative language sample was elicited from each participant using the wordless picture book *Frog Goes to Dinner* (Mayer, 1974). Prior to telling their story, participants viewed each page of the book to get a sense of the progression of the story. The examiner then prompted the participant to start from beginning and tell the story page by page. The examiner provided minimal prompts throughout the story-telling according to a standardized script. Each narrative sample was audio-taped and transcribed by specially trained research assistants using standard Systematic Analysis of Language Transcripts conventions (SALT; Miller & Chapman, 2000). The transcripts were segmented into communication units (C-units), which include an independent clause and its modifiers (Loban, 1976). Each narrative was transcribed by a primary coder. While viewing the primary coder's transcript, a secondary coder listened to the audio-tape and marked transcription disagreements. The primary coder reviewed the disagreements, checked discrepancies against the audio-tape, and corrected the transcript as appropriate. The final corrected transcript was used for the DSS coding.

Eight (13%) of the narrative transcripts were randomly selected and transcribed by an independent coder for reliability purposes. These transcripts included three from the DS participants, three from the FXS participants, and two from the TD participants. The independent coders' transcripts were compared to the primary coders' original transcripts. The mean percent agreement for utterance segmentation was 86% (range = 78% - 94%), for number

of bound morphemes per utterance was 100% (range = 98% - 100%), and for number of words per utterance was 95% (range = 85% - 100%).

Developmental Sentence Scoring (DSS; Lee, 1974): DSS is a language analysis procedure that considers performance across a number of grammatical categories (e.g., personal pronoun, main verb, conjunctions). All of the utterances in each participant's narrative language sample were coded using DSS procedures (see Tables 1 and 2 for the range of C-units comprising the participants' samples subjected to coding). Computerized Profiling Version 9.7.0 (Long, Fey, & Channell, 2006) software was used to facilitate the coding. Only utterances that contained a subject and verb in subject-predicate order were scored. Each transcript was coded by two trained research assistants independently. A primary and secondary coder was then randomly assigned to each transcript. Discrepant scores were re-evaluated by the primary coder. The secondary reviewed the primary coder's changes and made any corrections. The primary coder then had a final chance to correct the codes. When coding the transcripts, each coder was blind to the diagnostic study group to which the participant belonged. The measures derived from the DSS coding used for study analyses included the Indefinite Pronoun/Noun Modifier Score, Personal Pronoun Score, Main Verb Score, Conjunction Score, and Negative Score. Additionally, the Sentence Point, which is an average based on the number of utterances judged to be grammatical divided by the total number of utterances, and the DSS Total, which is an average of the points awarded across categories, were also included in the study analyses.

Intra-class correlation coefficients (ICC) were used to assess the reliability of the dependent study measures. ICCs reflect a calculation of the proportion of variance in each dependent variable that can be attributed to true participant differences and those attributed to interactions between the coders and participants (Berk, 1979; Suen & Ary, 1989). ICCs can range from 0 to 1.0 with values closer to 1.0 indicating greater variance associated with true participant differences. Thus, for each DSS dependent measure, Coder 1's scores and Coder 2's scores for each participant were included in the ICC calculation. Using the consistency definition, the ICCs for the DSS measures ranged from 0.86 to 0.99, which indicate that the proportion of variance in the participants' scores associated with the coders was very small.

Statistical Design

This study involved two sets of analyses. In the first set of analyses, designed to answer Study Questions 1, 2, and 3, a separate analysis of variance (ANOVA) was conducted for each measure to assess differences in language performance across the DS, FXS, and TD study groups. In all analyses, diagnostic group (DS, FXS, TD) served as the independent variable. Levene's Test for equality of variance was completed for each analysis. Unless otherwise noted, this statistical assumption was met for each analysis ($p > .05$). Although this analysis design increased the risk of Type I error, the .05 significance level was maintained due to relatively small sample sizes. Significant ANOVAs were followed by pairwise comparisons using Tukey's Honest Significant Difference Test for unequal samples, with alpha set at .05. Effect sizes (d) were calculated and interpreted using Cohen's standards of .20 to represent a small effect size, .50 a medium effect size, and .80 a large effect size (1988).

The second set of analyses, designed to answer Study Question 4, evaluated performance differences based on gender in the FXS group. Thus, only individuals with FXS were included in these analyses. Because the study sample comprised only 5 females with FXS, these analyses were conducted for exploratory hypothesis-generation purposes using nonparametric statistics. Mann-Whitney U tests were conducted for these exploratory analyses using untransformed values.

