Narrative Development in Adolescents and Young Adults With Fragile X Syndrome

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Abstract
The narratives of 18 adolescents and young adults with fragile X syndrome were compared to those of 23 adolescents with Down syndrome and 21 typically developing children matched for nonverbal MA. Narratives were elicited using a wordless picture book and analyzed for use of narrative evaluation, linguistic productivity, and complexity. Results revealed that the individuals with fragile X syndrome produced significantly fewer different types of narrative evaluation, but more grammatically acceptable utterances than did the youth with Down syndrome. There was no significant difference between the participants with fragile X syndrome and their typically developing nonverbal-MA matches. Results suggest that a variety of language measures and contexts are needed to gain a full understanding of the language abilities of individuals with fragile X syndrome.

Fragile X syndrome is the most common inherited cause of mental retardation (Hagerman & Brunschwig, 1991). It is caused by a mutation of the FMR1 gene on the X chromosome and results in a decrease in, or absence of, the gene’s associated protein (FMRP). Lower levels of FMRP lead to lower IQ and impairments in specific aspects of cognition, such as executive functioning (Cornish, Sudhalter, & Turk, 2004; Dyer-Friedman et al., 2002; Kaufmann, Abrams, Chen, & Reiss, 1999; Loesch, Huggins, & Hagerman, 2004). Because the condition is X-linked, males are more often and, on average, more severely affected than females (Lachiewicz & Mirrett, 2000; Mazzocco, Pennington, & Hagerman, 1994). Language and communication are impaired in individuals with this syndrome, with receptive language being relatively better than expressive language, although the difference between the two modalities increases with age (Abbeduto & Hagerman, 1997; Roberts, Mirrett, & Burchinal, 2001). Many of the researchers examining expressive language have found that males with fragile X syndrome have especially poor pragmatic skills, at least when assessed in conversational contexts (Abbeduto & Hagerman, 1997; Murphy & Abbeduto, 2003; Sudhalter & Belser, 2001). It is unclear, however, whether the deficits observed in conversation are evident in other language tasks or contexts, such as narration or story-telling. In light of the important role played by narrative skills in the acquisition of literacy as well as in the construction and representation of self (Ninio & Snow, 1996), we designed the present study in order to examine narrative abilities in youth with fragile X syndrome.

Numerous empirical studies have demonstrated that these individuals perform more poorly on several important dimensions of conversational ability than do individuals with different developmental disabilities (e.g., Down syndrome or autism) who were matched for chronological age (CA) or mental age (MA) (Belser & Sudhalter, 2001; Lachiewicz, Spiridigiozzi, Gullion, & Ransford, 1994; Wolf-Schein et al., 1987). Sudhalter et al. (1990) reported that males with fragile X syndrome had more difficulty with topic initiation and maintenance, as well as with producing pragmatically appropriate responses than did peers with other forms of mental retardation. During conversation, males with fragile X syndrome also have been found to produce more tangential comments than peers with autism or peers with mental retardation from a mixed-etiology group (Sudhalter & Belser, 2001).

Other investigators have found that males...
with fragile X syndrome have higher rates of perseveration, that is “excessive self-repetition” (Abbeduto & Hagerman, 1997, p. 317) during conversation than developmental level-matched typically developing children or youth with other developmental disabilities (Belser & Sudhalter, 2001; Ferrier, Bashir, Meryash, Johnston, & Wolff, 1991; Loesch et al., 2004; Murphy & Abbeduto, 2003). Belser and Sudhalter (2001) compared the conversational performance of males with fragile X syndrome to males with autism and individuals with mental retardation from other causes. In conversational language samples, the males with fragile X syndrome produced significantly more repetitions of phonemes, words, and phrases than either comparison group.

Language use in conversation, however, is not always an accurate reflection of language use in other contexts. Abbeduto, Benson, Short, and Dolish (1995), for example, demonstrated that some dimensions of language, including syntax and talkativeness, vary between conversation and narration both for typically developing children and youth with mental retardation. Expanding the range of contexts in which the language challenges of youth with fragile X syndrome are examined, therefore, will lead to a greater understanding of the full range of communication skills that define the phenotype of the syndrome. However, there is little empirical information about the communicative abilities of individuals with fragile X syndrome in other contexts, such as narration (Abbeduto & Hagerman, 1997; Murphy & Abbeduto, 2003).

