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Macrostructural Narrative Language of Adolescents and Young Adults with Down Syndrome or Fragile X Syndrome

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Abstract

Purpose—To gain a better understanding of language abilities, the expressive macrostructural narrative language abilities of verbally expressive adolescents and young adults with Down syndrome (DS) and those with fragile X syndrome (FXS) were examined.

Method—The authors evaluated 24 adolescents and young adults with DS, 12 male adolescents and young adults with FXS, and 21 younger children with typical development (TD). Narrative samples were assessed at the macrostructural level using the Narrative Scoring Scheme (NSS; Heilmann, Miller, Nockerts, & Dunaway, 2010). Three group comparisons were made using: (a) the full sample matched on nonverbal mental age, (b) a subset of the participants individually matched on nonverbal mental age, and (c) a subset of participants individually matched on mean length of utterance.

Results—Study analyses revealed that the DS and FXS groups significantly outperformed the TD group on a limited number of NSS measures. No significant differences emerged between the DS and FXS groups.

Conclusions—Study results suggest that some aspects of macrostructural narrative language may be relative strengths for adolescents and young adults with DS and those with FXS. These results can be used to create a more nuanced and informed approach to assessment and intervention for these populations.

Language development is significantly impaired in nearly all individuals with Down syndrome (DS; Abbeduto, Warren, & Conners, 2007) as well as in most males with fragile X syndrome (FXS; Abbeduto, Brady, & Kover, 2007). DS and FXS are the two leading genetic causes of intellectual disabilities, with approximately 1 in 733 infants born with DS (“Improved national prevalence estimates for 18 selected major birth defects—United States, 1999–2001,” 2006), and 1 in 4,000 males and 1 in 8,000 females born with FXS (Crawford, Acuna, & Sherman, 2001). From an early age, children with DS and children with FXS experience difficulty and delay in their development of all aspects of expressive and receptive language compared to children of the same chronological age with typical development. However, much of the research conducted to date has focused on language skills at the utterance level (e.g., vocabulary, phrase structure). Thus, this study focused on the macrostructural narrative language abilities (e.g., use of setting, character development,

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conclusion) of verbally expressive adolescents and young adults with DS and those with FXS.

Language Development in Down Syndrome

DS results from an extra copy of all or part of chromosome 21. This genetic difference affects many aspects of cognitive development, including language. Beginning at an early age and continuing well into adolescence, language development for individuals with DS proceeds at a significantly slower pace compared to chronological-age peers with typical development (TD; Berglund, Eriksson, & Johansson, 2001). Children and adolescents with DS have difficulties across multiple language domains, including vocabulary, grammar, and pragmatics (Martin, Klusek, Estigarribia, & Roberts, 2009). Given the broad range of language learning difficulties, it is important to consider conversational and narrative language abilities of adolescents and young adults with DS at both the utterance level (i.e., microstructure) and the global, supra-utterance, level (i.e., macrostructure) (Kintsch & van Dijk, 1978).

At the microstructural level, children with DS demonstrate significant deficits compared to children with TD of similar nonverbal mental age on expressive and receptive measures of morphology and syntax, such as mean length of utterance (MLU), number of different words, and total number of words in language samples derived from conversation (Chapman, Seung, Schwartz, & Bird, 1998; Price et al., 2008; Rosin, Swift, Bless, & Kluppel Vetter, 1988). Similar microstructural deficits also are present based on narrative language samples obtained from children with DS (Boudreau & Chapman, 2000; Chapman, et al., 1998). In comparison to children with TD of similar nonverbal mental age, individuals with DS also demonstrate significantly greater language-learning weaknesses based on examinations of the use and comprehension of specific grammatical forms, such as past tense, passives, and *wh*- questions (Chapman, et al., 1998; Eadie, Fey, Douglas, & Parsons, 2002; Joffe & Varlokosta, 2007; Price, et al., 2008). Other studies have found that adolescents and young adults with DS include significantly less complex (Finestack & Abbeduto, 2010) and fewer grammatically correct (Finestack & Abbeduto, 2010; Keller-Bell & Abbeduto, 2007) utterances in their narrative language samples compared to younger TD children with similar nonverbal mental ages.

There is variability across studies, however, and individuals with DS have sometimes been found to perform no differently or in some cases significantly better than matched TD children on microstructural language measures. This is especially true of microstructural measures derived from narrative language samples obtained from adolescents and young adults. For example, Thordardottir et al. (2002) found no significant difference between adolescents with DS and children with TD matched on MLU as regards the proportion of complex sentences in their expressive narratives. In a comparison by Keller-Bell and Abbeduto (2007) involving TD children matched to adolescents and young adults with DS on nonverbal mental age, no differences were found on measures of MLU, number of different words, and clausal density derived from narrative language samples. Bird and colleagues (2008) also found no significant differences between children and adolescents with DS and TD children matched on reading ability on measures of MLU, total number of words, and number of different words in spoken narratives. There is even evidence of children and adolescents with DS outperforming comparison groups. Both Bird et al. (2008) and Boudreau and Chapman (2000) found that children and adolescents with DS included significantly more utterances in their oral narratives compared to younger TD children with similar reading abilities and similar MLUs.

Relative to utterance-level language abilities, less is known about the macrostructural language abilities of individuals with DS. Several studies have suggested that children and

adolescents with DS use a significantly greater number of unclear references (Boudreau & Chapman, 2000) and include significantly fewer thematic elements (Miles & Chapman, 2002) in their oral narrative retellings than younger TD children of similar nonverbal mental ages. However, other investigations of the expressive macrostructural abilities of individuals with DS have yielded no significant differences relative to matched comparison groups, and in many cases indicate better performance by individuals with DS. For example, the comparison of children and adolescents with DS to younger children with TD matched on reading ability conducted by Bird and colleagues (2008), yielded no significant differences between the DS and TD groups on a measure of narrative episodic structure. Keller-Bell and Abbeduto (2007) found no significant differences between adolescents and young adults with DS and TD children matched on nonverbal mental age on a number of narrative macrostructure measures, including the use of mental state verbs, character names, dialogue, repetition, and exaggeration. However, Keller-Bell and Abbeduto also found that the individuals with DS used onomatopoeia and exclamations significantly more frequently and had a greater diversity and density of narrative devices than the TD comparison group. Additionally, Roberts and colleagues (2007) found that children and adolescents with DS used significantly more instances of adequate topic maintenance during conversation than mental-age matched TD children. Moreover, there is evidence that compared to younger TD children matched on MLU, individuals with DS include significantly more events (Boudreau & Chapman, 2000) as well as plot, theme, and episodic content in their narratives (Miles & Chapman, 2002).

In summary, it appears that at the microstructure level, individuals with DS have considerable difficulty with conversational language, but have better performance in narrative contexts. Individuals with DS demonstrate some strengths in their macrostructural language abilities compared to TD children, especially in narrative production. In light of the inconsistent findings, however, further examination of the language abilities of adolescents and young adults with DS is needed.

Language Development in Fragile X Syndrome

FXS is caused by an expansion of the sequence of trinucleotide (CGG) repeats included in the *fragile X mental retardation 1 (FMR1)* gene located on the X chromosome (Hagerman, 2008). Children with FXS as young as 9 months of age exhibit delays in development that continue throughout childhood and adolescence (J. E. Roberts et al., 2009). Cognitive and language development of children with FXS occurs at a significantly slower pace than that of children with TD (Hall, Burns, Lightbody, & Reiss, 2008; Prouty et al., 1988). By age 20, many individuals with FXS use some complex sentences (Bailey, Raspa, Holiday, Bishop, & Olmsted, 2009), whereas some remain minimally verbal well into adulthood (Philofsky, Hepburn, Hayes, Hagerman, & Rogers, 2004; J. Roberts, Mirrett, & Burchinal, 2001). Research indicates that males with FXS are more consistently and severely affected, on average, than females with FXS on virtually all measures of neurocognitive functioning, which is due in part to X chromosome inactivation in females (see Abbeduto, Brady, et al., 2007).

Relative to DS, only a small number of studies have examined the language abilities of individuals with FXS. At the microstructural level, the morphological and syntactic skills of children and adolescents with FXS are generally significantly poorer than younger TD children with similar nonverbal mental ages. For example, studies examining conversational language samples have documented significantly lower MLUs and mean number of different words in males with FXS than children with TD after controlling for nonverbal mental age and maternal education (Price, et al., 2008; J. Roberts, Hennon, et al., 2007). This pattern has been found for more specific measures of morphology and syntax, such as the Index of

Productive Syntax (IPSyn; Scarborough, 1990), obtained from conversational language samples (Price, et al., 2008; Prouty, et al., 1988; J. Roberts, Hennon, et al., 2007).

Some examinations of morphology and syntax use based on narrative language samples have yielded similar results; however, other studies have not. For example, Finestack and Abbeduto (2010) found that adolescents and young adults with FXS used significantly less complex syntactic forms in their narrative language when compared to younger TD children with similar mental ages. In contrast, Keller-Bell and Abbeduto (2007) did not find significant differences between adolescents and young adults with FXS and younger mental-age matched TD children on microstructural measures, such as MLU, percent of grammatical C-units, clause density, and mean number of causal and conditional connectors. Moreover, Finestack and Abbeduto (2010) found no significant group differences between the FXS group and mental age-matched TD group based on the rate of grammatical utterances. In terms of microstructural language, it appears that children and adolescents with FXS are less likely to use complex language forms than TD children with similar mental ages in both conversational and narrative contexts; however, the forms they use tend to be grammatical.

