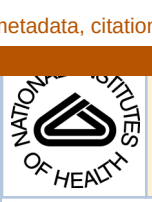

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An Investigation of Narrative Ability in Boys with Autism and Fragile X Syndrome

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Abstract

Whereas pragmatic language difficulties are characteristic of both autism and Fragile X syndrome, it is unclear whether such deficits are qualitatively similar or whether certain skills are differentially affected. This study compared narrative competence in boys with autism, Fragile X syndrome, Down syndrome, and typical development. Results revealed that an interaction between diagnosis and nonverbal mental age predicted narrative microstructure (e.g., complex syntax) but not macrostructure (e.g., thematic maintenance). Correlations with *FMR1*-related variation were investigated in children with Fragile X syndrome. While CGG repeat length was associated with many language characteristics, nonverbal IQ appeared to mediate these relationships. These findings are an important step toward understanding narrative abilities in boys with and without the *FMR1* mutation.

Keywords

Autism; Fragile X syndrome; narrative; mental age; *FMR1*; Down syndrome

Fragile X syndrome is the most common inherited form of intellectual disability (Crawford, Acuna, & Sherman, 2001), caused by a mutation in the Fragile X mental retardation 1 gene (*FMR1*), located on the X chromosome (Brown, 2002). This mutation results in cognitive deficits and language difficulties, as well as behaviors often associated with autism, such as social avoidance, gaze aversion, and repetitive and stereotyped behaviors (Hagerman & Hagerman, 2002). Autism is a neurodevelopmental disorder characterized by significant impairments in communicative and social functioning and the presence of restricted interests and repetitive behaviors (American Psychiatric Association, 1994). Autistic disorder along with Asperger syndrome and pervasive developmental disorder——not otherwise specified (PDD-NOS) compose a family of disorders known as autism spectrum disorder (ASD). For the purpose of this article, the term “autism” will be used to refer more generally to any autism spectrum disorder, unless specified otherwise.

Previous studies have estimated that the percentage of males with Fragile X syndrome who also meet “gold standard” diagnostic criteria for autistic disorder is between 18% and 52%, with prevalence rates as high as 74% when all autism spectrum disorders are considered

(Clifford et al., 2007; Hall, Lightbody, & Reiss, 2008; Kaufmann et al., 2004; Philofsky, Hepburn, Hayes, Hagerman, & Rogers, 2004; Rogers, Wehner, & Hagerman, 2001). Furthermore, approximately 2% to 6% of children with autism also have Fragile X syndrome (Hagerman, 2006).

Despite the increased rates of autism symptomatology in children with Fragile X syndrome, it is unclear whether autism in the context of Fragile X syndrome represents the same etiological basis and/or clinical presentation as idiopathic autism. Thus, cross-population studies directly comparing children with autism and those with Fragile X syndrome and comorbid autism can provide valuable insight into whether these symptoms are indicative of a shared disorder and can have important implications for intervention as well as studies of the causal basis of autism. The present study aims to compare narrative abilities in children with autism and Fragile X syndrome (with and without autism) in an attempt to better characterize the pragmatic language phenotypes of the two disorders.

Language deficits are hallmark features of both autism and Fragile X syndrome. In children with autism, both receptive and expressive language impairments are common (Tager-Flusberg, 2000), although receptive language abilities appear to be more severely impacted than expressive language abilities (Gillum & Camarata, 2004; Hudry et al., 2010; Kjelgaard & Tager-Flusberg, 2001; Luyster, Kadlec, Carter, & Tager-Flusberg, 2008; Paul & Sutherland, 2005). Some studies have reported that children with autism also demonstrate syntactic deficits, although others have found intact syntactic development relative to matched controls. For example, Eigsti, Bennetto, and Dadlani (2007) found that, in the context of free play with an examiner, 5-year-old children with autism produced spontaneous language that was less complex than the language produced by developmentally delayed and typically developing children matched on nonverbal IQ and receptive vocabulary. Specifically, the children with autism produced shorter utterances, as indicated by shorter mean length of utterance, and lower scores on the Index of Productive Syntax (IPSyn; Scarborough, 1990), despite intact lexical abilities and receptive vocabulary. Other studies, however, have found that children with autism do not demonstrate syntactic impairments relative to either established norms (Condouris, Meyer, & Tager-Flusberg, 2003) or language-matched typically developing children (Anderson et al., 2007).

Children with Fragile X syndrome tend to have receptive and expressive language abilities that are either commensurate with or delayed relative to their nonverbal mental age (Abbeduto, Brady, & Kover, 2007; Abbeduto & Hagerman, 1997; Abbeduto et al., 2003; Price, Roberts, Vandergrift, & Martin, 2007; Roberts, Chapman, Martin, & Moskowitz, 2008; Roberts, Price, et al., 2007). Their syntactic development also tends to be impaired relative to younger typically developing children matched on cognitive abilities. For example, children with Fragile X syndrome demonstrate shorter mean length of utterance and tend to produce fewer complex phrases, even when nonverbal mental age and maternal education are controlled (Roberts, Hennon, et al., 2007; Sudhalter, Scarborough, & Cohen, 1991).

Less is known about the language abilities of children with Fragile X syndrome and comorbid autism. Some studies have found that these children demonstrate more severe language deficits than children with Fragile X syndrome only (Bailey, Hatton, Skinner, & Mesibov, 2001; Lewis et al., 2006; Philofsky et al., 2004), whereas others have not reported such differences (Kaufmann et al., 2004; Kover & Abbeduto, 2010; Roberts, Mirrett, & Burchinal, 2001). Furthermore, the lack of idiopathic autism control groups in these studies leaves unanswered questions concerning the extent of phenotypic similarity between autism in the context of Fragile X syndrome and autism of idiopathic etiology.

This is also true for the domain of pragmatic language (i.e., the use of language for social interaction), which has rarely been compared across these disorders. Pragmatic language appears to be universally impaired in individuals with autism, even in the absence of structural language impairment and/or intellectual disability. These pragmatic impairments include, but are not limited to, difficulty initiating and maintaining a conversational topic (Baron-Cohen, Leslie, & Frith, 1988; Bishop & Adams, 1989; Fine, Bertolucci, Szatmari, & Ginsberg, 1994; Tager-Flusberg & Anderson, 1991), difficulty with conversational turn-taking (Botting & Conti-Ramsden, 2003), and inclusion of irrelevant or inappropriate information in conversation (Adams, Green, Gilchrist, & Cox, 2002; Landry & Loveland, 1989; Tager-Flusberg & Anderson, 1991; Volden, 2004).