Narrative ability has received increasing attention as a measure of linguistic performance in various clinical populations, such as children with specific language impairment (Gillam & Johnston, 1992; Liles, Duffy, Merritt, & Purcell, 1995) and children and adolescents with Down syndrome (Boudreau & Chapman, 2000; Miles & Chapman, 2002), Williams syndrome (Reilly, Losh, Bellugi, & Wulfeck, 2004) or autism (Capps, Losh, & Thurber, 2000). In order to produce a narrative, an individual must use his or her knowledge of the many dimensions of language (e.g., pragmatics, syntax, morphology, phonology, and semantics) to create a coherent discourse that engages and informs the listener (Gillam & Pearson, 2004). Researchers and clinicians have examined the use of these language skills at two different levels in narration: the macrostructure and the microstructure. Macrostructure refers to the global organization of narrative content, whereas microstructure refers to the amount of linguistic complexity at the lexical or sentence levels (see Hughes, McGillivray, & Schmidt, 1997; McFadden & Gillam, 1996).

Children with various types of language problems perform differently on measures of narrative microstructure, such as grammatical accuracy and complexity, total number of words used, and number of different words employed (Fey, Catts, Proctor-Williams, Tomblin, & Zhang, 2004; Gillam & Carlile, 1997; Gillam, Cowan, & Day, 1995; Gillam & Johnston, 1992; Liles et al., 1995). Norbury and Bishop (2003) investigated the narrative skills of four groups of children: specific language impairment, pragmatic language impairments, high-functioning autism, and those who were typically developing. The children were shown a wordless picture book (i.e., *Frog, Where Are You?) and produced a narrative describing the events in the story. The clinical groups differed from the typically developing children in the syntactic complexity of the utterances comprising their narratives.

There is also evidence that children with various types of language problems differ from typically developing children on various measures of macrostructure. Manhardt and Rescorla (2002) reported that 9-year-old children who were diagnosed with an expressive language delay as toddlers produced narratives that not only were less grammatically complex at the microstructure level, but they also contained fewer elements of evaluation than similarly aged children who were typically developing. Narrative evaluation, a measure of macrostructure, refers to a person’s ability to interpret the psychological states of characters or events that are not explicitly stated in the story (Bamberg & Reilly, 1996; Manhardt & Rescorla, 2002; McCabe & Peterson, 1984; Peterson & McCabe, 1991). Examples include the use of character dialogue, references to the character’s emotional or mental state, and causal connectors.

Tager-Flusberg (1995) reported differences at the macrostructure and macrostructure levels in the narratives of children with autism when compared to children with mental retardation and typically developing children matched for language age. The children with autism produced narratives that were shorter in length, less grammatically complex, and contained fewer causal connectors. The children with autism also had more difficulty labeling the characters’ emotions. Capps et al. (2000) found that children with autism and children with other types of developmental delays used fewer different types of narrative evaluation...
than did typically developing children, although the two groups did not differ in the overall frequency of evaluative devices.

Using a wordless picture book, Losh, Bellugi, Reilly, and Anderson (2000) compared the narratives of children with Williams syndrome to typically developing children matched for age and gender. The children with Williams syndrome produced more grammatical errors and less complex syntax. Despite these problems at the level of the microstructure, the children with Williams syndrome produced significantly more evaluative devices when compared to the typically developing children. Losh et al. suggested that this profile of strengths and weaknesses reflects the fact that children with Williams syndrome are highly social and interactive.