There are three published studies that have examined the language of individuals with FXS at the macrostructural level. First, Simon and colleagues (2001) examined the ability of adult women with FXS to complete short stories/jokes with coherent endings. In that study, it was found that women with FXS made more errors in selecting endings, especially for jokes, than adult women without FXS. Second, Roberts and colleagues (2007) found few significant differences between boys with FXS and younger TD boys, after controlling for nonverbal mental age, on macrostructure measures assessing topic maintenance and change derived from conversational language samples. The only significant difference found between the FXS and TD groups in the Roberts et al. study was on the rate of elaborate topic maintenance with the TD boys outperforming the FXS boys. Third, Keller-Bell and Abbeduto (2007) found that compared to younger TD children matched on nonverbal mental age, the adolescents and young adults with FXS did not perform significantly differently as regards their use of narrative forms, such as mental state verbs, character names, and character dialogue. In summary, although individuals with FXS have difficulty with some aspects of macrostructural language (e.g., topic maintenance), they exhibit more difficulties relative to TD children of the same nonverbal mental age with language at the microstructural level.

Group Comparisons

A few of the aforementioned studies included both DS and FXS participant groups and directly compared the microstructural language abilities of individuals with DS and FXS. Most of these studies have not revealed significant group differences based on either broad (e.g., MLU, number of different words) or more targeted (e.g., IPSyn) measures of morphology and syntax (Finestack & Abbeduto, 2010; Keller-Bell & Abbeduto, 2007; Price, et al., 2008); however, a few significant differences have emerged. For example, Price and colleagues (2008) found that boys with FXS performed at significantly higher levels relative to boys with DS on IPSyn and MLU measures obtained from a conversational language sample. In two separate analyses of morphological and syntactic abilities of adolescents and young adults with DS and those with FXS using narrative language samples, Keller-Bell and Abbeduto (2007) and Finestack and Abbeduto (2010) found that the participants with FXS produced proportionally more grammatical utterances than the participants with DS. Thus, it appears that individuals with FXS have stronger skills than those with DS in some aspects of the microstructure of language, especially at younger ages.

Only two studies have compared the macrostructural language abilities of individuals with DS and those with FXS and these studies yielded contrasting findings. In a study comparing the conversational skills of children and adolescents with DS to mental-age matched FXS boys, Roberts and colleagues (2007) found that boys with DS demonstrated significantly more instances of adequate topic maintenance than boys with FXS. In their examination of narrative language performance, Keller-Bell and Abbeduto (2007) found no significant DS and FXS group differences on most measures, including use of mental state verbs, character name, character dialogue, repetition, and fantasy/exaggeration; however, relative to the adolescents and young adults with FXS, the adolescents and young adults with DS had significantly more uses of onomatopoeia/exaggeration and a higher density and diversity of narration forms. In sum, based on this limited evidence, there is a tendency for individuals with DS to outperform individuals with FXS on macrostructural measures of language ability.

Current Study

Given the sparse and inconsistent findings regarding both the micro- and macrostructural language abilities of individuals with DS and those with FXS, it is clear that much remains to be understood about the language abilities of these populations. This is especially true of narrative language. For individuals with DS, current research indicates a need to focus intervention on microstructural forms and build upon macrostructural language strengths. However, the precise nature of these macrostructural strengths is not well understood. Although there is some indication of particular microstructural weaknesses for individuals with FXS, adolescents and young adults with FXS generally perform similarly to nonverbal mental-age matched children with TD. Based on these findings, it appears that intervention should broadly target all aspects of language in FXS. However, only one study to date has investigated narrative language abilities of individuals with FXS at the macrostructural level (i.e., Keller-Bell & Abbeduto, 2007). Moreover, that study examined very specific high-level narrative features (e.g., use of mental state verbs, repetition, fantasy) and did not consider the overall narrative structure. The current study was designed to evaluate the oral narratives of adolescents and young adults with DS and those with FXS using a more comprehensive measure of narrative structure; namely, the Narrative Scoring Scheme (NSS; Heilmann, Miller, Nockerts, et al., 2010).

NSS (Heilmann, Miller, Nockerts, et al., 2010) comprises the evaluation of seven narrative macrostructure components, including Introduction, Character Development, Mental States, Referencing, Conflict/Resolution, Cohesion, and Conclusion. Examination of these narrative elements is critical to determine if there are essential narrative elements that are frequently omitted or poorly constructed and that would negatively affect comprehension of the narrative being conveyed. Thus, we assessed the narratives of adolescents and young adults with DS, those with FXS, and younger TD children matched on nonverbal mental age or language ability using NSS procedures.

Study Questions and Predictions

This study aimed to increase our understanding of the narrative language abilities of verbally expressive adolescents and young adults with DS and those with FXS. Specifically, this study was designed to answer the following questions:

1. Are there differences in NSS performance between adolescents and young adults with DS who produce multi-word utterances and children with TD with similar nonverbal mental ages and/or MLUs?

2. Are there differences in NSS performance between adolescents and young adults with FXS who produce multi-word utterances and children with TD with similar nonverbal mental ages and/or MLUs?
3. Are there differences in NSS performance between adolescents and young adults with DS and adolescents and young adults with FXS with similar nonverbal mental ages and/or MLUs?

Three sets of analyses were used to answer these questions. The first set of analyses used nonparametric techniques with group matching based on nonverbal mental age. The second set of analyses included a subset of participants who were individually matched based on nonverbal mental age to control even more precisely for cognitive ability. The third set of analyses included a subset of participants who were individually matched based on MLU to control for morphosyntactic language ability. Based on previous findings, it was predicted that the DS group would significantly outperform the TD and FXS groups, with no significant group differences between the TD and FXS groups.

Method

Participants

The participants in this study included the same sample of individuals as in the Finestack and Abbeduto (2010) study of grammatical development. The characteristics of each study group are presented in Table 1. This sample comprised 24 adolescents and young adults with DS (mean age = 16.9 years), 12 adolescents and young adults with FXS (mean age = 14.95 years), and 21 younger children with typical cognitive development (TD; mean age = 4.82 years). All of the participants were involved in a larger study designed to examine the language development of individuals with DS or FXS. Results of other examinations, which include overlapping participant samples, have been previously reported (e.g., Abbeduto et al., 2003; Abbeduto et al., 2006; Keller-Bell & Abbeduto, 2007; Kover & Abbeduto, 2010; Lewis et al., 2006). The larger study included a pool of 236 individuals (77 DS; 55 FXS; 104 TD). The participants with TD were recruited locally through public postings and area pre-schools. 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, and mailings to special educators and genetic clinics.

To be included in the present study, participants had to complete each of the study measures, demonstrate 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; American Speech-Language-Hearing 1996 Audiologic Assessment Panel, 1997) 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, parents of the children with TD had to indicate that their children had no diagnosed disability and that they were not receiving special education services other than speech-articulation therapy. A total of 50 individuals (15 DS; 3 FXS; 32 TD) were excluded from the present analyses because they did not complete all study measures. After this exclusion, 5 males with FXS were excluded for meeting autism diagnostic criteria, and 8 individuals (4 DS; 2 FXS; 2 TD) were excluded for failing to meet other study inclusion criteria. In addition, three children reported to have typical development who received standard scores on the nonverbal Stanford-Binet (Thorndike, Hagen, & Sattler, 1986) composite below 80 were also excluded from the study. Moreover, only males with FXS were included in this study due to significant heterogeneity between genders (see Abbeduto, Brady, et al., 2007); thus, 10 females with FXS were excluded from the present analyses. Females were not excluded from the other diagnostic groups.

Because the focus of this study was on narrative language development, it was important that each narrative sample provide a sufficient corpus of utterances to analyze and that the utterances were at a level of complexity at which an analysis of narrative language would be appropriate. Thus, 102 individuals (34 DS; 22 FXS; 46 TD) were excluded because their narrative language sample did not include 50 complete and intelligible utterances and/or their mean length of utterance was less than 3.0. Additionally, one male with FXS was excluded because he had over 400 complete and intelligible C-units in his narrative sample, which was well over the group's mean of 70 C-units. The remaining sample, which was the focus of the current study, thus, comprised a highly selective subset of adolescents and young adults with DS or FXS who had relatively high levels of expressive language abilities.

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

To ensure similar characteristics across the DS, FXS, and TD groups on key characteristics and inclusion criteria, the chronological age, nonverbal mental age and IQ, and number of utterances and MLU based on the narrative sample were compared using nonparametric Kruskal-Wallis tests. Table 1 presents summative descriptions for each of these variables. The groups were well matched based on the number of utterances in their narratives and MLU, with analyses yielding p -values greater than .50 (Mervis & Robinson, 2003). Although not significantly different, the groups were not as well matched on nonverbal mental age, indicated by a nonsignificant p -value of 0.29, which is less than the .50 criterion level suggested by Mervis and Robinson (2003). Expected significant group differences were identified based on chronological age and nonverbal IQ, characterized by a significantly younger TD group with significantly higher nonverbal IQ standard scores relative to the DS and FXS groups. No significant differences were identified between the DS and FXS groups ($ps > .07$) on any of these latter measures. Chi-square analyses of group differences based on race and maternal education yielded no significant group differences. Analyses including these matched groups are referred to as *full-sample* analyses.