Results

Diagnostic Group Analyses

Expressive Language—Three ANOVAs were completed to examine expressive language ability measured with the OWLS raw score, DSS Sentence Point, and DSS Total. The means, standard deviations, and effect sizes for each analysis are presented in Table 3. For the analysis of the OWLS, Levene's Test indicated significant differences in variances across groups, $F(2, 59) = 6.87$, $p = .002$; thus, a square root transformation of the data was conducted for the ANOVA. Results indicated significant group differences, $F(2, 59) = 13.82$, $p < .001$. Post-hoc analyses revealed significant differences between the FXS and DS groups ($p < .001$) and between the FXS and TD groups ($p = .02$). The comparison between the DS and TD groups just missed the significance level ($p = .06$). In this analysis, the FXS group significantly outperformed both the DS and TD groups. The Levene's Test for equality of error variance for the Sentence Point analysis was significant, $F(2, 59) = 4.34$, $p = .02$; thus, a natural logarithmic transformation was used to adjust for skewness in this variable prior to conducting the ANOVA. The ANOVA yielded a significant main effect of diagnostic group for the Sentence Point measure, $F(2, 59) = 8.15$, $p = .001$. Post-hoc analyses revealed significant differences between the DS and FXS groups ($p = .002$) and the DS and TD groups ($p = .004$). In both analyses, the DS group performed significantly poorer than the other group. There was a significant main effect of diagnostic group for the overall DSS measure ($F(2, 59) = 5.99$, $p = .004$). This effect was characterized by the TD group significantly outperforming the DS group ($p = .004$).

Developmental Sentence Score Grammatical Categories—Five ANOVAs were completed to assess expressive language ability using the DSS subcategories of Indefinite Pronoun/Noun Modifier, Personal Pronoun, Main Verb, Conjunction, and Negative scores. The means, standard deviations, and effect sizes for each of the DSS subcategory analyses are presented in Table 3. No main effects were found for the Indefinite Pronoun/Noun Modifier ($F(2, 59) = 1.43$, $p = .25$), Personal Pronoun ($F(2, 59) = .84$, $p = .44$), Main Verb ($F(2, 59) = 2.00$, $p = .15$), and Negative ($F(2, 54) = 2.87$, $p = .07$) ANOVAs. However, it is important to note that the effect sizes for comparisons of the DS and TD groups and the FXS and TD groups were medium to large for the Main Verb and Negative analyses. For both measures, the TD group had higher mean scores than the DS or FXS groups. The analysis of Conjunction scores was significant ($F(2, 59) = 4.17$, $p = .02$), with post-hoc analyses revealing that the TD group significantly outperformed the DS group on this measure ($p = .016$).

FXS Group Gender Analyses

Expressive Language—Expressive language ability of females and males with FXS based on the OWLS raw score, DSS Sentence Point, and DSS Total measures were evaluated using the Mann-Whitney *U* test. The means, standard deviations, mean ranks, *U* values, and *p* values are presented in Table 4. Significant group differences were found for the OWLS analysis ($z = -3.17$, $p = .002$) and the DSS Total analysis ($z = -2.64$, $p = .008$). In both analyses, the females outperformed the males. No significant gender differences were found based on the DSS Sentence Point analysis ($z = -1.90$, $p = .06$), although the difference favored females and approached significance.

Developmental Sentence Score Grammatical Categories—The means, standard deviations, mean ranks, *U* values, and *p* values for each of the five DSS FXS gender analyses are presented in Table 4. The Mann-Whitney *U* tests revealed that the females with FXS significantly outperformed the males with FXS on the Indefinite Pronoun/Noun Modifier ($z = -1.95$, $p = .05$) and Conjunction ($z = -2.44$, $p = .02$) analyses. No significant gender differences were found based on the Personal Pronoun ($z = -1.58$, $p = .11$), Main Verb ($z = -1.90$, $p = .06$),

or the Negative ($z = -0.31$, $p = .76$) analyses, although the former two comparisons approached significance.

Because the female FXS mean scores tended to be greater than the male FXS scores and in four cases these differences reached conventional levels of significance, the Group analyses addressing Study Questions 1, 2, and 3 were conducted again omitting the five females from the FXS group. The analyses results were identical to the first set, with one exception. The OWLS performance difference between the FXS group and TD group was no longer significant ($p = .72$).