Children with Fragile X syndrome also have pervasive difficulties with pragmatic language (Roberts, Martin, et al., 2007; Sudhalter & Belser, 2001; Sudhalter, Cohen, Silverman, & Wolf-Schein, 1990; Wolf-Schein et al., 1987). Like children with autism, they tend to include irrelevant and tangential utterances (Sudhalter & Belser, 2001), and they also demonstrate more perseverative language than typically developing children (Roberts, Martin, et al., 2007). However, children with Fragile X syndrome appear better at conversational turn-taking than children with autism when matched on chronological and mental age (Sudhalter et al., 1990). Importantly, children with Fragile X syndrome with comorbid autism have been shown to demonstrate more severe pragmatic impairments than those without comorbid autism, in that they have difficulties continuing the conversation with appropriate and relevant information, and they more frequently change conversational topics without signaling to the listener (Roberts, Martin, et al., 2007).

To our knowledge, only one study (Losh, Martin, Klusek, Hogan-Brown, & Sideris, 2012) has directly compared pragmatic language abilities in children with autism and children with Fragile X syndrome (with and without autism). This study found that children with autism and children with Fragile X syndrome and comorbid autism performed more poorly on a standardized assessment of pragmatic language than children with Fragile X syndrome without autism and typical developing children. Thus, there is some evidence that children with Fragile X syndrome and autism exhibit similar patterns of pragmatic language difficulties.

Narrative discourse is an important pragmatic language skill as it requires integration of a diverse range of pragmatic skills, including appropriate introduction of a topic, assessment of a listener's knowledge and comprehension, and inclusion of relevant information (Bamberg, 1997; Karmiloff-Smith, 1985; Labov & Waletsky, 1967). Socially, narrative storytelling plays a critical role in daily life, in that it is through stories that individuals interpret and communicate their experiences in meaningful ways (Bamberg & Reilly, 1996; Berman & Slobin, 1994; Bruner, 1987, 1990; 1991, 1997; Ochs & Capps, 2001), Thus, impairments in narrative ability can significantly impact a child's social interactions, and can undermine both social and communicative competence.

Because narrative abilities provide insight into linguistic, cognitive, and social-cognitive capacities, narrative production has been studied extensively in individuals with autism, who demonstrate pervasive difficulties in all three domains. Children with autism and children with Fragile X syndrome both have demonstrated impairments at the microstructure level, which refers to the structure of and relationship between propositions used to construct the narrative (Kintsch & van Dijk, 1978). Analysis of complex syntax in narratives has been commonly used to evaluate narrative coherence and organization (Bamberg & Damred-Frye, 1991; Bamberg & Marchman, 1991; Bamberg & Reilly, 1996; Berman & Slobin, 1994). In narratives, complex syntax is a marker of linguistic complexity and also serves the critical role of tying events and episodes together to form a cohesive discourse (Berman & Slobin,

1994). Previous studies have found that children with autism produce fewer propositions and use less complex syntax than typically developing children (Capps, Losh, & Thurber, 2000). Adolescents and young adults with Fragile X syndrome also use less complex syntax than typically developing mental age-matched participants in their narratives (Finestack & Abbeduto, 2010), but they do not differ on other aspects of narrative microstructure (e.g., mean length of utterance, grammaticality of utterances; Finestack & Abbeduto, 2010; Keller-Bell & Abbeduto, 2007).

Children and adolescents with autism have also been shown to demonstrate consistent impairments in narrative macrostructure, which refers to features such as cohesion, story structure, and overall narrative quality, and therefore focuses on the holistic organization of the discourse (Kintsch & van Dijk, 1978). In the context of narratives elicited by wordless storybooks, children with autism tend to use less evaluation (e.g., referring to characters' thoughts and emotions) to add meaning and perspective to narratives, and less frequently employ causal-explanatory frameworks to integrate and elaborate narrated events (Capps, Losh, & Thurber, 2000; Diehl, Bennetto, & Young, 2006; Losh & Capps, 2003; Tager-Flusberg, 1995). They are also more likely to include irrelevant, inappropriate, or otherwise bizarre utterances (Diehl et al., 2006; Loveland, McEvoy, & Tunali, 1990). Higher functioning adolescents with autism demonstrate difficulties with personal narratives in conversational contexts but are able to produce storybook narratives comparable to those of typically developing peers in most respects (Losh & Capps, 2003). In general, individuals with autism tend to produce narratives that lack coherence and fail to meet the cultural expectations of storytelling, implying a lack of understanding of the use of narratives as a tool for organization and communicating experiences (Bruner & Feldman, 1993; Diehl et al., 2006; Loveland et al., 1990).

Studies of narrative macrostructure in children with Fragile X syndrome report inconsistent results, with some suggesting impaired performance and others reporting intact or even enhanced abilities, relative to control groups. For example, Finestack, Palmer, and Abbeduto (2012) found that adolescents with Fragile X syndrome and with Down syndrome produced narratives with better story introduction and better overall quality than those produced by mental age-matched typically developing participants. However, Keller-Bell and Abbeduto (2007) found that children with Fragile X syndrome and typically developing controls used less evaluation and fewer types of evaluation than children with Down syndrome. Another study examining story grammar using a narrative recall task (Estigarribia et al., 2011), found that children with Fragile X syndrome (with and without comorbid autism) recalled characters' actions toward a goal less frequently than typically developing children. Furthermore, children with Fragile X syndrome and comorbid autism had a lower overall story structure score than typically developing children, suggesting that the presence of autism in these children was related to poorer narrative construction during recall.