Because the p -value for nonverbal mental age was less than 0.50, a group of DS, FXS, and TD participants, individually matched on nonverbal mental age, was created. Table 2 contains the characteristics of these matches. Participants were matched within .33 years (mean difference for FXS/DS groups = 0.06, range = 0 – 0.14; FXS/TD groups = 0.15; range = 0 – 0.31). There was one participant with FXS, for whom no nonverbal mental age match was available. This participant was excluded. Thus, there were 11 matches with each comprising an individual with DS, an individual with FXS, and an individual with TD for a total of 33 individuals. Kruskal-Wallis tests were conducted to compare the matched groups on key variables. The tests revealed no significant group differences based on nonverbal mental age, $\chi^2(2, N = 33) = 0.38, p = 0.83$; total number of utterances, $\chi^2(2, N = 33) = 0.17, p = 0.92$; and MLU, $\chi^2(2, N = 33) = 2.53, p = 0.28$. Analyses involving this subset of participants are referred to as *nonverbal mental age-matched* analyses.

Similarly, a group of participants individually matched on MLU also was created. Table 3 contains the characteristics of these matches. Participants were matched within .50 morphemes (mean difference for FXS/DS groups = 0.15, range = 0.03 – 0.38; FXS/TD groups = 0.08, range = 0 – 0.32). Again, there was one participant with FXS for whom no MLU match was available. This participant was excluded, yielding 11 matches. Kruskal-

Wallis tests revealed no significant group differences based on nonverbal mental age, $\chi^2(2, N = 33) = 1.28, p = 0.53$; total number of utterances, $\chi^2(2, N = 33) = 3.00, p = 0.22$; and MLU, $\chi^2(2, N = 33) = 0.04, p = 0.98$. Analyses involving this subset of participants are referred to as *MLU-matched* analyses.

Procedures

Following procedures approved by an Institutional Review Board of the University of Wisconsin-Madison, a parent of each of the participants consented to their child's participation. The study testing was in most cases completed across two test sessions that occurred in a single day with a 1- to 2-hour break between sessions. For some participants, the sessions were spread across two days. In these cases, all testing was completed in no more than a 3-week time period. A quiet room was used to test participants individually, with parents having the option to view testing through an observation window. For each participant, the same examiner typically administered the complete test battery.

Study Measures

Nonverbal Intelligence—Nonverbal cognitive ability was assessed using the Bead Memory, Pattern Analysis, and Copying subtests of the Stanford-Binet Intelligence Scale, 4th Edition (Thorndike, et al., 1986). These subtests require few verbal instructions and examinee responses are all nonverbal. A nonverbal partial composite IQ score was calculated using the mean standard score from each of the subtests. The mean of the age equivalents obtained from the three subtests yielded the nonverbal mental age for each participant (Abbeduto, et al., 2003; Abbeduto et al., 2008; Chapman, et al., 1998).

Number of Utterances/Mean Length of Utterance—The number of complete and intelligible utterances and MLU in morphemes was derived from a narrative language sample elicited from participants using the wordless picture book *Frog Goes to Dinner* (Mayer, 1974). After reviewing the book once, the participants retold the story, page by page, in their own words to an examiner who was seemingly unfamiliar with the storyline. The examiner provided minimal prompts throughout the story-telling. Each narrative sample was audio-taped and transcribed by trained research assistants. Following the standard Systematic Analysis of Language Transcripts conventions (SALT; Miller & Chapman, 2000), utterances were segmented into communication units (C-units), defined as an independent clause and its modifiers, which can include dependent clauses (Loban, 1976). Sentence fragments and elliptical utterances were also transcribed and counted as separate C-units. Each narrative was transcribed by a primary coder and checked by a secondary coder who, while viewing the primary coder's transcript, 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. SALT software was used to compute the number of complete and intelligible C-units as well as the MLU in morphemes for C-units.

An independent coder randomly selected and transcribed eight (14%) of the narrative transcripts for reliability purposes. These transcripts included three from the DS participants, three from the FXS participants, and two from the TD participants. The independent coder's transcripts were compared to the primary coders' original transcripts. The mean percent of point-to-point agreement for segmentation into C-units was 86% (range = 78% – 94%) and for number of bound morphemes per utterance was 100% (range = 98% – 100%).

Narrative Scoring Scheme—NSS (Heilmann, Miller, Nockerts, et al., 2010) comprises the evaluation of seven narrative macrostructure components, including Introduction, Character Development, Mental States, Referencing, Conflict/Resolution, Cohesion, and

Conclusion. Individual scores from each of these components are combined to yield a total NSS score, with each category having equal weighting. The NSS component scores as well as the total NSS score were used in the analyses for the present study.

Appendix A contains sample rubrics used to assign scores for the macrostructure components. The rubrics assisted the coders in consistently assigning appropriate scores for each story component. Although these rubrics were modeled from the NSS rubrics available on the SALT website (<http://www.saltsoftware.com/training/handcoded/nss.cfm>), the rubrics used in this study were modified so as to be appropriate for *Frog Goes to Dinner*. Scores of 0 through 5 were given for each narrative component, with a score of 0 indicating poor performance and a score of 5 indicating proficient use. The scored components reflected participants' ability to encode the following in their productions: (a) Introduction - incorporate new main and sub-settings (e.g., house, restaurant, frog in wine glass) as well as main and sub-characters (e.g., frog, boy, dog); (b) Character Development - mention and provide details regarding main and sub-characters; (c) Mental States – use a variety of mental state words to convey characters' emotions and thought processes; (d) Referencing – consistently and accurately use pronouns and their antecedents; (e) Conflict/Resolution – clearly describe key conflicts and their corresponding resolutions for plot development; (f) Cohesion – logically sequence story events and provide sufficient transitions between events; and (g) Conclusion – provide a description of the final story event and wrap-up the entire story.

Each narrative transcript was independently scored by two trained coders (the first and second authors), who were blind to participant group assignment. The same two coders scored all of the transcripts. The coders compared their scores and noted discrepancies. The coders then met and resolved all scoring differences. A third coder was trained and independently scored 20% ($n = 13$) of the transcripts. NSS coding reliability was calculated using Krippendorff's alpha, which takes into account chance agreement and the degree of difference between coders (Hayes & Krippendorff, 2007; Krippendorff, 2004). Krippendorff's alpha has been used in other studies using NSS measures (e.g., Heilmann, Miller, & Nockerts, 2010; Heilmann, Miller, Nockerts, et al., 2010). Krippendorff's alpha should be interpreted such that comparisons greater than .67 are acceptable for tentative conclusions and that values greater than .80 indicate adequate agreement. Statistical analyses yielded the following alpha values for the NSS measures: Introduction $\alpha = .86$; Character Development $\alpha = .98$; Mental States $\alpha = .97$; Referencing $\alpha = .53$; Conflict/Resolution $\alpha = .97$; Cohesion $\alpha = .67$; and Conclusion $\alpha = .72$. Because of the low reliability of the Reference measure, it was omitted from all subsequent analyses and did not contribute to the Total NSS measure. Krippendorff's alpha for the Total NSS measure (excluding the Reference measure) was $\alpha = .96$. All other agreement levels were considered acceptable.

Statistical Analyses

This study involved three sets of nonparametric Kruskal-Wallis analyses. The first set of analyses compared the entire sample of DS, FXS, and TD study participants. The second set of analyses included a sample of 33 participants (11 per diagnostic group) individually matched based on nonverbal mental age. The third set of analyses included a sample of 33 participants (11 from each diagnostic group) individually matched based on MLU. In each of these sets of analyses, a separate Kruskal-Wallis test was conducted for each of the seven NSS measures (i.e., Introduction, Character Development, Mental States, Conflict/Resolution, Cohesion, Conclusion, and Total NSS Score) to compare the groups. Significant Kruskal-Wallis tests were followed by Mann-Whitney U tests with alpha set at .05. Effect sizes (d) were calculated and interpreted using Cohen's (1988) standards of .20 to represent a small effect size, .50 a medium effect size, and .80 a large effect size. It is important to

note that due to the relatively small sample sizes no formal adjustments were made to control for Type I error. Thus, all results should be viewed as preliminary.