In contrast to the wealth of data on other disorders, studies of narrative in individuals with fragile X syndrome are virtually nonexistent. Medved and Brockmeier (2004) examined the ability of a single female with fragile X syndrome to produce autobiographical narratives given prompting and support. The authors reported that the participant had difficulty using causal relations and temporal cohesion, although they did not report quantitative data. Simon, Keenan, Pennington, Taylor, and Hagerman (2001) investigated story comprehension in high functioning women with fragile X and found that even those with normal-range IQs had difficulty selecting appropriate story endings, suggesting that they had problems constructing complete and coherent representations of the stories they heard. It is likely that such deficits will extend to narrative production and be more severe in lower functioning individuals with fragile X syndrome; however, there are no relevant data.

In summary, results of studies to date indicate that individuals with fragile X syndrome, both males and females, exhibit impairments in critical conversational abilities even when compared to individuals with other forms of mental retardation. Although conversational abilities represent one form of expressive language, studies of children and adolescents with other types of language impairments indicate that narrative ability is another aspect of language that should be investigated. As suggested by Losh et al. (2000), narratives provide an opportunity to examine the relationship between language ability, affect, and pragmatics and may well be an area of special difficulty for individuals with fragile X syndrome (Simon et al., 2001). The present study was designed to extend previous research and investigate the narrative ability of children and adolescents with fragile X syndrome, focusing on both the microstructure and macrostructure levels.

Method

Participants

Language samples were collected from 65 children and adolescents as part of a larger study of language development in individuals with mental retardation (Abbeduto et al., 2003). All participants had a mean pure tone threshold across the frequencies of 500, 1000, and 2000 Hz of no worse than 30 dB in their better ear. Thresholds could not be determined for two noncompliant typically developing children, but their parents reported no hearing problems. The sample overlaps with those reported on in other papers (e.g., Abbeduto et al., 2003; Abbeduto et al., 2006), although this is the first report on the participants’ narrative abilities.

The participants with fragile X syndrome and Down syndrome were recruited nationally through professional organizations and family-support groups as well as advertisements placed in newspapers in selected urban areas and locally through a university-based registry of potential research participants with developmental disabilities. Typically developing children were recruited through local preschools and school systems as well as by postings in local public places (e.g., shops).

The initial group with fragile X syndrome was comprised of 19 youth; however, a preliminary examination of the data revealed that one participant with fragile X syndrome produced an unusually long narrative, with a total of 412 utterances, which was 6.98 SDs above the mean length for all participants. Given the influence a substantially longer narrative would have on measures such as number of different words, this individual was considered an outlier and excluded from the study. The remaining 18 participants (12 males, 6 females) had a diagnosis of fragile X syndrome confirmed by a genetic test, with virtually all participants having had a molecular genetic test to confirm the presence of the full mutation (i.e., CGG repeats of 200 or greater), although cases of mosaicism were not excluded. For inclusion in the study, the adolescents had to use speech as their primary means of communication and have no physical impairments that would limit their ability to perform required tasks. Table 1 presents data.
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Table 1. Means, SDs, and Ranges for Age, Intelligence, and Mental Age by Group

<table>
<thead>
<tr>
<th>Characteristic</th>
<th>Fragile X (n = 18)</th>
<th>Down syndrome (n = 23)</th>
<th>Typically developing (n = 21)</th>
</tr>
</thead>
<tbody>
<tr>
<td>CA&lt;sup&gt;a&lt;/sup&gt;</td>
<td>16.68 ± 3.24</td>
<td>16.83 ± 2.92</td>
<td>4.91 ± 0.80</td>
</tr>
<tr>
<td>Nonverbal MA&lt;sup&gt;a&lt;/sup&gt;</td>
<td>5.02 ± 1.67</td>
<td>4.93 ± 1.01</td>
<td>4.68 ± 1.15</td>
</tr>
<tr>
<td>Nonverbal intelligence</td>
<td>44.31 ± 10.40</td>
<td>41.09 ± 6.3</td>
<td>90.55 ± 15.60</td>
</tr>
</tbody>
</table>

<sup>a</sup>In years.

for each group on age, nonverbal IQ, and nonverbal MA (as determined by our administration of the Copying, Bead Memory, and Pattern Analysis subtests from the Stanford-Binet Test of Intelligence, 4th edition (Thorndike et al., 1986). Any participant who met the Diagnostic and Statistical Manual of Mental Disorders—DSM-IV (American Psychiatric Association, 1994) criteria for an autism spectrum disorder was also excluded (see Abbeduto et al., 2003, for details of the autism diagnostic procedures).