To our knowledge, studies to date have not directly compared narratives from children with Fragile X syndrome with and without autism relative to children with idiopathic autism, leaving unclear how this critical skill is impacted across these disorders and whether similar difficulties may be evident in autism and Fragile X syndrome. The primary objective of this study was to characterize narrative ability in children with autism, children with Fragile X syndrome and comorbid autism, and children with Fragile X syndrome only, and to compare performance to children with Down syndrome and children with typical development, who served as controls for intellectual disability and developmental level, respectively. Based on prior research on narrative abilities in idiopathic autism (Capps et al., 2000; Diehl et al., 2006; Losh & Capps, 2003; Tager-Flusberg, 1995; Tager-Flusberg & Sullivan, 1995) and Fragile X syndrome with and without autism (Estigarribia et al., 2011; Finestack & Abbeduto, 2010; Finestack et al., 2012; Keller-Bell & Abbeduto, 2007), as well as research

on the phenotypic overlap between autism and Fragile X syndrome (Bailey, Hatton, Skinner, et al., 2001; Bailey et al., 2004; Demark, Feldman, & Holden, 2003), it was hypothesized that children with autism and children with Fragile X syndrome and comorbid autism would demonstrate similar patterns of narrative ability at both the microstructure and macrostructure levels. It was also predicted that narratives produced by children with only Fragile X syndrome would resemble those produced by children with Down syndrome.

Regarding narrative microstructure, it was predicted that children with idiopathic autism and Fragile X syndrome with autism would produce fewer propositions and use less complex syntax than the other children. At the macrostructure level, it was hypothesized that children with idiopathic autism and Fragile X syndrome with comorbid autism would produce narratives with less evaluation and poorer story structure, as evidenced by reduced use of thematic maintenance devices and fewer references to main story episodes. Finally, it was predicted that children with autism and Fragile X syndrome and comorbid autism would include more inappropriate/irrelevant, off-topic, and unintelligible utterances in their narratives.

A secondary objective of this study was to investigate the relationship between *FMR1*-related genetic variation and measures of general linguistic competence, intellectual ability, autism severity, and narrative ability in the children with Fragile X syndrome. Bailey and colleagues (2001) found that expression of FMRP, the protein encoded by *FMR1*, was related to level of development on a global measure of communicative ability in boys with Fragile X syndrome, and one recent study reported that *FMR1*-related variation was associated with pragmatic language, nonverbal cognitive abilities, and receptive and expressive vocabulary (Losh et al., 2012). However, other studies have found that FMRP expression does not relate to symptoms of autism (including social and communication symptoms) after accounting for nonverbal or full-scale IQ (Loesch et al., 2007; McDuffie et al., 2010). Thus, it was predicted that genetic characteristics would be correlated with language characteristics, autism severity, and narrative performance, but that such relationships may be mediated by nonverbal IQ.

Method

Participants

Twenty-one children with autism (i.e., an autism spectrum disorder), 23 children with Fragile X syndrome and comorbid autism, 19 children with Fragile X syndrome only, 19 children with Down syndrome, and 17 typically developing children were initially included in this study as part of a larger longitudinal study of pragmatic language development. Children with developmental disabilities were recruited through the Research Registry of the Carolina Institute for Developmental Disabilities at the University of North Carolina at Chapel Hill, genetic clinics, developmental evaluation clinics, and/or parent support groups. Typically developing children were recruited from pediatricians' offices, schools, and childcare centers in North Carolina.

To be included in this study, all children were required to be at or beyond Brown's (1973) Stage III (i.e., generally using three or more words per utterance), with English being the primary language spoken in their home. All children were required to have an average hearing threshold of less than or equal to 30 dB HL in the better ear, determined from a hearing screening across 500, 1,000, 2,000, and 4,000 Hz with a MAICO MA 40 audiometer. Typically developing children were required to be within 1.5 standard deviations of the age-appropriate mean for expressive vocabulary, receptive vocabulary, and nonverbal IQ. Language level, expressive and receptive vocabulary, and nonverbal IQ were assessed prior to enrollment in the study, and if children did not meet these criteria, they

were excluded from the study. Children with autism, Fragile X syndrome, and Down syndrome were included in this study only if they had been previously diagnosed with one of these disorders. The Social Communication Questionnaire (SCQ; Rutter, Bailey, & Lord, 2003) was used to screen out autism in the typically developing participants and those with Down syndrome.

Autism spectrum disorder classifications were determined using the Autism Diagnostic Observation Schedule (ADOS; Lord et al., 2000) revised diagnostic algorithms (Gotham, Risi, Pickles, & Lord, 2007). In the autism group, 17 children (81%) met the cutoff score for autistic disorder, and three children (14.3%) met cutoff for autism spectrum disorder. One child (4.7%) did not meet cutoff on the ADOS and was therefore dropped from the study. Of the children in the Fragile X syndrome with autism group, 19 (82.6%) met cutoff for autistic disorder on the ADOS, and four (17.4%) met cutoff for autism spectrum disorder. No children in the Fragile X syndrome without autism group or in the typically developing group met cutoff for autism spectrum disorder on the ADOS. One child with Down syndrome did, however, score above cutoff on the ADOS and was therefore dropped from the study.

Groups were initially matched on a composite score of expressive and receptive vocabulary skills, obtained through administration of the Expressive Vocabulary Test (EVT; Williams, 1997) and the Peabody Picture Vocabulary Test—Third Edition (PPVT-III; Dunn & Dunn, 1997). However, after the participants described above were dropped from the study, groups were no longer matched. To achieve proper matching, one child with Fragile X syndrome without autism and one child with typical development were dropped because their scores represented the highest composite language scores (2.91 and 1.92 SD greater than their group means, respectively), and one child with Down syndrome was dropped because of a low composite score (1.93 SD less than the group mean). After these participants were dropped from the study, independent samples *t*-tests were employed to compare groups on the EVT/PPVT composite score, and all *p* values were greater than .50, indicating that they were well matched on this measure (Mervis & Robinson, 2003). The final study sample was composed of 20 children with autism, 23 children with Fragile X syndrome and comorbid autism, 18 children with Fragile X syndrome without autism, 17 children with Down syndrome, and 16 typically developing children.

Expressive and receptive vocabulary age equivalent scores were also calculated based on performance on the EVT and PPVT, respectively. The Leiter International Performance Scale—Revised (Leiter-R; Roid & Miller, 1997) was used to obtain nonverbal mental age equivalent and IQ scores for each child. Standardized ADOS severity scores were calculated based on the procedures described by Gotham, Pickles, and Lord (2009). This score provides a continuous, quantitative estimate of autism symptom severity that is relatively independent from participant characteristics such as age and verbal language and that allows for comparison across different modules of the ADOS (Gotham et al., 2009). Table 1 depicts mean language scores, nonverbal mental age, IQ, ADOS severity, and chronological age of the participants in each diagnostic group.