Results

Full-Sample Analyses

Seven Kruskal-Wallis tests were completed to examine expressive narrative language ability based on NSS scores. The means, standard deviations, and effect sizes for each analysis are presented in Table 4. Results indicated significant group differences for Introduction, $\chi^2(2, N = 57) = 9.70, p < 0.01$, and NSS Total Score, $\chi^2(2, N = 57) = 5.97, p = 0.05$. No statistically significant differences emerged for Character, $\chi^2(2, N = 57) = 4.40, p = .11$, Mental States, $\chi^2(2, N = 57) = 1.04, p = 0.60$, Conflict/Resolution, $\chi^2(2, N = 57) = 3.00, p = 0.22$, Cohesion, $\chi^2(2, N = 57) = 5.21, p = 0.07$, or Conclusion, $\chi^2(2, N = 57) = 5.67, p = 0.06$. Post-hoc Mann-Whitney *U* tests for Introduction revealed significant differences between the DS and TD groups ($p = .03$) and between the FXS and TD groups ($p < .01$), characterized by the DS and FXS groups outperforming the TD group. Post-hoc analyses for NSS Total Score revealed a significant difference between the DS and TD groups ($p = .02$), with the DS group outperforming the TD group. With exception of the Mental State measure, the effect sizes for the DS and TD comparisons and the FXS and TD comparisons were all medium-sized (d range = .47 – 1.32). Thus, despite the lack of statistically significant group differences, there was a strong trend for the DS and FXS groups to outperform the TD group.

Nonverbal Mental Age-Matched Analyses

Expressive narrative language ability of adolescents and young adults with DS, adolescents and young adults with FXS, and children with TD who were individually matched on mental age was evaluated using seven NSS measures. A separate Kruskal-Wallis test was conducted for each variable. Figure 1 displays boxplots for the DS, FXS, and TD groups for each NSS variable. Significant group differences were found for Introduction, $\chi^2(2, N = 33) = 6.09, p = 0.05$, Conflict/Resolution, $\chi^2(2, N = 33) = 6.86, p = 0.03$, Cohesion, $\chi^2(2, N = 33) = 7.07, p = 0.03$, and Total Score, $\chi^2(2, N = 33) = 8.22, p = 0.02$. The follow-up Mann-Whitney *U* tests for these significant analyses revealed that the DS group significantly outperformed the TD group (all $ps < .05$) on all measures except Introduction ($p = .14$) and that the FXS group significantly outperformed the TD group on the Introduction and Total NSS measures ($ps < .05$). Significant group differences were not found between the DS and FXS groups on any measure.

MLU-Matched Analyses

Similar to the evaluation of groups individually matched on nonverbal mental age, analyses were conducted to evaluate narrative language ability of adolescents and young adults with DS, adolescents and young adults with FXS, and children with TD, individually matched based on mean length of C-unit. A separate Kruskal-Wallis test was conducted for each of the NSS measures. A significant group difference was found for the Introduction, $\chi^2(2, N = 33) = 8.03, p = 0.02$. The follow-up Mann-Whitney *U* test revealed that the FXS group significantly outperformed the TD group ($p < .01$). There were no significant differences between the DS and TD groups or the DS and FXS groups.

Discussion

The aim of this study was to gain a better understanding of the narrative language abilities of verbally expressive adolescents and young adults with DS or FXS. The narrative performance of adolescents and young adults with DS and those with FXS were compared

to younger TD children with similar nonverbal mental ages and/or language abilities. Narrative language was assessed at the macrostructure level using the Narrative Scoring Scheme (NSS; Heilmann, Miller, Nockerts, et al., 2010). Overall, the analyses revealed some relative strengths in macrostructural narrative language for adolescents and young adults with DS and those with FXS. Across all analyses, the DS or FXS groups outperformed the younger TD group on the Introduction measure. The DS and FXS groups also outperformed the TD group on the Total NSS measure based on two of the analyses. The most group differences emerged when the participants were individually matched on nonverbal mental age and the fewest emerged when the participants were individually matched on MLU. Thus, it appears that macrostructural language ability in these populations is more closely associated with microstructural language ability than nonverbal cognitive ability.

DS Narrative Macrostructure Language

Based on findings from previous examinations of the narrative language abilities of individuals with DS that included participants similar to our own (e.g., Boudreau & Chapman, 2000; Miles & Chapman, 2002), it was predicted that the DS group would significantly outperform the matched TD group. That prediction was partially supported in this study. In the full sample analysis, the adolescents and young adults with DS obtained significantly higher scores on the Introduction and Total NSS measures with the group comparisons yielding medium effect sizes for six of the seven NSS measures. Additionally, in the individually-matched nonverbal mental age comparison, the DS group received significantly higher scores on the Conflict/Resolution, Cohesion, and Total NSS measures. In the individually-matched MLU comparison there were no significant DS and TD group differences on any of the NSS measures, although visual image of the box charts presented in Figure 1 reveal a trend for the DS participants to outperform the TD participants on each measure. These results suggest that without accounting for cognitive and language abilities, elements of narrative macrostructure are areas of strength for many adolescents and adults with Down syndrome and support our prediction. However, when grossly controlling for language ability, these strengths no longer emerge, which suggests that there is a close association between micro- and macro-structural narrative language performance and that the macrostructural strengths of the individuals with DS is driven by their microstructural language skills.

The findings of the current study and those of the Keller-Bell and Abbeduto (2007) study, which used the same narratives from an overlapping participant sample, yielded similar performance patterns. Keller-Bell and Abbeduto analyzed the narrative samples using a high-point analysis, which evaluates specific narrative skills, such the use of mental state verbs, character names, dialogue, repetition, and exaggeration. Keller-Bell and Abbeduto identified significant differences between the participants with DS and the TD children of similar nonverbal mental age on the onomatopoeia/exclamation measure, but no differences measures examining use of mental state verbs, character names, character dialogue, repetition, or fantasy/exaggeration. Similarly, in the current study, the results of the analyses in which nonverbal mental age was controlled, the DS group outperformed the TD group on some (i.e., Conflict/Resolution, Cohesion, Total), but not all (i.e., Introduction, Character Development, Mental States, Conclusion) NSS measures. Findings from these two studies suggest that when matched on nonverbal cognitive abilities, the relative strengths of individuals with DS may be limited to certain aspects of narration; however these strengths diminish when microstructural language abilities are considered. Thus, it is important that language interventions for individuals with DS focus on both microstructural and macrostructural narrative language abilities.

Some of the present results, however, run counter to findings from previous investigations. In the current study, for example, the MLU-matched analysis did not yield a significant difference between the DS and TD groups based on the NSS Conflict/Resolution measure, which is designed to evaluate the inclusion of elements essential for advancing the story plot. In contrast, in an investigation involving adolescents and young adults (aged 12 through 26 years) with DS that included several measures of narrative content which were similar to the NSS Conflict/Resolution measure, Miles and Chapman (2002) found that individuals with DS demonstrated significant strengths in plot and theme performance compared to younger TD children with similar MLUs. Differences in participant groups, narrative elicitation procedures, and measures are all possible reasons for this discrepancy. For example, although both the current study and the Miles and Chapman study included participants with DS and those with TD of similar ages, the MLUs of the participants in the current study were considerably higher (DS: 6.53 vs. 4.23; TD: 6.45 vs. 4.29). The higher MLUs in the current study are a good indication that our participants had better overall language abilities resulting in stronger performance in thematic context. Thus, thematic context may be a less sensitive measure of narrative ability for individuals with more advanced language abilities. In contrast to the current study, participants in the Miles and Chapman study did not have the opportunity to view each page in the wordless picture book before telling this story. Thus, the differences in thematic content found in the Miles and Chapman study may have reflected differences in online processing and memory, which the current study was not designed to examine. It is also important to note that the current study and the Miles and Chapman study used different measures of thematic content and these measures may have captured slightly different skills.

FXS Narrative Macrostructure Language

Very few studies have examined the narrative language abilities of individuals with FXS at the macrostructure level. In general, the few studies that have been conducted have not revealed differences relative to younger TD children with similar nonverbal mental ages. Thus, we predicted that the adolescents and young adults with FXS in our study would not perform differently than younger TD children matched on nonverbal mental age or language ability. However, our study results collectively indicate a relative strength for individuals with FXS to provide introductory details. In each of the study comparisons, the FXS participants outperformed the TD participants based on the NSS Introduction measure. Additionally, in the comparison involving nonverbal mental-age-matched TD children, the FXS group's Total scores were significantly higher than the TD group's scores. Narrative samples from a set of MLU-matched participants (MLU7) are presented in Appendix B. Examination of the first few lines of the FXS sample and the TD sample clearly demonstrates how the FXS participant purposefully sets up the story context, while the TD participant immediately jumps into the story providing vague initial setting and character details. These samples in conjunction with the study results suggest that adolescents and young adults with FXS have strengths in some aspects at the narrative macrostructural level relative to expectations based on their nonverbal mental ages and MLUs.

With this narrative profile of individuals with FXS in mind, it is important for language interventionists to not limit the narrative goals of adolescents and young adults to standards based on nonverbal mental age and MLU. Results from the current study support a relative strength of the inclusion of introduction components in the narratives of individuals with FXS and indicate that individuals with FXS are capable of exceeding putative "constraints." In many cases, it may be appropriate for interventions to target more complex narrative aspects, such as character and plot development. This narrative strength may be used to build up other, less advanced narrative areas. For example, clinicians may target character development by teaching individuals with FXS to include more details of characters when

first introduced in the story. The narrative context may serve as an exceptionally good context in which to target specific language weaknesses such as grammatical complexity.

DS and FXS Narrative Macrostructure Language Comparison

Based on findings from Roberts and colleagues (2007) and Keller-Bell and Abbeduto (2007), we predicted that the adolescents and young adults with DS would outperform the adolescents and young adults with FXS on the NSS measures; however, this was not the case. No significant differences emerged between the DS and FXS groups when matched on nonverbal mental age or microstructural language ability. Thus, the NSS measures analyzed in this study were not sensitive to group differences, if they exist.