Individuals with Down syndrome had a diagnosis of trisomy 21 according to parental report, which in most cases was verified by a medical report shared with the researchers. Twenty-five adolescents with Down syndrome (12 males, 13 females) participated in this study and served as a comparison group. Two participants with Down syndrome were excluded because they refused to participate in the task and did not produce enough utterances to be meaningfully analyzed. The remaining 23 participants with Down syndrome were matched groupwise to the participants with fragile X syndrome on the range and mean for CA and, as measured by the Stanford-Binet subtests for nonverbal IQ and MA. These participants, like those with fragile X syndrome, used speech as their primary means of communication and had no physical impairments that would have limited their ability to perform required tasks.

A second comparison group was comprised of 15 female and 6 male typically developing children. This group had a mean CA of 4.9 years. They were selected to be matched groupwise to the individuals with fragile X syndrome and Down syndrome on the mean and range of their nonverbal MAs, according to results on the Stanford-Binet subtests. None of the typically developing children had received special education services (other than occasional speech therapy) according to parental report and scored within normal limits on the Stanford-Binet subtests.

Data-Collection

As part of a more comprehensive protocol, a narrative was elicited from each participant using the wordless picture book, Frog Goes to Dinner (Mayer, 1977). Other books in this series, which are similar in structure and length, have been used to examine narrative development in cross-linguistic populations (e.g., typically developing children) and clinical populations (e.g., individuals with autism [Capps et al., 2000], Down syndrome [Miles & Chapman, 2002], and specific language impairment [Botting, 2002]). The participant viewed the book page by page once to become acquainted with the story, and during a second viewing, he or she told the story page by page to the examiner. The examiner, whose behavior was highly scripted, provided only minimal prompts.

Each narrative was audiotaped and ranged in length from 2.9 to 35 minutes in length (M = 6.07). The narratives were transcribed according to conventions of the Systematic Analysis of Language Transcripts software—SALT (Miller & Chapman, 2000). According to Miller and Chapman, individuals with language and speech impairments may have poor intelligibility and prosody, decreasing the accuracy and reliability of using intonation contour to segment utterances. Moreover, the tendency of older children to link their utterances by the use of and can unduly inflate summary measures of syntactic complexity based on utterance length. Thus, narratives were segmented into communication units (C-units), which are syntactically defined. In accordance with Loban (1976), a C-unit was defined as an independent clause and its modifiers.

Each narrative was recorded by a primary transcriber, and the transcript was independently

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checked against the audiotape by a second transcriber, who noted points of disagreement on the transcript. The primary transcriber then reviewed the annotated transcript against the audiotape and made any changes deemed necessary.

Seventeen percent of the narrative samples were randomly selected for intertranscriber reliability (4 each from the groups with fragile X syndrome and Down syndrome and 3 from the typically developing group). The mean percentage of agreement for the narratives was 89.9 for segmentation into C-units, number of morphemes and words per C-unit, identity of words in the C-unit, C-units with mazes, and C-units with mazes or unintelligible portions.

Data Analysis

The analyses were designed to examine narrative ability at both the microstructure (i.e., linguistic productivity and complexity) and macrostructure levels (i.e., evaluation devices). Only complete and fully intelligible C-units were used in the analyses. In order to ensure that the sample represented spontaneous language production, C-units that were produced in response to questions were excluded.

Linguistic productivity. Two measures of linguistic productivity, total number of C-units and number of different words root, were calculated using SALT software (Miller & Chapman, 2000). The number of C-units was an index of the length of the narrative sample. The number of different word roots measured lexical diversity.