All experimental testing was completed by trained examiners. ADOS administrations were videotaped and later coded from video. A research assistant, who was trained by and reliable with an independent ADOS trainer, coded 82 of the administrations. A second research assistant, who was formally trained through the ADOS research training course, and who was reliable with the other coder at the 80% agreement threshold, coded the remaining 12 videos.

Genetic Variables for Children with Fragile X Syndrome

Several measures of *FMR1*-related variation were examined in relationship to narrative performance among those children for whom molecular data were available. These included a the number of CGG expansion repeats in the 5' region of the *FMR1* gene, percentage of methylation of the *FMR1* gene, and the percentage of lymphocytes producing the gene's protein, FMRP, which has been shown to be associated with the cognitive and language impairments observed in Fragile X syndrome (Bailey, Hatton, Tassone, et al., 2001; Dyer-Friedman et al., 2002; Loesch, Huggins, & Hagerman, 2004; Losh et al., 2012). Table 2 depicts the group means for these genetic variables.

Narratives—Narratives were elicited using the popular children's storybook, *A Bed Full of Cats* (Keller, 2003), which was adapted into a wordless picture book. The book, which is about a boy searching for his lost pet cat, was chosen because of its developmental appropriateness and because it offers the narrator several opportunities to describe cognitive and affective states and behaviors. For example, in one scene the boy is sitting on the floor surrounded by his toys, sad because he cannot find his cat. Some of the pictures in the book were modified slightly to add visual interest and offer additional opportunities for description (e.g., a picture of a spider was superimposed under a table where the boy is searching for his cat).

Children were instructed to look at the book as the examiner slowly turned the pages. If a child became distracted, the examiner would prompt his attention back to the pictures. After looking through the book once, children were then instructed to tell the story as the child and the examiner slowly flipped through the book. Examiners provided nonspecific prompts such as "What happened next?" or "And then…?" if children were struggling to narrate the story. Only one prompt was provided for each 2-page spread.

Narratives were videotaped and audiotaped and then transcribed using the Systematic Analysis of Language Transcripts (SALT; Miller & Chapman, 2008) software. Transcribers, who were blind to the group status of all participants, were trained to 80% reliability on morpheme-level transcription. Transcription reliability was later assessed for 24 (25.5%) transcripts. Files were randomly selected from each of the groups, resulting in five transcripts from the autism, Fragile X syndrome with autism, Fragile X syndrome without autism, and typically developing groups, and four transcripts from the Down syndrome group checked for reliability. Mean morpheme-level agreement was 86% (range = 78%–92%).

Microstructure

Microstructure variables included mean length of utterance (MLU), narrative length, and complex syntax total and diversity. MLU was defined as the number of morphemes in each utterance and was calculated using SALT. Narrative length was quantified as the number of propositions used to construct the narrative. As in previous narrative studies (e.g., Capps et al., 2000), a "proposition" was defined as a verb and its arguments, which together form a semantically related unit of speech (Sato, 1988). For example, "The cat is sleeping" would be coded as one proposition whereas "The cat is sleeping and the boy is playing" would constitute two propositions.

Complex syntax was defined as the combination of at least two simple clauses into a single utterance contour. The types of complex syntax included coordinate clauses ("The boy looked under but only found a spider"), verb complements ("He tried to find it"), adverbial clauses ("When he woke up, there was a cat on his bed"), relative clauses ("He was a little sad of his little kitten that got lost"), and passive constructions ("The cat was cuddled by the

boy"). The frequency of complex syntax was calculated, along with the number of different types of complex syntax employed (i.e., syntactic diversity, ranging from 0 to 5).

Macrostructure

At the macrostructure level, evaluation, story structure, and thematic maintenance were assessed. The evaluative coding scheme, originally developed by Reilly, Klima, and Bellugi (1990) and subsequently used in several studies of narrative abilities in children with autism (e.g., Capps et al., 2000; Losh & Capps, 2003) and other developmental disabilities (Losh et al., 2001; Reilly et al., 1998; Reilly, Losh, Bellugi, & Wulfeck, 2004), included several difference types of evaluation (e.g., causality, emotion and cognition, intensifiers and attention getters, and storytelling devices). Definitions and examples of the types of evaluation examined can be found in the Appendix. The number of times evaluation was used was tallied, along with the number of different types of evaluation employed (i.e., evaluative diversity, ranging from 0 to 10).

As a measure of story structure, the total number of main episodes described was tallied. Main episodes of the story's plot were identified a priori, based on pilot work with typically developing adults to identify the gold standard narrative structure of the story. The story included 11 main episodes that depicted a boy's adventures searching for his missing pet cat, resulting in the boy reuniting with the cat and her litter of kittens.

Additionally, the initiation and maintenance of the central theme (i.e., searching for the cat) and mention of the story resolution (i.e., the cat returns home with her kittens) were coded. A total thematic maintenance score was created by tallying one point for initiation of the theme, one point for each time the theme was mentioned, and one point for story resolution.

Inappropriate, Irrelevant, and Unintelligible Utterances

Language features that detracted from narrative quality, such as inappropriate/irrelevant, off-topic, and unintelligible utterances were coded, and the frequencies of these features were tallied.

Reliability

All coding was conducted blind to group status by the first author. A second coder (the fourth author, a graduate student studying speech-language pathology) who was also blind to group status coded 22.7% ($n = 23$) of the narratives for reliability. Files were randomly selected from each of the groups, resulting in four transcripts being second-coded for the autism, Fragile X syndrome with autism, and Down syndrome groups, and five transcripts being second-coded for the Fragile X syndrome without autism and typically developing groups. Intraclass correlations were run, and agreement on all but one variable ranged from .91 to .99. Agreement on the total number of inappropriate/irrelevant utterances was .52. Because there were relatively few instances of inappropriate/irrelevant utterances, we examined rater agreement dichotomously (i.e., whether or not instances of such utterances occurred), rather than by utterance counts, and found that for 21 out of 23 reliability files (91.3% of cases), coders agreed on whether or not the participant included any inappropriate or irrelevant utterances. All disagreements were discussed and resolved.