Appendix B comprises the narrative samples from a set of individually MLU-matched participants (MLU7). The DS and FXS participants received the same Total NSS score (19), although there are small differences in the scores awarded for individual NSS components. The TD participant received a lower NSS Total score (14). Descriptively, in this sample, the scoring differences were largely due to the TD receiving lower scores on the Character Development, Conflict/Resolution, and Cohesion measures in comparison to the DS and FXS participants. Review of the narrative samples and results from this study suggest that verbally expressive adolescents and young adults with DS and those with FXS have similar narrative language profiles when considered at the macrostructure level. This similarity is further supported by the small effect sizes (d range = .05 to .38) based on the full-sample analyses (see Table 4).

Clinical Implications and Conclusions

This is the first study to examine the macrostructural narrative language abilities of adolescents and young adults with DS and adolescents and young adults with FXS using NSS. NSS is a holistic rating that proved to be sensitive to language differences between the groups with DS or FXS and the TD group on some measures (i.e., Introduction, NSS Total). Thus, NSS may serve as a good tool to use in treatment planning for individuals with DS and those with FXS. However, because of its holistic nature, NSS may be insensitive to some areas of deficit in DS and FXS; therefore, it may be necessary to supplement NSS with measures that analyze narrative language in finer detail when planning a comprehensive language intervention for an individual. Thus, until more is learned about the macrostructural language abilities of individuals with DS and those with FXS, it is important for clinicians to use a battery of macrostructural as well as microstructural narrative measures in treatment planning.

This study included a relatively small sample of adolescents and young adults with DS and male adolescents and young adults with FXS. Therefore, study results should be viewed as preliminary. The individuals with DS and those with FXS included in the study were highly specified subtypes that were verbally expressive with MLUs greater than 3.0. Because the specific speech-language services that these individuals were receiving is unknown, clinicians should be careful not to generalize these findings to all individuals with DS or FXS. However, if working with an adolescent or young adult with DS or FXS who is constructing complete utterances and exhibits some story-telling skills, the results of this study may help clinicians design appropriate assessment and treatment approaches.

Although the study results reveal that some aspects of narrative language may be relative strengths for adolescents and young adults with DS and those with FXS, it is clear that most verbally expressive individuals within these groups have not fully mastered narrative language. Figure 1 clearly depicts that these individuals are not performing at ceiling; rather, their narrative macrostructural language skills (e.g., Character Development, Cohesion)

appear to be still developing. Although we do not know if the participants in this study had previously received treatment focused on microstructural and/or macrostructural narrative language components, it is important for clinicians to understand the individual narrative language profiles of their clients with DS or FXS and to target skills that are relatively strong but still developing, such as those underlying the expression of the Introduction and Conflict/Resolution components, as well as those skills that are yet or just beginning to emerge, such as those underlying expression of the Character and Cohesion components. Moreover, narrative strengths may be exploited by embedding microstructure targets in narrative contexts.

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References

- Abbeduto L, Brady N, Kover ST. Language development and fragile X syndrome: Profiles, syndrome-specificity, and within-syndrome differences. *Mental Retardation and Developmental Disabilities Research Reviews*. 2007; 13(1):36–46. [PubMed: 17326110]
- Abbeduto L, Murphy MM, Cawthon SW, Richmond EK, Weissman MD, Karadottir S, O'Brien A. Receptive language skills of adolescents and young adults with Down syndrome or fragile X syndrome. *American Journal on Mental Retardation*. 2003; 108(3):149–160. [PubMed: 12691594]
- Abbeduto L, Murphy MM, Kover ST, Giles ND, Karadottir S, Amman A, Nollin KA. Signaling noncomprehension of language: A comparison of fragile X syndrome and Down syndrome. *American Journal on Mental Retardation*. 2008; 113(3):214–230. [PubMed: 18407723]
- Abbeduto L, Murphy MM, Richmond EK, Amman A, Beth P, Weissman MD, Karadottir S. Collaboration in referential communication: Comparison of youth with Down syndrome or fragile X syndrome. *American Journal on Mental Retardation*. 2006; 111(3):170–183. [PubMed: 16597184]
- Abbeduto L, Warren SF, Conners FA. Language development in Down syndrome: from the prelinguistic period to the acquisition of literacy. *Mental Retardation and Developmental Disabilities Research Reviews*. 2007; 13(3):247–261.10.1002/mrdd.20158 [PubMed: 17910087]
- American Speech-Language-Hearing 1996 Audiologic Assessment Panel. *Guidelines for Audiologic Screening*. Rockville, MD: 1997.
- Bailey DB Jr, Raspa M, Holiday D, Bishop E, Olmsted M. Functional skills of individuals with fragile X syndrome: A lifespan cross-sectional analysis. *American Journal on Intellectual and Developmental Disabilities*. 2009; 114(4):289–303. [PubMed: 19642710]
- Berglund E, Eriksson M, Johansson I. Parental reports of spoken language skills in children with Down syndrome. *Journal of speech, language, and hearing research*. 2001; 44(1):179–191.
- Bird EKR, Cleave PL, White D, Pike H, Helmkay A. Written and oral narratives of children and adolescents with Down syndrome. *Journal of Speech, Language, and Hearing Research*. 2008; 51(2):436–450.
- Boudreau DM, Chapman RS. The relationship between event representation and linguistic skill in narratives of children and adolescents with Down syndrome. *Journal of Speech, Language, and Hearing Research*. 2000; 43(5):1146–1159.
- Chapman RS, Seung HK, Schwartz SE, Bird EKR. Language skills of children and adolescents with Down syndrome: II Production deficits. *Journal of Speech, Language, and Hearing Research*. 1998; 41(4):861–873.
- Cohen, J. *Statistical power analysis for the behavioral sciences*. Hillsdale, NJ: Erlbaum; 1988.
- Crawford DC, Acuna JM, Sherman SL. FMR1 and the fragile x syndrome: Human genome epidemiology review. *Genetics in Medicine*. 2001; 3:359–371. [PubMed: 11545690]

- Eadie PA, Fey ME, Douglas JM, Parsons CL. Profiles of grammatical morphology and sentence imitation in children with specific language impairment and Down syndrome. *Journal of Speech, Language, and Hearing Research*. 2002; 45(4):720–732.
- Finestack LH, Abbeduto L. Expressive language profiles of verbally expressive adolescents and young adults with Down syndrome or fragile X syndrome. *Journal of Speech, Language, and Hearing Research*. 2010; 53:1334–1348.10.1044/1092-4388(2010/09-0125)
- Hagerman, RJ. Etiology, diagnosis, and development in Fragile X syndrome. In: Roberts, J.; Chapman, R.; Warren, SF., editors. *Speech and Language Development and Intervention in Down Syndrome and Fragile X Syndrome*. Baltimore: Paul H. Brookes Publishing Co; 2008. p. 27-49.
- Hall SS, Burns DD, Lightbody AA, Reiss AL. Longitudinal changes in intellectual development in children with fragile X syndrome. *Journal of Abnormal Child Psychology: An official publication of the International Society for Research in Child and Adolescent Psychopathology*. 2008; 36(6): 927–939.
- Hayes AF, Krippendorff K. Answering the call for a standard reliability measure for collecting data. *Communication Methods and Measures*. 2007; 1:77–89.
- Heilmann J, Miller JF, Nockerts A. Sensitivity of narrative organization measures using narrative retells produced by young school-age children. *Language Testing*. 2010; 27(4):603–626.10.1177/0265532209355669
- Heilmann J, Miller JF, Nockerts A, Dunaway C. Properties of the Narrative Scoring Scheme Using Narrative Retells in Young School-Age Children. *American Journal of Speech Language Pathology*. 2010; 19(2):154–166. [PubMed: 20008470]
- Improved national prevalence estimates for 18 selected major birth defects-United States 1999–2001. *Morbidity and Mortality Weekly Report*. 2006; 54(51&52):1301–1305. [PubMed: 16397457]
- Joffe V, Varlokosta S. Patterns of syntactic development in children with Williams syndrome and Down's syndrome: Evidence from passives and wh-questions. *Clinical Linguistics & Phonetics*. 2007; 21(9):705–727. [PubMed: 17701757]
- Keller-Bell YD, Abbeduto L. Narrative development in adolescents and young adults with fragile X syndrome. *American Journal on Mental Retardation*. 2007; 112(4):289–299. [PubMed: 17559295]
- Kintsch W, van Dijk TA. Toward a model of text comprehension and production. *Psychological Review*. 1978; 85(5):363–394.
- Kover ST, Abbeduto L. Expressive language in male adolescents with fragile X syndrome with and without comorbid autism. *Journal of Intellectual Disability Research*. 2010; 54(3):246–265.10.1111/j.1365-2788.2010.01255.x [PubMed: 20146742]
- Krippendorff K. Reliability in Content Analysis. *Human Communication Research*. 2004; 30(3):411–433.10.1111/j.1468-2958.2004.tb00738.x
- Lewis P, Abbeduto L, Murphy M, Richmond E, Giles N, Bruno L, Schroeder S. Cognitive, language and social-cognitive skills of individuals with Fragile X Syndrome with and without autism. *Journal of Intellectual Disability Research*. 2006; 50(7):532–545. [PubMed: 16774638]
- Loban, W. *Language development: Kindergarten through grade twelve*. Urbana, IL: National Council of Teachers of English; 1976.
- Martin GE, Klusek J, Estigarribia B, Roberts JE. Language characteristics of individuals with Down syndrome. *Topics in Language Disorders*. 2009; 29(2):112–132. [PubMed: 20428477]
- Mayer, M. *Frog Goes to Dinner*. NY: Dial Books; 1974.
- Mervis, CB.; Robinson, BF. Methodological issues in cross-group comparisons of language and cognitive development. In: Levy, Y.; Schaeffer, J., editors. *Language Competence Across Populations: Toward a Definition of Specific Language Impairment*. Mahwah, NJ: Lawrence Erlbaum Associates; 2003.
- Miles S, Chapman RS. Narrative content as described by individuals with Down syndrome and typically developing children. *Journal of Speech, Language, and Hearing Research*. 2002; 45(1): 175–189.
- Miller, JF.; Chapman, R. *SALT: Systematic Analysis of Language Transcripts [Computer software]*. Language Analysis Laboratory, Waisman Center, University of Wisconsin-Madison; 2000.