Discussion

This study aimed to gain a better understanding of the expressive language abilities of adolescents and young adults with DS and adolescents and young adults with FXS. The language skills of these groups were evaluated by comparing the language performance of verbally expressive adolescents and young adults with DS and those with FXS with a group of younger children at similar cognitive-development levels. An additional aim of this study was to begin exploration of FXS gender differences in expressive language. Group comparisons for each study aim included broad expressive language measures as well as more specific DSS measures derived from a narrative language sample.

DS Expressive Language Profile

It was predicted that for each of the global expressive language measures as well as for each of the DSS grammatical categories, that the TD group would outperform the DS group. This prediction held true for the global DSS Sentence Point and DSS Total measures. Although comparison between the DS and TD groups just failed to reach a conventional the level of statistical significance based on the OWLS raw score, this contrast resulted in a large effect size. Contrary to our prediction, in the analyses of the DSS subcategories, significant differences between the DS and TD group only emerged for the DSS Conjunction mean score. However, it is important to note that the effect sizes for the group comparisons for both the Main Verb and Negative subcategories were medium in size characterized by smaller DS group means, indicating that the lack of significance potentially could be attributed to low statistical power. Overall, these findings suggest that individuals with DS have significant weaknesses in expressive language including grammaticality and sentence complexity, and suggest particular difficulty in verb and negation usage.

These results support previous findings identifying weaknesses in expressive language ability based on more global measures in both conversational and narrative contexts (Boudreau & Chapman, 2000; Chapman et al., 1998; Price et al., 2008; Rosin et al., 1988), but run counter to the findings of Keller-Bell and Abbeduto (2007) and Thordardottir et al. (2002) who examined language in narrative contexts and failed to find significant DS and TD group differences. One possible reason for the difference between our findings and the findings of Keller-Bell and Abbeduto and Thordardottir et al. could be related to the age of the participants. However, the group of studies with findings similar to ours (Boudreau & Chapman, 2000; Chapman et al., 1998; Price et al., 2008; Rosin et al., 1988) included younger participants (age range = 4 through 26 years) and the studies with discrepant findings (Keller-Bell & Abbeduto, 2007; Thordardottir et al., 2002) included participants in a similar age range as our own (age range = 12 through 23 years). Both of these age ranges overlap with our own age (12 through 23 years); thus, age alone does not seem to be the distinguishing variable.

Another difference between studies is the sampling context. All of the studies that had previously found language performance discrepancies between individuals with DS and TD comparison groups included language measures based on conversational samples; the studies

in which no difference was found included measures based on narrative samples. Thus, the sampling context appeared to be a significant factor. However, in the current study, most language measures were based on narrative samples with resulting analyses indicating significant group differences.

The most plausible explanation for the discrepant findings is related to the specific measures included in the studies. Neither the studies with similar findings nor those with discrepant findings included exactly the same measures that were analyzed in the present study. Although both the Keller-Bell and Abbeduto (2007) study and the Thordardottir et al. (2002) study, which did not find significant language performance differences between individuals with DS and younger children with TD, included measures of sentence complexity (i.e., clausal density, proportion of complex sentences), these measures are gross estimates of language ability. DSS Total takes into account the complexity of specific grammatical forms and may, in fact, be better able to capture more nuanced grammatical weaknesses than measures previously included in studies of individuals with DS. It is also important to note that the Keller-Bell and Abbeduto study (2007) included some of the same participants as the current study, which further supports the explanation of differences in measurement sensitivity. Thus, the differences we found reinforce the notion that subtle, yet clinically important, differences among groups can be missed with gross measures, especially in studies of adolescents and young adults as was the case in the Keller-Bell and Abbeduto study and the Thordardottir et al. study.

FXS Expressive Language Profile

Compared with the TD group, the FXS group was found to have a significantly higher OWLS raw score, a significantly lower mean DSS Total score, and no significant difference in mean DSS Sentence Point score or any of the subcategory scores. Although the initial OWLS analysis revealed that the individuals with FXS significantly outperformed the younger children with TD, this effect was found to be primarily driven by the females in the FXS group. When the analysis was repeated excluding the FXS female data, the difference was no longer significant. A significant difference between the FXS group and the TD group based on the DSS Total score was found both when the females with FXS were included and when they were excluded. Thus, this difference appears to be robust and reflective of reduced complexity in the expressive language of adolescents and young adults with FXS. However, it was surprising that none of the DSS subcategory analyses resulted in statistically significant group differences that would explain the difference found on the DSS Total measure. Based on the medium, but non-significant effect sizes, the most likely subcategories contributing to the DSS Total effect are Main Verb and Negative.