Linguistic complexity. The measures of linguistic complexity were the mean length of C-units in words, percentage of grammatically acceptable C-units, clause density, and number of connectors per C-unit. The mean length of C-units in words was calculated automatically by the SALT software by dividing the total number of words by the total number of C-units and reflects syntactic complexity. A grammatically acceptable C-unit was defined as an utterance that was grammatically acceptable within Standard American English. The percentage of grammatically acceptable C-units was calculated by dividing the number of grammatically acceptable C-units by the total number of C-units in the narrative sample.

Clause density, another measure of syntactic complexity, was defined as the total number of main and dependent clauses divided by the number of C-units in the sample. The number of connectors per C-unit consisted of the number of causal and conditional connectors and divided by the total number of C-units. According to Gillam and Johnston (1992), conditional connectors denote that one clause modifies the occurrence of another clause (e.g., however, but, and if'), whereas a causal connector indicates that one clause explains or provides the motivation for another clause.

Narrative evaluation. Each C-unit was examined and coded for six evaluation devices, also known as high point analysis. Evaluation devices express or describe implicit elements of a story, such as opinions or mental states (Hughes et al., 1997). The following coding scheme, which were adapted from McCabe and Peterson (1991), Capps et al. (2000), and Losh et al. (2000), reflects frequency of occurrence in number of C-units:

- Mental state verbs: The number of verbs used that indicate or express a character’s internal state (e.g., believe, recognize, and forget).
- Character name: The number of C-units in which the narrator refers to characters by personal name (e.g., “The boy, Jack, went to the door”).
- Character dialogue: The number of C-units that reflect a character’s voice or speech (e.g., “Then the boy said, ‘Get out of the barn’”)
- Repetition: The number of C-units that contain a word that is repeated for emphasis, not to clarify information (e.g., “He was very, very angry”).
- Onomatopoeia, sound effects, or exclamations: The number of C-units that contain an onomatopoeic reference or sound effects (e.g., ‘The frog went bong’) or that are exclamations, such as “Oh, boy!”
- Fantasy or exaggeration: Exaggeration is defined as the number of C-units that refer to an action or event that is beyond the reality, such as “The frog jumped out of the room” when in actuality the frog only jumped off the bed. Fantasy reflects the number of C-units depicting a fictional event, such as “The boy’s grandmother went to the store” when there is no evidence of a grandmother in the story.
- Evaluation density: The total number of evaluative devices used divided by the total number of C-units in the sample.

Two graduate students in speech-language pathology received training on the coding system, and intercoder reliability for the narratives codes was calculated for a randomly selected set of samples (17%). Point-by-point intercoder reliability was 91% for Name, 96% for Dialogue, 90% for Repetition, 83% for Onomatopoeia, 100% for Fantasy, and 91% for Mental State Verbs. After determining the point-by-point agreement, we calculated Cohen’s kappa statistic (Cohen, 1960) to correct for chance agreement. According to Fleiss (1981), kappas above .60 are considered good and kappas above .75 are
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Table 2. Means and SDs for Linguistic Productivity and Complexity by Group

<table>
<thead>
<tr>
<th>Variable</th>
<th>Fragile X syndrome (n = 18)</th>
<th>Down syndrome (n = 23)</th>
<th>Typically developing (n = 21)</th>
</tr>
</thead>
<tbody>
<tr>
<td>No. of C-unitsa</td>
<td>41.83 20.00</td>
<td>56.96 25.76</td>
<td>38.67 19.51</td>
</tr>
<tr>
<td>No. of different words</td>
<td>87.78 41.30</td>
<td>104.04 45.79</td>
<td>78.81 25.10</td>
</tr>
<tr>
<td>Mean length of C-units in words</td>
<td>5.80 2.10</td>
<td>5.02 1.28</td>
<td>5.78 1.04</td>
</tr>
<tr>
<td>Percentage of grammatically acceptable C-units</td>
<td>82.69 12.47</td>
<td>69.46 13.59</td>
<td>88.64 12.80</td>
</tr>
<tr>
<td>Clause density</td>
<td>.79 .15</td>
<td>.77 .15</td>
<td>.83 .14</td>
</tr>
<tr>
<td>Mean number connectors per C-unit</td>
<td>0.13 .1</td>
<td>0.11 .11</td>
<td>0.10 .12</td>
</tr>
</tbody>
</table>

aCommunication units.