Analysis Plan

For both microstructure and macrostructure comparisons, one-way multivariate analyses of variance (MANOVAs) were performed to investigate group differences. To examine the effect of nonverbal mental age on narrative performance, multivariate analyses of covariance (MANCOVAs) were then employed, with group entered as the independent

variable and nonverbal mental age entered as a covariate. The interaction between group and nonverbal mental age was also included in the models.

For analyses of narrative microstructure, four dependent variables were included: MLU, number of propositions, complex syntax total, and complex syntax diversity. For the macrostructure comparisons, four dependent variables were included: evaluation total, evaluation diversity, number of main episodes, and thematic maintenance total.

The distributions of the three different types of off-topic or irrelevant remarks were highly positively skewed, because the majority of children either displayed these behaviors infrequently or not at all. Transformations were not successful in normalizing the data. Therefore, chi-square analyses were employed as a nonparametric alternative to investigate group differences on these variables.

Finally, in exploratory analyses, Pearson correlations were employed to examine whether genetic variables correlated with language level, ADOS severity scores, and narrative features in children with Fragile X syndrome. To investigate the potentially mediating role of nonverbal IQ, the same relationships were reanalyzed as partial correlations, with nonverbal IQ entered as the control variable.

Results

The MANOVA revealed no main effects of group on narrative microstructure, $F(16, 263.37) = 1.64$, $p = .06$, Wilks' lambda = .75. Similarly, no main effect of group emerged on narrative macrostructure, $F(16, 263.37) = 1.32$, $p = .18$; Wilks' lambda = .79. When nonverbal mental age was entered as a covariate, a main effect of nonverbal mental age emerged for narrative macrostructure, $F(4, 81) = 7.13$, $p < .001$, but there was no significant group effect. For narrative microstructure, however, main effects emerged for both group, $F(16, 248.10) = 2.20$, $p < .01$; Wilks' lambda = .67, and nonverbal mental age, $F(4, 81) = 3.24$, $p < .05$; Wilks' lambda = .86. A significant interaction between group and nonverbal mental age also emerged, $F(16, 248.10) = 2.37$, $p < .01$; Wilks' lambda = .65. Tests of between-subjects effects showed that this interaction was significant for the use of complex syntax, $F(4, 84) = 3.23$, $p < .05$. Figure 1 illustrates this interaction. Groups did not differ on MLU or the length of narratives, defined by the number of propositions.

Examination of the slopes of the regression lines for complex syntax total indicate that the β values for the autism and Down syndrome groups were significantly larger than that of the typically developing group, $t(84) = 3.20$, $p < .01$; $t(84) = 2.23$, $p < .05$, respectively. No other group differences emerged for β values. To determine at what levels of nonverbal mental age the groups differed, estimated marginal means were compared at the following levels of the covariate (i.e., nonverbal mental age): 3 years, 4 years, 5 years, 6 years, 7 years, and 8 years. Results indicated that at the nonverbal mental age of 3 and 4 years, the estimated marginal means of the autism group were significantly lower than those of the typically developing children, $t(84) = -3.22$, $p < .05$; $t(84) = -3.00$, $p < .05$, respectively. However, the estimated marginal means of the autism group increased with nonverbal mental age, while the means of the typically developing group remained relatively stable. At the mental age of 8 years, the difference between the autism group and the typically developing group approached significance, $t(84) = 2.65$, $p = .09$.

To further explore the use of different types of complex syntax, one-way analyses of covariance (ANCOVA) were used to compare types of complex syntactic constructions across groups, with nonverbal mental age included as a covariate. For coordinate clauses, main effects of group, $F(4, 84) = 3.82$, and mental age, $F(1, 84) = 8.20$, as well as an interaction, $F(4, 84) = 3.86$ emerged, ps < .01 (see Figure 2). Again, β values were

compared, and estimated marginal means were examined at various levels of the covariate (i.e., nonverbal mental age). These analyses indicated several group differences. The autism group demonstrated a steeper slope than the typically developing group, $t(84) = 2.64$, $p < .01$, and the Down syndrome group demonstrated a steeper slope than both the typically developing children and those with Fragile X syndrome without comorbid autism, $t(84) = 3.44$, $p < .01$; $t(84) = 2.33$, $p < .05$, respectively. Furthermore, the estimated marginal means for the autism and Down syndrome groups were significantly lower than that of the typically developing children at the nonverbal mental age of 3 years, $t(84) = -2.92$, $p < .05$; $t(84) = -3.14$, $p < .05$, respectively. At the nonverbal mental ages of 7 and 8 years, the estimated marginal means of the Down syndrome group were significantly higher than those of the typically developing group, $t(84) = 3.00$, $p < .05$; $t(84) = 3.25$, $p < .05$, respectively.

For frequency of relative clauses, a main effect of group, $F(4, 84) = 3.21$, $p < .05$, and an interaction between group and nonverbal mental age, $F(4, 84) = 3.84$, $p < .01$, emerged (see Figure 3). Examination of the β values showed that the slope of the autism group was significantly steeper than the slopes of all other groups (all ts > 2.03, ps < .05). Pairwise comparisons of the estimated marginal means revealed that the means for the children with autism were significantly lower than those of the children with Fragile X syndrome without autism at 3 years, $t(84) = -2.88$, $p < .05$, but higher than the children with Fragile X syndrome without autism at 6, 7, and 8 years (ts > 3.05, ps <.05), and higher than the children with Down syndrome at 7 and 8 years (ts > 3.14, ps < .05). The percentage of children who used relative clauses varied by group, with 35% of the children with autism, 13% of the children with Fragile X syndrome and autism, 16.7% of the children with Fragile X syndrome without autism, 17.6% of the children with Down syndrome, and 6.3% of the typically developing children including relative clauses in their narratives However, chi-square analyses did not reveal significant group differences in the percentage of children using relative clauses ($\chi^2 = 5.80$, $p > .05$).