With a few exceptions, these findings align closely with the results of the Price et al. (2008) study and the Roberts et al. (2007) study in which younger boys between the ages 2 and 7 years with FXS earned statistically significant lower IPSyn Total, Sentence Structure, and Verb Phrase scores compared to children with TD based on conversational language samples. In contrast to these studies, we did not find significant group differences or medium/large effect sizes based on noun phrase measures (i.e., Indefinite Pronouns/Noun Modifier and Personal Pronoun subcategories). Moreover, the effect size derived from the Negative subcategory scores in the current study was large, most likely reflecting true differences; whereas the Price et al. (2008) and Roberts et al. (2007) studies failed to find significant differences between the FXS and TD groups on a similar measure (i.e., IPSyn Questions/Negations subscale).

There are several possible explanations for the discrepant findings between our study and the Price et al. (2008) and Roberts et al. (2007) studies. First, as previously described, the expressive grammatical measures in our study were based on narrative language samples, whereas the measures in the Price at al. and Roberts et al. studies were based on conversational

language samples obtained during the ADOS. Because narrative language samples have been found to elicit more complex language from individuals with intellectual impairment (Abbeduto et al., 1995), it is plausible that our narrative context "pulled" the most complex language from our participants; thus, eliminating FXS and TD group differences on most measures, particularly the noun phrase measure.

Another possible explanation for the differences between studies is related to the age of the participants. The participants in the Price et al. (2008) and Roberts et al. (2007) studies were considerably younger than the participants with FXS in the current study. Thus, it may be that as individuals with FXS age, their expressive language profiles change with noun phrases becoming more improved and with lingering weaknesses in main verb usage. Additionally, other grammatical aspects, such as negation, may become more pronounced with age. However, because this study was not designed to evaluate language growth and other studies have not examined language change in individuals with FXS, this explanation is merely speculative at this point.

The significant group differences in the current study run counter to the findings of the Keller-Bell and Abbeduto (2007) study in which no differences were found between a group of adolescents and young adults with FXS and a group of younger children with TD based on narrative language sample measures including MLU, percent of grammatical C-units, and clausal density. Both our study and the Keller-Bell and Abbeduto drew study groups from the same pool of participants and used the same language sampling context. Thus, the discrepant study findings are unlikely related to participant or context factors. Instead, the most plausible explanation is that the DSS Total measure used in the current study was more sensitive to the performance differences than the more general measures of MLU and percent of grammatical C-units included in the Keller-Bell and Abbeduto study.

The exploratory FXS gender analyses revealed that the females in the FXS group had a significantly higher OWLS raw score and a significantly higher means on the DSS Total, Indefinite Pronoun/Noun Modifier, and Conjunction scores than the males with FXS. Additionally, the *p* values for the Main Verb and Sentence Point approached the .05 level of significance. Thus, it is likely with a larger sample size, for these analyses to reach statistically significant levels. For all measures, except the Negative score, the females with FXS had higher mean scores than the males with FXS.

Although there were only five females with FXS in our sample, the differences observed were sensible given our current understanding of the phenotypic and genetic differences between females and males with FXS. The gender differences were not surprising for two main reasons. First, it is important to note that females with FXS tend to have less severe intellectual impairment than males with FXS (Hagerman, 1999), which was certainly the case in the present study. The mean nonverbal mental ages of the females and males in the present study were 7.24 and 4.38 years, respectively. We could not control for this difference because there were only five female participants with FXS. Second, although there are a limited number of studies comparing the language skills of females and males with FXS, previous examinations have found females with FXS to outperform males with FXS based on a variety of language-based measures including measures of overall language ability (Fisch et al., 1999), receptive language (Abbeduto et al., 2003), and conversational repair (Abbeduto et al., 2008). Thus, our findings extend the literature to expressive grammar and uniformly align with the findings of previous studies; however, large-scale examinations of the language profiles of females with FXS, especially controlling for gender-related cognitive differences, are still needed to gain a better understanding of FXS gender effects.