Results

Although the groups were matched for nonverbal MA, they differed in gender distribution. As previously discussed, gender is closely related to cognitive ability in fragile X syndrome; therefore, we sought to examine the influence of gender and nonverbal MA on the dependent measures.

Linguistic Complexity and Productivity

A multivariate analysis of variance (MANCOVA) was used to examine the effect of group membership (fragile X syndrome vs. Down syndrome vs. typically developing) on the following six dependent variables: number of C-units, number of different words, mean length of C-units in words, percentage of grammatically acceptable C-units, clause density, and number of causal connectors. Nonverbal MA and gender were the covariates. The Levene’s test of equality of error variances for number of different word roots was significant, \( F(2, 59) = 4.76, p < .01 \); therefore, the natural logarithm of this variable was used in all analyses.

There was a significant effect for group, Wilks’ Lambda \( F(12, 100) = 3.11, p = .001 \), partial \( \eta^2 .72 \). Table 2 contains the means and SDs for the dependent variables by group. Follow-up tests to the MANCOVA were evaluated with the Holm sequential procedure (Holm, 1979; Levin, Serlin, & Seaman, 1994) to control for Type I errors. Using this method, we ranked the univariate tests from the largest to the smallest. The largest \( F \) had to reach an alpha of .008 (.05 divided by 6) to be significant, the next largest \( F \), an alpha of .01 (.05 divided by 5), and so on, with the smallest \( F \) needing to reach an alpha .05 for significance. Only the univariate test for percentage of acceptable C-units was significant, \( F(2, 55) = 14.13, p < .001 \). Post hoc comparisons were conducted using Fisher’s LSD. The results revealed that the participants with fragile X syndrome, \( p = .001 \), and the typically developing MA-matches, \( p < .001 \), produced significantly more grammatically correct C-units than did the individuals with Down syndrome.

Narrative Evaluation

Table 3 presents the summary statistics for the amount and type of evaluation devices used by the participants. Significant Kolmogorov-Smirnov tests, \( p < .0001 \), indicated that the six evaluation devices were not normally distributed; thus, we used nonparametric Kruskall-Wallis one-way ANOVA tests in narrative analyses. Mann-Whitney \( U \) tests were employed to follow-up any significant findings. We analyzed group differences first and then examined gender differences within each group.

Kruskal-Wallis tests were conducted to evaluate differences among the three groups for mental state verbs, character name, character dialogue, repetition, onomatopoeia or exclamations, and fantasy or exaggeration. The Holm sequential procedure was used to control for Type I error. In this procedure, the largest \( \chi^2 \) had to reach an alpha of .008 (i.e., .05/6); the next, an alpha of .01 (i.e., .05/5); and continuing until the smallest \( \chi^2 \) required an alpha of .05 to reach significance.

As shown in Figure 1, significant between-group differences were detected in the use of onomatopoeia or exclamations, \( \chi^2(2, N = 62) = \) excellent. Intercoder reliability across all narrative codes resulted in a kappa of .72.
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Table 3. Means and SDs for Measures of Narrative Evaluation and Plot Themes by Group

<table>
<thead>
<tr>
<th>Evaluation and themes</th>
<th>Fragile X syndrome (n=18)</th>
<th>Down syndrome (n=23)</th>
<th>Typically developing (n=21)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Mean</td>
<td>SD</td>
<td>Mean</td>
<td>SD</td>
</tr>
<tr>
<td>Evaluation devices</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Mental state verbs</td>
<td>2</td>
<td>3.43</td>
<td>1.17</td>
</tr>
<tr>
<td>Character name</td>
<td>0.33</td>
<td>1.41</td>
<td>0.91</td>
</tr>
<tr>
<td>Character dialogue</td>
<td>2.39</td>
<td>4.03</td>
<td>5.3</td>
</tr>
<tr>
<td>Repetition</td>
<td>0</td>
<td>0.17</td>
<td>0.39</td>
</tr>
<tr>
<td>Onomatopoeia or exclamations</td>
<td>0.17</td>
<td>.51</td>
<td>1.87</td>
</tr>
<tr>
<td>Fantasy or exaggeration</td>
<td>0.17</td>
<td>.71</td>
<td>0.17</td>
</tr>
<tr>
<td>Evaluation density</td>
<td>.10</td>
<td>.13</td>
<td>.17</td>
</tr>
<tr>
<td>Evaluation diversity (6 maximum)</td>
<td>1.39</td>
<td>1.14</td>
<td>2.39</td>
</tr>
</tbody>
</table>