Only a main effect of nonverbal mental age emerged for adverbial clauses, $F(1, 84) = 9.98$, $p < .01$, although the effect of group and the interaction effect approached significance (ps < .09). No main or interaction effects emerged for frequency of verb complements. No child in the study used a passive clause when constructing his narrative; therefore, frequency of passive clauses was not analyzed. Chi-square analyses were used to examine differences between diagnostic groups on presence of inappropriate/irrelevant utterances, off-topic utterances, and unintelligible utterances. No significant differences emerged across diagnostic groups for any of these variables, χ^2s < 2.74, ps > .05.

Pearson correlations revealed several significant relationships between the number of CGG repeats and linguistic variables. Specifically, CGG repeat length was correlated with receptive vocabulary age ($r = -.46$), narrative length ($r = -.47$), evaluation total ($r = -.51$), and complex syntax total ($r = .45$), all ps < .05. Methylation percentage and percentage of lymphocytes producing FMRP were not correlated with any other language or narrative characteristics. However, when nonverbal IQ was entered as a control variable, no correlations remained significant.

Discussion

This study compared narrative abilities in children with autism, Fragile X syndrome with and without autism, Down syndrome, and typical development. Previous studies have shown that children with idiopathic autism and children with Fragile X syndrome demonstrate impairments in narrative microstructure (e.g., MLU, complex syntax) as well as narrative macrostructure (e.g., evaluation, thematic maintenance; Capps et al., 2000; Diehl et al., 2006; Estigarribia et al., 2011; Finestack & Abbeduto, 2010; Finestack et al., 2012; Keller-

Bell & Abbeduto, 2007; Losh & Capps, 2003; Tager-Flusberg, 1995; Tager-Flusberg & Sullivan, 1995). However, little is understood about the degree to which narrative impairments overlap between the two disorders. For the current study, it was predicted that children with autism and children with Fragile X syndrome with autism would demonstrate qualitatively similar narrative impairments, whereas narratives produced by children with Fragile X syndrome without comorbid autism would more closely resemble those produced by children with Down syndrome.

Comparisons of narrative microstructure revealed no clear group differences in narrative length, MLU, and syntactic complexity. However, for complex syntax use, nonverbal cognitive abilities and diagnosis interacted to produce divergent patterns across the diagnostic groups. Interestingly, children with autism and children with Down syndrome demonstrated similar patterns, with steep increases in complex syntax use as nonverbal mental age increased, driven by the use of coordinate and relative clauses. The trajectory of coordinate clause use followed a pattern similar to that of overall complex syntax use in both groups; however, the relationship between nonverbal mental age and the use of relative clauses was less straightforward. In the children with autism, frequency of relative clauses increased sharply as nonverbal mental age increased, whereas in other groups it increased more gradually (as was the case for the typically developing children and the children with Fragile X syndrome and comorbid autism) or even decreased (as was the case for the children with Down syndrome and children with Fragile X syndrome without autism).

Relative clause use was relatively infrequent in all but the idiopathic autism group, where 35% of the children with autism included relative clauses in their narratives (versus an average of 13.5% of the children in the other groups). However, even those children who did use relative clauses did so sparingly, and the maximum number of relative clauses included in any narrative was two. Furthermore, qualitative review of the transcripts that included relative clauses did not expose any striking patterns of use in the autism group (e.g., formulaic/perseverative use of relative clauses). Thus, although these results are unexpected, they do not appear to be driven by any outlying data points or striking anomalies in the children with autism.

In some respects, these findings differ from those of previous studies, which have reported that children with autism and Fragile X syndrome use less complex syntax (Capps et al., 2000; Finestack & Abbeduto, 2010), and children with autism produce fewer propositions (Capps et al., 2000) relative to typically developing controls. However, the findings of the present study are in line with previous reports that MLU in children with autism and Fragile X syndrome does not differ from that of typically developing children in the context of narrative production (Finestack & Abbeduto, 2010; Keller-Bell & Abbeduto, 2007).

The results pertaining to narrative macrostructure were also unexpected. When carefully matched on expressive and receptive vocabulary abilities, and when similar on nonverbal mental age, narratives produced by these cognitively young groups of children look very much alike at the macrostructure level. These findings were surprising, given previous reports of pervasive difficulties with narrative macrostructure in children with idiopathic autism (Capps et al., 2000; Losh & Capps, 2003, 2006). However, earlier studies often included older children, adolescents, or adults, and many of the participants were diagnosed with high-functioning autism or Asperger syndrome. In the current study, the majority of the children in the diagnostic groups had intellectual disability with an average nonverbal mental age of approximately 5 years. Interestingly, nonverbal mental age emerged as a strong predictor of narrative macrostructure across groups. Thus, in this sample of children, it appears that nonverbal mental age, as opposed to diagnostic status, is an important determinant of macrostructure-level performance. However, it is important to consider that,

although the developmental disability groups demonstrated no significant narrative impairments relative to younger typically developing children, their narratives would most certainly differ from those produced by chronological age-matched controls.

Several correlations emerged with *FMR1*-related variation in the Fragile X syndrome group. However, consistent with prior research (Loesch et al., 2007; McDuffie et al., 2010), relationships were found to be attenuated when controlling for mental age. This suggests that although variation in the *FMR1* gene may influence social communication, this relationship is complexly interrelated with general cognitive ability in Fragile X syndrome. Future work should examine such relationships in higher functioning groups in which more precise relationships between narrative abilities and other social communicative skills, underlying genetics, and protein variation might be delineated.

Some limitations should be considered when interpreting these results. First, as noted above, it is possible that the children selected for inclusion in this study were too impaired for meaningful differences in narrative ability to be observed using the storybook elicitation procedure and coding scheme used. Also, narratives were assessed and compared at only one time point, and therefore this study does not provide insight into the developmental trajectories of narrative ability in these populations. Given the cross-sectional nature of this study, our findings of increased use of complex syntax in a subgroup of cognitively older children must be interpreted cautiously. Future studies should follow children with autism and Fragile X syndrome longitudinally to examine patterns of narrative competence throughout development.