DS and FXS Expressive Language Profile Comparison

It was anticipated that the individuals with FXS would outperform those with DS based on the OWLS, DSS Sentence Point, DSS Total, and each of the DSS subcategory measures. However, significant differences between the DS and FXS group were found only on the OWLS and the DSS Sentence Point scores. In both cases, the FXS group significantly outperformed the DS group. None of the DSS subcategory analyses yielded significant group differences. These findings are very similar to the Price et al. (2008) study results in which the participants with FXS had a significantly higher mean MLU and IPSyn Total scores than the participants with DS and no group differences were found on any of the IPSyn composites. Additionally, our findings align closely with the results of the Keller-Bell and Abbeduto (2007) study that only found differences between the DS and FXS participant groups based on percent of grammatical C-units, a measure of grammaticality similar to our DSS Sentence Point measure. Thus, across studies that have used both conversational and narrative language samples, individuals with FXS have consistently outperformed individuals with DS on broad grammatical measures; however, more detailed grammatical measures included in both the Price et al. study as well as our study, have failed to identify specific areas of grammatical weakness.

We can think of two plausible explanations for this discrepancy. First, it may be the case that true differences in expressive grammatical abilities exist between individuals with DS and individuals with FXS that are not captured in the DSS subcategory coding. For example, none of the DSS categories assign points for use of plurals or articles; however, if omitted, no sentence point would be assigned, decreasing the overall Sentence Point score. Additionally, DSS excludes utterances that do not contain a main verb; thus, sentences lacking a copula are omitted from analyses precluding a thorough evaluation of copula use. Despite the exclusion of these forms in the DSS coding system, such forms are coded in the IPSyn system. Thus, if there were true difference between individuals with DS and those with FXS on such measures, we would have expected them to be revealed in the Price et al. study (2008), but this was not the case.

The second possible explanation for the discrepancy between gross and specific grammatical measures, which is consistent with both the Price et al. (2008) findings and our own, is that the expressive grammatical abilities of individuals with DS are not dramatically different from individuals with FXS and that there are not specific areas of grammatical weaknesses for individuals with DS relative to those with FXS. Rather it may be the case that the language abilities of individuals with DS are generally weaker than those of individuals with FXS and that when accounted for together are great enough to expose significant expressive language weaknesses between the two diagnostic groups. Further examination of the grammatical abilities of individuals with DS and those with FXS using even more detailed grammatical measures are needed to properly understand this discrepancy and to gain a better depiction of the similarities and differences of the language phenotypes associated with DS and FXS.

Study Strengths and Limitations

This is one of the first studies to closely examine expressive language, including grammatical complexity, in verbal adolescents and young adults with DS and verbal adolescents and adults with FXS. This study included measures based on both standardized, norm-referenced tests and narrative language samples which were coded using DSS procedures. Both the OWLS and DSS measures were sensitive to detecting differences between the study groups; however, the DSS subcategory analyses did not detect group differences. Although this study was designed to identify subtle differences in the use of grammar, the nature of the group differences found in the DSS Sentence Point and DSS Total analyses were not illuminated by the specific constructions examined. Similarly, the source of performance differences between the study groups on the OWLS remains unknown. Thus, future studies should aim to better understand

underlying mechanisms for grammatical strengths and weaknesses in individuals with FXS and DS as well as the differences in overall expressive language, grammatical errors, and grammatical complexity between these groups.

In an effort to evaluate the best language efforts of the participants, this study included measures based on narrative language samples. Previous investigations have used either conversational or narrative contexts and have yielded somewhat conflicting results. The results of the current study partially align with the studies based on conversational language (e.g., Price et al., 2008) as well as the studies based on narrative language (e.g., Keller-Bell & Abbeduto, 2007). Thus, the influence of language sampling context on language performance needs to be examined further.

Relatively little is known regarding the expressive language abilities of adolescents and young adults with developmental disabilities, especially females with FXS; thus, the participants in the current study comprise a unique under-represented sample. Despite this study advantage, the overall study sample size was modest and the female sample size was very small. However, even with these limitations, statistically significant effects were found. To a gain better understanding of the language abilities of individuals with DS and individuals with FXS it is essential to analyze the language abilities of a larger pool of individuals with FXS, especially females. To increase potential generalizability of study findings, it is also important for future samples to be equally distributed across age ranges as well as levels of cognitive and language abilities. Moreover, longitudinal studies are needed to examine language development in these populations. Ideally, such studies would begin with individuals at early language developmental stages (e.g., individuals with MLUs less than 3.0, such as those excluded from the present investigation) and trace the earlier origins of the patterns revealed in this study.