15.40, \(p < .008\). We conducted follow-up tests using Mann-Whitney \(U\) test. The typically developing MA matches (range 0 to 6 C-units) and the participants with fragile X syndrome (range 0 to 13 utterances) did not differ in their use of onomatopoeia or exclamations. The individuals with Down syndrome, however, used this evaluation device more frequently (range 0 to 31 C-units) than did either the group with fragile X syndrome, \(Z = -3.69, p < .001\), or the MA-matched typically developing participants, \(Z = -2.45, p < .05\).

There were no differences between the groups in number of mental state verbs, character names, character dialogue, repetition, or the amount of fantasy or exaggeration used in the narrative.

Kruskall-Wallis tests were also used to assess evaluation density, \(\chi^2(2, N = 62) = 7.80, p < .05\), and evaluation diversity, \(\chi^2(2, N = 62) = 7.79, p < .05\), both of which were significant (see Table 3). Mann-Whitney \(U\) follow-up tests revealed that individuals with fragile X syndrome had lower evaluation density scores than did the participants with Down syndrome, \(Z = -2.25, p < .05\), but did not differ from the MA-matched typically developing group, \(Z = -.64, p < .05\). The group with fragile X syndrome also produced fewer different types of evaluative devices, reflected in lower evaluation diversity scores, than did the group with Down syndrome, \(Z = -2.66, p < .01\), but performed similarly to the typically developing MA-matches. The participants with Down syndrome produced higher scores than did the MA-matches on evaluation diversity, \(Z = -2.1, p < .05\), and evaluation density, \(Z = -2.48, p < .05\).

Separate nonparametric Kruskall-Wallis tests were conducted for each evaluation measure to determine whether there were differences between genders within each of the three groups. No effects were significant at the .05 level on any measure.

Discussion

Our aim in this study was to investigate the narrative ability of individuals with fragile X syndrome and their performance relative to peers matched for nonverbal MA. By using two control groups, one with and one without mental retardation, we were able to determine whether the results were due to cognitive ability or reflected characteristics that were possibly syndrome specific. We focused on measures that assessed the microstructure (at the word and C-unit level) and macrostructure (at the text level) of the narratives. Overall, our findings suggest that the narratives of the participants with fragile X syndrome resembled those of the typically developing control group more than those of the group with Down syndrome.
When compared on measures of linguistic complexity and productivity, for which we examined the microstructure, the three groups differed only on the percentage of grammatically acceptable utterances. The participants with fragile X syndrome produced significantly more grammatically correct utterances than did the group with Down syndrome. This finding is consistent with the extensive literature documenting the expressive language deficits, particularly morphology and syntax, in children and adolescents with Down syndrome (Abbeduto et al., 2001; Chapman & Hesketh, 2000; Chapman, Seung, Schwartz, & Bird, 1998; Fowler, 1990; Miller, 1999). However, the groups did not differ on other linguistic measures (i.e., number of C-units, number of different words, mean length of C-units in words, clause density, and mean number of connectors) that other studies have shown discriminate children with language disorders (Fey et al., 2004; Gillam & Johnston, 1992; Liles et al., 1995; Scott & Windsor, 2000). The lack of difference on these measures may be due to sampling context. Abbeduto, Benson, Short, and Dolish (1995) found that children and adolescents, both those who had mental retardation and those who were developing typically, produced fewer total utterances but used more syntactically complex language in narration than in spontaneous conversation. Another possible explanation is that in the present study, the focus on only spontaneous utterances and exclusion of responses to questions may have reduced the length of the narratives and minimized differences between the participants.