- Philofsky A, Hepburn SL, Hayes A, Hagerman R, Rogers SJ. Linguistic and cognitive functioning and autism symptoms in young children with Fragile X syndrome. *American Journal on Mental Retardation*. 2004; 109(3):208–218. [PubMed: 15072521]
- Price J, Roberts J, Hennon EA, Berni MC, Anderson KL, Sideris J. Syntactic complexity during conversation of boys with fragile X syndrome and Down syndrome. *Journal of Speech, Language, and Hearing Research*. 2008; 51(1):3–15.
- Prouty LA, Rogers RC, Stevenson RE, Dean JH, Palmer KK, Simensen RJ, Schwartz CE. Fragile X syndrome: growth, development, and intellectual function. *American Journal of Medical Genetics*. 1988; 30(1–2):123–142. [PubMed: 3177438]
- Roberts J, Hennon EA, Price JR, Dear E, Anderson K, Vandergrift NA. Expressive language during conversational speech in boys with Fragile X syndrome. *American Journal on Mental Retardation*. 2007; 112(1):1–17. [PubMed: 17181388]
- Roberts J, Martin GE, Moskowitz L, Harris AA, Foreman J, Nelson L. Discourse skills of boys with fragile X syndrome in comparison to boys with Down syndrome. *Journal of Speech, Language, and Hearing Research*. 2007; 50(2):475–492.
- Roberts J, Mirrett P, Burchinal M. Receptive and expressive communication development of young males with fragile X syndrome. *American Journal on Mental Retardation*. 2001; 106(3):216–230. [PubMed: 11389664]
- Roberts JE, Mankowski JB, Sideris J, Goldman BD, Hatton DD, Mirrett PL, Bailey DB Jr. Trajectories and predictors of the development of very young boys with Fragile X Syndrome. *Journal of Pediatric Psychology*. 2009; 34(8):827–836. [PubMed: 19074489]
- Rosin MM, Swift E, Bless D, Kluppel Vetter D. Communication profiles of adolescents with Down syndrome. *Journal of Childhood Communication Disorders*. 1988; 12(1):49–64.
- Scarborough HS. Index of productive syntax. *Applied Psycholinguistics*. 1990; 11:1–22.
- Simon JA, Keenan JM, Pennington BF, Taylor AK, Hagerman RJ. Discourse processing in women with fragile X syndrome: Evidence for a deficit establishing coherence. *Cognitive Neuropsychology*. 2001; 18(1):1–18. [PubMed: 20945204]
- Thorndike, RL.; Hagen, EP.; Sattler, JM. *Stanford-Binet Intelligence Scale*. 4. Chicago: Riverside; 1986.

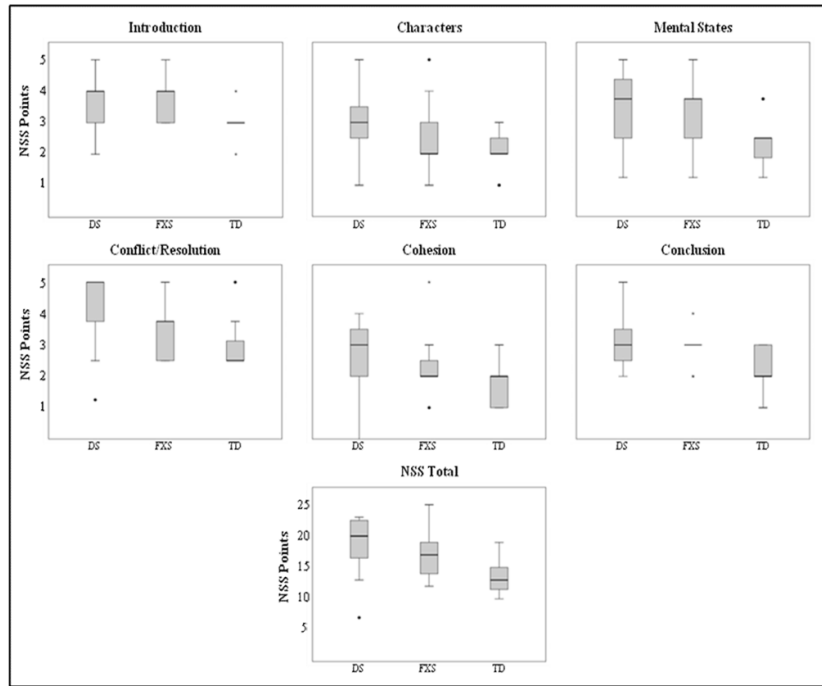


Figure 1. Box plots of the Nonverbal Mental Age-matched DS, FXS, and TD groups for the NSS measures

Table 1

Participant Characteristics for the Full-Sample DS, FXS, and TD Groups

Characteristic	Group			p	d
	DS (n = 24)	FXS (n = 12)	TD (n = 21)		
Chronological Age (years)				< 0.001	DS/FXS: 0.68 DS/TD: 5.29 FXS/TD: 5.31
Mean	16.90	14.95	4.82		
SD	3.14	2.59	0.76		
Min-Max	12.08–23.37	11.38–19.74	3.61–6.66		
Nonverbal Mental Age (years) ^a				0.29	DS/FXS: 0.54 DS/TD: 0.06 FXS/TD: -0.44
Mean	4.94	4.38	4.87		
SD	1.04	1.03	1.21		
Min-Max	3.31–7.06	2.86–7.11	3.42–7.84		
Nonverbal IQ ^a				< 0.001	DS/FXS: 0.34 DS/TD: -6.81 FXS/TD: -5.37
Mean	41.71	39.50	98.00		
SD	6.87	6.05	9.45		
Min-Max	36–57	36–56	84–115		
Number of C-units ^b				0.68	DS/FXS: 0.12 DS/TD: -0.23 FXS/TD: -0.34
Mean	67.42	65.67	71.43		
SD	14.44	13.83	19.58		
Min-Max	50–96	51–95	50–121		
Mean Length of Utterance (morphemes) ^b				0.51	DS/FXS: 0.36 DS/TD: 0.05 FXS/TD: -0.44
Mean	6.53	5.97	6.45		
SD	1.95	1.01	1.15		
Min-Max	3.40–11.20	4.43–8.10	3.86–8.06		
Race				0.08	Cramer's V = 0.30
White:Other	24:0	11:1	17:4		

Characteristic	Group			<i>p</i>	<i>d</i>
	DS (n = 24)	FXS (n = 12)	TD (n = 21)		
Maternal Education ^c				0.10	<i>Cramer's V</i> = 0.28
High School or Less: Some College or More	10:14	5:7	3:18		
Gender				NA	NA
Female:Male	11:13	0:12	13:8		

^aMean 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);

^bBased on a narrative language sample;

^cPaternal education level used for one participant in the DS group because maternal education level was unknown

Table 2
Participant Characteristics for the Nonverbal Mental Age-Matched DS, FXS, and TD Groups