Conclusions

This study contributes to the ongoing construction of the language phenotypes of individuals with DS and individuals with FXS. Through a focus on grammar abilities, this study aimed to better characterize the language profiles of adolescents and young adults with DS and those with FXS with relatively high levels of language ability. The study results support the existence of distinct language profiles between these groups based on global expressive language measures. Specifically, we found that individuals with DS demonstrated weaker language skills compared to individuals with TD and FXS across broad expressive language measures. Additionally, compared with individuals with TD, individuals with FXS demonstrated significant weaknesses in grammatical complexity. Thus, both individuals with DS and individuals with FXS exhibited weaknesses in grammatical complexity; however, individuals with FXS appeared to have better overall use of language including grammatical language ability.

These findings suggest that DS and FXS may differ in the degree, but not the nature of their grammatical difficulties. Moreover, based on the language profiles that emerged from this study, grammatical complexity appears to be an appropriate treatment target for both verbally expressive individuals with DS and verbally expressive individuals with FXS with a focus on increasing verb, negative, and conjunction complexity. Strengthening the grammatical language weaknesses of individuals with DS or FXS is likely to facilitate the effectiveness of their communication and to have positive effects on reading and writing abilities (Mackie & Dockrell, 2004). Although our findings raise the possibility that the targets of the intervention are likely to be similar for the two groups, the specific treatment approaches best suited for each diagnostic group require future examination. Finally, the present findings suggest that the constraints on the course of language development are fairly substantial as very different genetic anomalies result in similar patterns of grammar development.

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Table 1

Participant characteristics for each diagnostic group

Characteristic	Group			*p*	*d*
	DS (n = 24)	FXS (n = 17)	TD (n = 21)		
Chronological Age (years)				< 0.001	FXS/DS: −0.34
					FXS/TD: 5.19
Mean	16.90	15.79	4.82		DS/TD: 5.29
SD	3.14	2.89	0.76		
Min-Max	12.08-23.37	11.38-21.54	3.61-6.66		
Nonverbal Mental Age (years)[a]				0.68	FXS/DS: 0.20
					FXS/TD: 0.24
Mean	4.94	5.22	4.87		DS/TD: 0.06
SD	1.04	1.65	1.21		
Min-Max	3.31-7.06	2.86-7.97	3.42-7.84		
Nonverbal IQ[a]				< 0.001	FXS/DS: 0.31
					FXS/TD: −5.49
Mean	41.71	44.35	98.00		DS/TD: −6.81
SD	6.87	10.09	9.45		
Min-Max	36-57	36-65	84-115		
Number of C-units[b]				0.77	FXS/DS: 0.14
Mean	67.42	70.29	71.43		FXS/TD: −0.05
SD	14.44	24.50	19.58		DS/TD: −0.23
Min-Max	50-96	51-151	50-121		
Gender				0.14	Φ = 0.25
Female:Male	11:13	5:12	13:8		
Race				0.09	Φ = 0.28
White:Other	24:0	15:2	17:4		
Maternal Education[c]				0.10	Φ = 0.30
High School or Less: Some College or More	10:14	7:10	3:18		

[a] Mean of the age-equivalents for the Copying, Pattern Analysis, and Bead Memory subtests of the Stanford-Binet Intelligence Scale, 4th Edition (Thorndike et al., 1986)

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[b] Based on a narrative language sample

[c] Paternal education level used for one participant in the DS group because maternal education level was unknown

Table 2

FXS group characteristics according to gender

	Female (n = 5)	**Male (n = 12)**	***U***	***p***
Chronological Age (years)			13.00	0.07
Mean	17.79	14.95		
SD	2.79	2.59		
Min-Max	14.25-21.54	11.38-19.74		
Mean Rank	12.40	7.58		
Nonverbal Mental Age (years)[a]			2.00	0.003
Mean	7.24	4.38		
SD	0.90	1.03		
Min-Max	5.78-7.97	2.86-7.11		
Mean Rank	14.60	6.67		
Nonverbal IQ[a]			4.00	0.005
Mean	56.00	39.50		
SD	8.16	6.05		
Min-Max	44-65	36-56		
Mean Rank	14.20	6.83		
Number of C-units[b]			29.50	0.96
Mean	81.40	75.67		
SD	40.69	13.83		
Min-Max	54-151	51-95		
Mean Rank	9.10	8.96		
Race			0.50	$\Phi = 0.17$
White:Other	4:1	11:1		
Maternal Education			0.95	$\Phi = 0.02$
High School or Less: Some College or More	2:3	5:7		