Also, as this study did not include female children with autism and Fragile X syndrome, the interactive effects of gender and diagnosis on narrative ability remain unknown. Previous studies have found that girls with Fragile X syndrome tend to be higher functioning than their male counterparts (Clifford et al., 2007; de Vries et al., 1996; Keysor & Mazzocco, 2002; Loesch et al., 2003; Loesch et al., 2002), because of the protective effect of their second (unaffected) X chromosome. However, the gender differences in autism are not quite so straightforward, with some studies finding phenotypic differences between males and females (Bolte, Duketis, Poustka, & Holtmann, 2011; Holtmann, Bolte, & Poutska, 2007; McLennan, Lord, & Schopler, 1993) and others reporting no such differences (Lord, Schopler, & Revicki, 1982; Tsai & Beisler, 1983; Volkmar, Szatmari, & Sparrow, 1993). Studying narrative abilities in females with autism and Fragile X syndrome will be an important step toward delineating the effects of gender on pragmatic language abilities in these disorders.

Finally, as noted, it will be important to compare children's narrative abilities across different discourse contexts, including conversation and narratives elicited using different types of stimuli. Particular patterns of strengths and weaknesses have been documented across contexts in prior studies of autism (Losh & Capps, 2003), and it will be important to determine whether similar patterns are observed in Fragile X syndrome with and without autism.

In sum, these findings suggest that at younger developmental levels these groups did not demonstrate striking differences in narrative performance and further suggest that mental age, as opposed to diagnosis, was a critical predictor of narrative ability at such young cognitive developmental levels. To our knowledge, this is the first study to compare directly the narrative abilities of children with idiopathic autism to those of children with Fragile X syndrome with and without comorbid autism. This type of cross-population comparison is critical to elucidate the phenotypic expression of autism in Fragile X syndrome. Results may also have implications for the identification and treatment of pragmatic language difficulties

in autism and Fragile X syndrome. From a clinical and educational standpoint, results suggested that narrative abilities should be assessed in all children with developmental disabilities, regardless of diagnosis. Narrative storytelling is a critical aspect of social interaction, as it is through narrative practices that children make sense of their world and communicate their experiences with others (Bamberg & Reilly, 1996; Berman & Slobin, 1994). Impairments in narrative ability, therefore, can greatly affect a child's social interactions. As such, clinicians and educators should consider narrative and pragmatic language impairments in assessment and treatment plans for children with developmental disabilities.

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Appendix

Evaluation Coding Scheme

Variable	Fragile X syndrome with autism	Fragile X syndrome without autism
Causality. Causal statements include those in which the narrator presents the cause or motivation of events or behaviors ("He looked everywhere to find his cat").		
Emotion and cognition. References to the characters' internal states were tallied. This category includes descriptions of characters' affective states ("He is sad") and behaviors ("The boy cried") as well as cognitive states ("He know that") and behaviors ("His Mama saw the cats"). In addition, statements that included causal explanations of internal states ("And so he cried about the cat") were coded as such.		
Negatives. Negative propositions, such as "Cat's not there," communicate the narrator's perspective by indicating that events or behaviors belie underlying expectations.		
Hedges. Hedges ("Maybe the mom knows") are employed to communicate narrator uncertainty, thus inferring multiple possible interpretations or perspectives of an event.		
Character speech, onomatopoeia, and sound effects. These devices capture and direct the listener's attention and assume the perspective of the story's characters through the use of *character speech* ("And the boy said, 'goodnight Kitty'"), *onomatopoeia* ("Meow"), and *sound effects* ("Plop").		
Intensifiers and attention getters. Intensifiers such as *emphatic markers* ("He was so happy"), and *repetition* ("And he look look look everywhere for his kitty"), and *attention getters* ("Hey look, there's a spider") point to the significance of an event by drawing the listener's attention to a specific behavior or episode.		
Storytelling devices. These devices (e.g., "Once upon a time," "The end") help the narrator to frame the events and episodes as one related story unit and also infer the narrator's understanding of the cultural expectations of storytelling.		

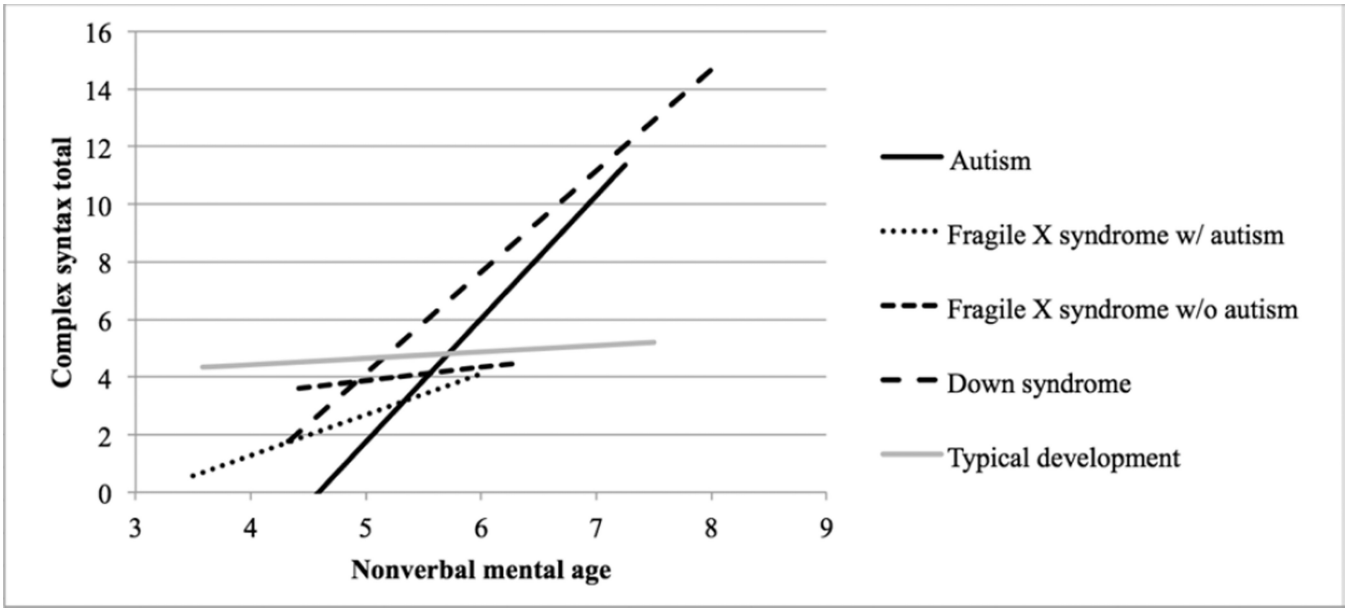

Figure 1.
Relationship between nonverbal mental age and complex syntax.