ID	DS						FXS						TD					
	Gender	NMA	CA	Uttr	MLU	MLU	Gender	NMA	CA	Uttr	MLU	MLU	Gender	NMA	CA	Uttr	MLU	
MA 1	M	3.53	12.08	55	5.09	MA 1	M	3.56	14.41	64	2.69	MA 1	M	3.42	4.17	80	3.24	
MA 2	M	3.72	15.99	50	4.59	MA 2	M	3.83	13.35	58	5.00	MA 2	M	3.72	4.15	62	4.89	
MA 3	M	3.89	18.24	92	2.71	MA 3	M	3.92	12.18	73	3.37	MA 3	F	3.72	4.52	121	6.96	
MA 4	F	4.17	18.53	56	2.41	MA 4	M	4.03	12.49	51	2.42	MA 4	F	4.11	4.37	50	3.50	
MA 5	M	4.19	21.84	65	2.63	MA 5	M	4.25	16.85	69	2.59	MA 5	F	4.14	4.28	53	2.65	
MA 6	M	4.39	18.66	50	2.47	MA 6	M	4.33	11.38	54	3.79	MA 6	M	4.19	5.19	60	2.82	
MA 7	F	4.47	14.98	55	2.84	MA 7	M	4.39	19.74	52	2.47	MA 7	M	4.19	4.40	53	3.33	
MA 8	F	4.56	18.39	96	2.65	MA 8	M	4.56	14.55	55	4.00	MA 8	F	4.25	4.77	83	4.27	
MA 9	M	4.72	16.77	58	4.03	MA 9	M	4.81	13.39	95	2.41	MA 9	F	4.81	4.84	103	5.03	
MA 10	M	4.89	13.87	79	3.56	MA 10	M	4.94	15.83	76	2.59	MA 10	F	5.00	4.89	63	3.56	
MA 11	F	7.06	23.37	70	3.06	MA 11	M	7.11	17.67	58	2.88	MA 11	F	7.42	6.21	54	2.32	
M		4.51	17.52	66	7.06	M		4.52	14.71	64.09	5.89	M		4.45	4.70	71.09	6.41	
SD		0.94	3.30	16.37	2.35	SD		0.95	2.57	13.33	1.02	SD		1.08	0.60	23.22	0.99	

Table 3

Participant Characteristics for the MLU-matched DS, FXS, and TD Groups

ID	DS				FXS				TD								
	Gender	NMA	CA	Uttr	MLU	Gender	NMA	CA	Uttr	MLU	Gender	NMA	CA	Uttr	MLU		
MLU 1	F	4.86	14.73	55	4.80	MLU 1	M	4.39	19.74	52	4.69	MLU 1	M	5.20	3.73	81	5.01
MLU 2	M	4.89	13.87	79	5.09	MLU 2	M	3.56	14.41	64	5.19	MLU 2	F	3.72	4.52	121	5.19
MLU 3	M	3.31	13.21	64	5.28	MLU 3	M	3.92	12.18	73	5.47	MLU 3	M	3.72	4.15	62	5.53
MLU 4	F	6.25	19.58	72	5.68	MLU 4	M	4.94	15.83	76	5.64	MLU 4	F	4.25	4.77	83	5.63
MLU 5	F	3.36	19.45	70	5.69	MLU 5	M	4.81	13.39	95	5.83	MLU 5	M	5.94	5.76	87	5.85
MLU 6	F	4.56	18.39	96	6.17	MLU 6	M	4.33	11.38	54	6.11	MLU 6	M	3.42	4.17	80	6.15
MLU 7	M	4.72	16.77	58	6.19	MLU 7	M	7.11	17.67	58	6.16	MLU 7	M	4.19	5.19	60	6.17
MLU 8	M	5.69	14.79	64	6.20	MLU 8	M	4.56	14.55	55	6.51	MLU 8	M	7.84	6.66	95	6.34
MLU 9	M	6.46	15.18	91	6.47	MLU 9	M	3.83	13.35	58	6.69	MLU 9	F	5.36	4.98	81	6.88
MLU 10	F	5.39	12.49	75	6.48	MLU 10	M	2.86	17.61	83	6.86	MLU 10	F	4.11	4.37	50	6.90
MLU 11	F	4.17	18.53	56	8.20	MLU 11	M	4.25	16.85	69	8.10	MLU 11	M	5.39	4.84	70	8.06
<i>M</i>		4.88	16.09	70.91	6.02	<i>M</i>		4.41	15.18	67.00	6.11	<i>M</i>		4.83	4.83	79.09	6.16
<i>SD</i>		1.03	2.57	13.62	0.92	<i>SD</i>		1.07	2.59	13.68	0.93	<i>SD</i>		1.29	0.82	19.19	0.88

Table 4
Ms, SDs, Kruskal-Wallis p-values, and Effect Sizes for Full-sample Comparisons

NSS Measure	Group			p	d
	DS (n = 24)	FXS (n = 12)	TD (n = 21)		
Introduction				<.01*	DS/FXS: -0.38
Mean	3.46	3.75	2.95		DS/TD: 0.68*
SD	0.88	0.62	0.59		FXS/TD: 1.32*
Character				.11	DS/FXS: 0.15
Mean	2.92	2.75	2.24		DS/TD: 0.66
SD	1.14	1.14	0.89		FXS/TD: 0.50
Mental State				.60	DS/FXS: -0.13
Mean	2.46	2.58	2.24		DS/TD: 0.22
SD	1.06	0.79	0.94		FXS/TD: 0.39
Conflict/Resolution				.22	DS/FXS: 0.17
Mean	2.50	2.33	1.90		DS/TD: 0.63
SD	1.06	0.99	0.83		FXS/TD: 0.47
Cohesion				.07	DS/FXS: 0.17
Mean	2.50	2.33	1.90		DS/TD: 0.63
SD	1.06	0.99	0.83		FXS/TD: 0.47
Conclusion				.06	DS/FXS: 0.05
Mean	2.96	2.92	2.29		DS/TD: 0.64
SD	1.08	0.52	1.01		FXS/TD: 0.78
Total Score (No Reference)				.05*	DS/FXS: 0.07
Mean	17.21	16.92	14.10		DS/TD: 0.69*
SD	5.00	3.75	3.97		FXS/TD: 0.73

* Significant at .05 level or better

Appendix A

Select Sections of *Frog Goes to Dinner* NSS Scoring Rubric

INTRODUCTION					
INTRODUCTION: Presence/absence of main characters and qualitative depiction of setting components throughout the story --Beginning house/bedroom can be included if there is good detail used to describe first scene even if the words "home" or "bedroom" are left out (i.e., getting ready to go out/leave)					
Main Settings	Beginning bedroom/house or getting dressed/ready Frog in Pocket	Restaurant or eating (not just ordering). Outside Restaurant Frog in salad	Car/ending house/bedroom Inside restaurant/sitting at table/ordering/menu Frog in wineglass		
Sub-Settings	Frog in saxophone				
Main Characters	Boy (at beginning or end)	Frog			
5 Proficient Use	4 Most details Included	3 Emerging/Inconsistent	2 Minor Details Included	1 Immature or Minimal	0 Poor Performance
- Clearly provides all 3 main settings and 5-6 sub-settings AND -Both main characters are clearly mentioned AND All components are well developed	-Clearly provides all 3 main settings and 3-4 sub-settings AND -Both main characters are clearly mentioned OR Includes all components, but not well developed	-4-5 settings mentioned (main or sub) AND mentions at least 1 main character OR -Mentions 2 characters AND 2 main settings OR -Mentions 1 character AND 3 main settings	-Provides 2-3 settings (main or sub) AND -Mentions 1 main character	-Provides 1 or less setting element OR -Mentions 1 or less main character	-Provides no setting or character elements of story -Child errors such as conversing with examiner, not completing/refusing task, unintelligibility
CONFLICT RESOLUTION					
CONFLICT RESOLUTION: Presence/Absence of conflicts and resolutions required to express the story as well as how thoroughly each is described. Take whole story into account.					
Conflicts	1. Boy is leaving with family, saying goodbye (NOT getting dressed) 2. Family goes to nice restaurant 3. Frog jumps into saxophone 4. Frog jumps onto salad plate in waiter's hand 5. Frog jumps into wine glass 6. Waiter tries to catch frog 7. Boy wants frog back from waiter 8. Father gets angry at boy and sends him to his room				
Resolutions	1. Frog jumps into boy's pocket 2. Family sits down for dinner at restaurant/eating/ordering 3. Musician expresses reaction (mad, surprised) 4. Lady expresses reaction (startled by frog on plate) 5. Man expresses reaction (surprised, upset) 6. Waiter takes frog away 7. Waiter gives frog back to boy (demands family to leave restaurant) 8. Frog and boy laugh about the day's events				
5 Proficient Use	4 Most details Included	3 Emerging/Inconsistent	2 Minor Details Included	1 Immature or Minimal	0 Poor Performance

CONFLICT RESOLUTION					
-Clearly states all conflicts and resolutions critical to advancing the plot of the story -All 8 pairs of conflicts/resolutions mentioned, must have both aspects of pair	-Clearly states most conflicts and resolutions in story -6-7 pairs of conflicts/resolutions mentioned, must have both aspects of pair	-Under-developed description of conflicts and resolutions critical to advancing the plot of the story OR -Not all conflicts and resolutions critical to advancing the plot are present -At least 4-5 complete pairs of conflicts/resolutions mentioned	-Conflicts mentioned with little resolution or resolutions mentioned with little conflicts -4+ <i>unmatched</i> conflicts/resolutions OR -Little use of conflict/resolution agreement -2-3 complete pairs	-Many conflicts and resolutions critical to advancing the plot are not present -1-3 unmatched conflict/resolution pairs	-No conflicts or resolutions mentioned throughout story -Child errors such as conversing with examiner, not completing/refusing task, unintelligibility