In contrast to measures of linguistic complexity, substantial differences were seen in the use of evaluation devices among the groups. The performance of the participants with fragile X syndrome again resembled that of the typically developing MA-matched comparison group, but here the differences favored the individuals with Down syndrome. Both the fragile X syndrome and typically developing groups used less onomatopoeia or exclamations than did the individuals with Down syndrome, who also used more different types of evaluation devices, even when controlling for length of narrative, as reflected by evaluation density, than did the other groups. These results indicate that although individuals with Down syndrome may have more difficulty with the syntactic and morphological components of language, their narratives may reflect the use of other types of advanced language skills. This claim is further supported by the results of Miles and Chapman (2002), who found that individuals with Down syndrome produced more plot theme information than did typically developing controls matched for mean length of utterance (in morphemes). This suggests that, as claimed previously by Peterson and McCabe (1991), a variety of analysis methods are needed to gain an understanding of the range and complexity of language ability. Thus, although the group with fragile X syndrome produced more grammatically acceptable C-units, a measure of microstructure, they also produced fewer evaluation devices, a measure of macrostructure.

There are limitations that should be considered in conjunction with these results. First, replication with a larger sample of participants, particularly with regard to the number of females within the fragile X syndrome group, is needed. Although the males and females with fragile X syndrome did not differ significantly on any of the microstructure or macrostructure variables, a larger sample would increase probability of detecting differences that have been found in other studies of gender in fragile X syndrome (Abbeduto et al., 2003; Abbeduto & Hagerman, 1997).

Second, as discussed earlier, there was within-group variation in regard to the length of the narrative samples in C-units and duration. Differences in the length, as measured by number of C-units, could influence the performance on more traditional measures of microstructure, such as mean length of C-unit in words. However, as observed by Paul (2001) and Hughes et al. (1997), narrative language samples tend to be shorter in length than those collected in conversation. Our finding that the groups did not differ on the number of C-units or mean length of C-unit, is consistent with findings by Fey et al. (2004). In that study, the percentage of grammatically acceptable utterances yielded greater group differences than did story length or mean length of utterance between second and fourth grade. It is also notable that the mean number of C-units for an oral story was 11.24 for second graders and 13.15 for fourth graders (Fey et al., 2004), similar to the lower range of story length in this study.

A third limitation concerns the type of narrative task that we employed in this investigation. Although the Frog stories have been widely used to examine narrative ability, other narrative tasks such as story-retelling or story generation without picture support may have yielded different results (Gillam & Johnston, 1992; Hughes et al., 1997; Manhardt & Rescorla, 2002). Future researchers
should compare different elicitation methods to further investigate the narrative ability of individuals with fragile X syndrome.

In summary, these preliminary findings suggest that individuals with fragile X syndrome produce narratives similar to those of typically developing peers matched for nonverbal MA on microstructure (e.g., linguistic complexity and productivity) and measures of macrostructure (e.g., evaluation devices). Interestingly, the participants with Down syndrome used a greater variety of evaluation devices; suggesting that despite poor syntactic abilities, these individuals can produce narratives that are enriched in other ways. It is, perhaps, even more interesting that despite their relatively greater syntactic skills, the youth with fragile X syndrome did not make greater use of evaluation devices.

Paul (2001) described the use of evaluation or high point information as “story sparkle,” those elements that increase the overall quality of the narrative but are not adequately captured by traditional syntactic or lexical measures. Researchers examining typically developing children have found that with age, these children begin to use story sparkle to enhance their stories. It would be interesting to determine whether and when individuals with fragile X syndrome also begin to use these features or determine whether their narratives remain static, without further development. Future investigators should address other methods of analysis that may indicate other patterns of strengths and weakness in the narratives of individuals with fragile X syndrome. For example, because prior research has suggested that individuals with this disorder have difficulty maintaining top-

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