[a]Mean of the age-equivalents for the Copying, Pattern Analysis, and Bead Memory subtests of the Stanford-Binet Intelligence Scale, 4th Edition (Thorndike et al., 1986)

[b] Based on a narrative language sample

Table 3

*M*s, *SD*s, ANOVA *p*-values, and effect sizes for diagnostic group comparisons

Dependent Variable	Group DS (n = 24)	FXS (n = 17)	TD (n = 21)	*p*	*d*[b]
Expressive Language					
OWLS[a] (raw score)				<.001*†	DS/FXS: 1.51*†
Mean	31.75	54.06	40.14		DS/TD: 0.79†
SD	10.62	18.88	10.57		FXS/TD: 0.86*
DSS Sentence Point[a]				.001*†	DS/FXS: 0.97*†
Mean	0.72	0.84	0.83		DS/TD: 0.79*†
SD	0.14	0.09	0.06		FXS/TD: 0.39
DSS Total				004*†	DS/FXS: 0.25
Mean	6.29	6.77	8.22		DS/TD: 0.71*†
SD	1.72	2.09	1.98		FXS/TD: 0.92*†
Developmental Sentence Scoring Grammatical Categories					
Indefinite Pronoun/Noun				.25	DS/FXS: 0.44
Modifier					DS/TD: 0.39
Mean	1.71	2.05	1.95		FXS/TD: 0.16
SD	0.76	0.79	0.44		
Personal Pronoun				.44	DS/FXS: 0.16
Mean	2.10	2.15	2.22		DS/TD: 0.39
SD	0.31	0.33	0.30		FXS/TD: 0.22
Main Verb				.15	DS/FXS: 0.10
Mean	1.66	1.69	1.85		DS/TD: 0.53
SD	0.38	0.23	0.33		FXS/TD: 0.56
Conjunction				.02*†	DS/FXS: 0.50
Mean	3.82	4.47	4.85		DS/TD: 0.84*†
SD	1.45	1.14	0.96		FXS/TD: 0.36
Negative[c]				.07	DS/FXS: 0.09

Dependent Variable	Group			*p*	*d*[b]
	DS (n = 24)	FXS (n = 17)	TD (n = 21)		
Mean	4.60	4.46	5.43		DS/TD: 0.61
SD	1.66	1.29	1.00		FXS/TD: 0.84

[a] Based on untransformed values

[b] Based on transformed values, when appropriate

[c] Three participants in the DS Group and 2 participants in the FXS Group did not produce scoreable negative constructions in their narrative samples and are excluded from this analysis

[*] Significant at .05 level or better

[†] Significant when FXS females are removed from the analysis

Table 4

*M*s, *SD*s, Mann-Whitney test values, and *p*-values for FXS gender comparisons

	Females (n = 5)	Males (n = 12)	*U*	*p*
Expressive Language				
OWLS (raw score)			<.001*	0.002*
Mean	80.20	43.17		
SD	8.82	7.07		
Mean Rank	15.00	6.50		
Sentence Point			12.00	0.06
Mean	0.89	0.82		
SD	0.06	0.09		
Mean Rank	12.60	7.50		
DSS Total			5.00	0.008*
Mean	8.77	5.94		
SD	2.13	1.46		
Mean Rank	14.00	6.92		
Developmental Sentence Scoring Grammatical Categories				
Indefinite Pronoun/Noun Modifier			11.50	0.05*
Mean	2.50	1.86		
SD	0.53	0.82		
Mean Rank	12.70	7.46		
Personal Pronoun			15.00	0.11
Mean	2.41	2.05		
SD	0.24	0.31		
Mean Rank	12.00	7.75		
Main Verb			12.00	0.06
Mean	1.77	1.65		
SD	0.18	0.25		
Mean Rank	12.60	7.50		

		Females (n = 5)	Males (n = 12)	*U*	*p*
Conjunction				7.00	0.02*
	Mean	4.54	4.44		
	SD	0.83	1.29		
	Mean Rank	13.60	7.08		
Negative[a]				22.50	0.76
	Mean	4.23	4.58		
	SD	1.83	1.03		
	Mean Rank	7.50	8.25		

[a]Two male participants did not produce scoreable negative constructions in their narrative samples and are excluded from this analysis

*Significant at .05 level or better