Appendix B

Frog Goes to Dinner MLU7 Narrative Samples

DS Sample		
\$ Child, Examiner	C And then this guy was (:) hold/ing him.	C (Um) They won't (gonna) have dinner.
E How does the story start?	C And then (:) he fell into the (:)drum.	E Anything else?
C The (um) boy is look/ing in *the mirror.	E Anything <else>?	C No.
E Anything else?	C <And everyone> was stand/ing.	E Ok, here/'s the next page.
C Yeah.	E Anything else?	C And then (:) this guy took the frog outside.
C The dog and the turtle (:) and the frog is[ew:are] look/ing at the boy.	C Nope.	C And then the boy stop/ed him.
E Good, here/'s the next page.	E Ok, here/'s the next page.	E Anything else?
C And also (:) he/'s say/ing goodbye to his dog and the turtle.	C And then he get[ew:got] madder (:) about (uh) his saxophone (:) and drum.	C Nope.
C And then the boy is leave/ing.	C And then the frog (jump/ed:) jump/ed to the (um) salad and hide[ew:hid].	E Ok, here/'s the next page.
E Anything else?	E Anything else?	C And then (:) the parent/s and the son and the daughter was upset with him to give his (um) frog back [eu].
C Nope.	C Nope.	C And he did.
E What about the frog?	E Ok, here/'s the next page.	C And then he said, "out before I call the police".
C Oh, and the frog is in there too.	C And then this guy[ew:woman] eat/'s (of) her food.	E What else?
E Ok, here/'s the next page.	C And then she saw the frog.	C That/'s it.
C They went to a (rest*) restaurant to have dinner.	E Anything else?	E Ok, here/'s the next page.
C And (they) they look happy.	C Nope.	C The daughter and the son and the parent/s were mad.
E Anything else?	E Ok, here/'s the next page.	C Go/ing back (to) home.
C No.	C And then the frog jump/ed out (:to) into a glass.	C And that/'s it.
E Ok, here/'s the next page.	C And then (stare) stare/ed (:) again.	E Ok, here/'s the next page.
C The frog (um) fly[ew:flew] out to the (um) saxophone while (um) they are order/ing dinner.	E What else?	C And then (:) his father said, "go to your room"!.
E Anything else?	C That/'s it.	C And then she was unhappy about it probably because she does/n't care.
C Nope.	E Ok, here/'s the next page.	C And that/'s it.
E Ok, here/'s the next page.	C And then (: he) he look/ed upset.	E Ok, how does the story end?
C And then he blew the (um) saxophone really noisy[ew:noisily].	C And then (:) the frog almost kiss/ed him.	C The end.
C And then (he tip/ed out) he tip/ed the saxophone (:) to get the frog out.	E Anything else?	C (um) The dog was on the bed.
C And then he got mad.	C Nope.	C And that boy and the frog was[ew:were] on the floor with the turtle.
E What else?	E Ok, here/'s the next page.	C I bet they had fun.
C That/'s it.	C And then he got mad.	C And that/'s it.
E Ok, here/'s the next page.	C And then the parent/s was[ew:were] way upset.	

DS Sample		
C And then (:) the frog jump/ed out.	C And then they falled[ew:fell].	
C And (um) the guy look/*ed at him.		
FXS Sample		
\$ Child, Examiner	C (And the, one of the guy*) One of the band member/s see/3s (*f) something in (the sa* his f*) his instrument.	E Next page.
E Tell me everything about the story for <each> page.	E Next page.	C And the waiter catch/3s him.
C <(Th*)> The boy/'s get/ing *ready for dinner.	C aAnd the (*f) frog jump/3s onto (the ma* uh) the player/z face.	C And (the um) "I think I/'m gonna faint".
C Go/ing out for dinner.	C And he/'s land/ing to[ew:on] the drum.	C "Don't faint there".
C And that frog want/3s to go with him.	E Next page.	E Next page.
C With him to the dinner.	C The bandleader/'s mad at him for breaking his drum.	C The boy say/3s "wait, that/'s my frog".
C But frog cannot do it.	C And the waiter then see/3s the frog.	C "Don't throw him out".
C Ok, turn it.	C (Frog in the see/3s him) Does/n't see him.	C And the people (speak) tell him *to be quiet.
C Yeah, I/'m done with it.	C So he jump/3s in the side of the salad.	C Ok.
C I/'m done with it.	C Turn it.	E Next page.
E Good, here/'s the next page.	C I said turn it.	C "That/'s my frog".
C Fine.	C No, don't get <xx>.	C "Don't throw him out".
C Frog jump/3s into the boy/z coat.	E <Next> page.	C And the waiter say/3s "get out of here".
C And then the boy wave/3s to (his) the dog and the turtle.	C The waiter bring/3s the woman the food.	C "Don't come ever again with that dumb frog".
C (But it) But the boy did/n't see the frog.	C And she scream/3s "there/'s a frog in my salad"!.	C Ok.
C Did/n't see (the f* the) his frog.	C That salad.	E Next page.
C The whole family leave/3s for (to) the restaurant.	E Next page.	C The family is upset (because because the boy/'s) because the boy (did/n't) brought his frog to dinner.
C Restaurant.	C (Woman) Lady/'s have/ing a heartattack.	C Turn it.
E Next page.	C And frog (ju*) jump/3s up.	E Next page.
C The family arrive/3s at the fancy restaurant.	C And jump/3s into a man/z wine.	C (Da*) Father say/3s "go to your room".
C Restaurant.	E Next page.	C "Don't bring that frog here to (that d*) that resaurant again".
C Allright.	C "I want that frog outta here"!	C Xx.
E Next page.	C (Um) "I want that frog outta here *for what he did to me".	C Ok, turn it.
C (The b* the) The waiter/'s take/ing the order.	C And then the (frog) frog kiss/ed the man.	E How does the story end?
C And frog jump/*3s into a saxophone.		C They laugh.
E Next page.		C The boy and the frog laugh their head/s off.
C The frog/'s (is) inside *the saxophone.		
TD Sample		
\$ Child, Examiner	E Next page.	C This grown man xx.

TD Sample

E How does the story start?	C He went out his horn[ew:saxophone].	C (S*) And this guy got mad.
C (Um hmm) *He got his pants off.	C And he jump/ed on his face.	C (What* what* what* what*) Whatever that is.
C Yeah, and he put new pants on.	C And then he was scared.	C And that/'s all.
C And he got a frog and turtle and *a pet (: and (um) a doggie.	C (Then he:) Then the boy/ed[ew:boy] laugh/ *ed.	C Why are you do/ing this marker xx?
C And that/'s all maybe.	C He xx that scared (of) of frog.	E Next page.
E Good, here/'s the next page.	C I/'m not.	C And (: h*) the man took the frog and>
C In case he jump/ed into his pocket>	C Frog/s don't bite.	C What does that say?
C And he did/n't know he/'s in his pocket.	E Next page.	C I don't know what it <say/3s>.
C And then he said bye to his turtle and dog.	C (Um) He jump/ed to the (p*) salad, whatever that is.	E <We> Can talk about that later.
C Then he went out the door.	C And then he/'s carry/ing (in) it.	C Ok.
C And that/'s all.	C Then he jump/ed and he whatever that is.	C (He open/3s) He almost open/ed the door.
E Next page.	C (Um: he) His band[ew:drum] broke.	C And (he) he carry/ed his way [EU].
C (H*) He got[ew:was] in the pocket still.	E Next page.	C And>
C And (the girl) the boy and girl and mom and dad and the policeofficer.	C The sir gave it to him[ew:her].	E Next page.
C And that/'s all maybe.	C And then he jump/ed out.	C (He he walk/ed up to him) they walk/ed up to him.
C And flower/s.	C Then he was eat/ing (it) all of it.	C And then (:) that/'s all.
C That/'s all.	C (Then he:) Then the frog went over.	E Next page.
E Next page.	C He/'s beautiful.	C They/'re all mad at him.
C (He/'s) These guy/s were play/ing music.	C (Let/'s) She/'s (be*) beautiful.	C And that/'s all.
C And then he jump/ed out of his pocket.	C Let/'s pretend.	E Next page.
C And then he went into his horn[ew:saxophone] up to here, way up there, right here.	C And that/'s all.	C And (all the p* all his pet/s) all of his pet/s were all>
C And then (h*) he swoop/ed up.	E Next page.	C These two (were) got>
E Next page.	C He jump/ed out.	C And then he went in his room.
C And he thought those guy/s were mad.	C Then he went into there.	C He laugh/ed and laugh/ed and laugh/ed.
C (And:) And he/'s in a circle.	E Next page.	C And that/'s all.
C Then around[pron:round], round and around[pron:round], round, round, round, round.	C And (these these) these guy/s (these girl) and whatever (it) he is, (a band a band whatever he is t*) were talk/ing.	C I just poke/ed my eye.
C Then he (f* f*) flip/3s out on his>	C And the frog went out (and then) and then kiss/ed his nose.	E How does the story end?
	C Gotta kiss his lip/s, or his hair, or his ear/s.	C *He laughed *and laughed.
	E Next page.	

NSS Scores: Introduction = 3; Character Development = 3, Mental States = 3, Conflict/Resolution = 3, Cohesion = 4, Conclusion = 3, Total = 19

NSS Scores: Introduction = 4; Character Development = 3, Mental States = 2, Conflict/Resolution = 4, Cohesion = 3, Conclusion = 3, Total = 19

NSS Scores: Introduction = 3; Character Development = 2, Mental States = 2, Conflict/Resolution = 2, Cohesion = 2, Conclusion = 3, Total = 14