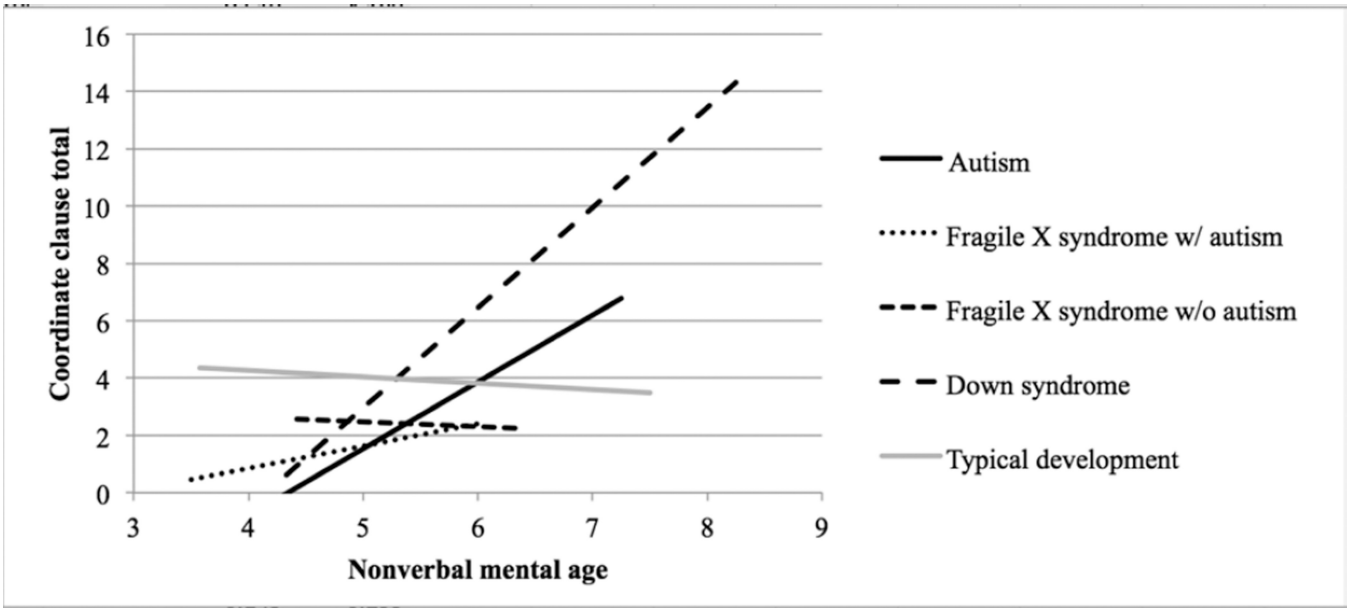

Figure 2.
Relationship between nonverbal mental age and use of coordinate clauses.
Note. Lines differ in length due to different nonverbal mental age ranges across groups.

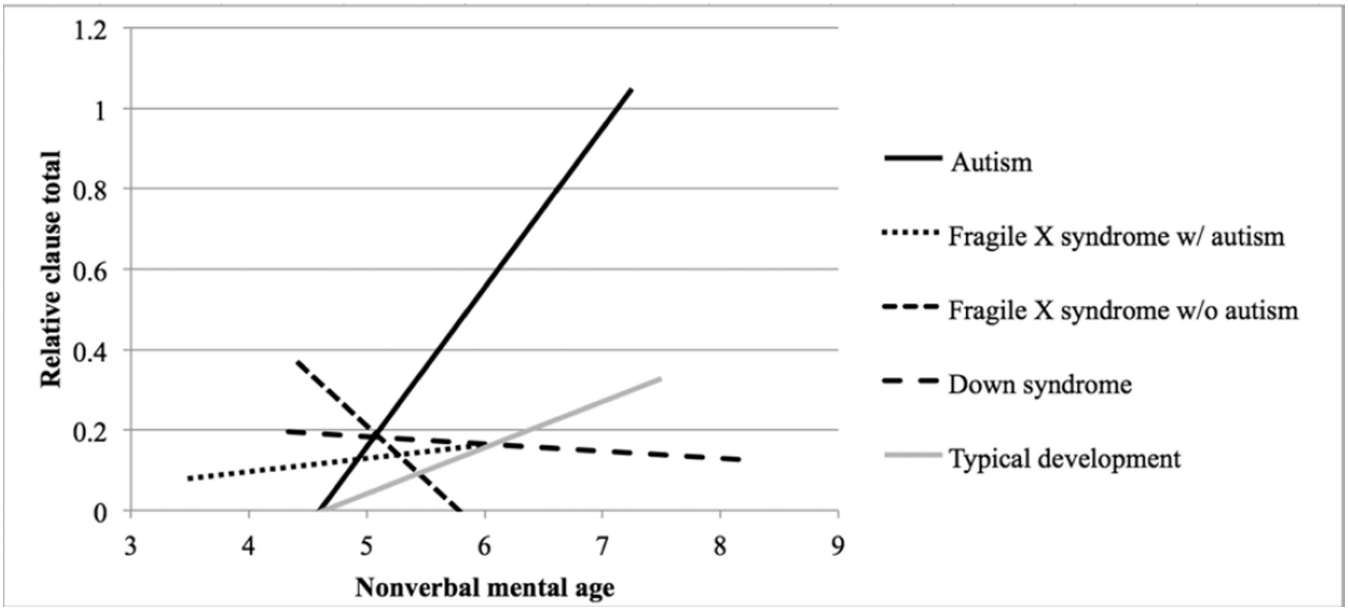

Figure 3.
Relationship between nonverbal mental age and use of relative clauses.
Note. Lines differ in length due to different nonverbal mental age ranges across groups.

Table 1

Participant Characteristics

Variable	Autism *n* = 20	Fragile X syndrome with autism *n* = 23	Fragile X syndrome without autism *n* = 18	Down syndrome *n* = 17	Typical development *n* = 16
	***M* (*SD*) Range**	***M* (*SD*) Range**	***M* (*SD*) Range**	***M* (*SD*) Range**	***M* (*SD*) Range**
EVT/PPVT composite score†	124.70 (35.71) 79–215	123.96 (29.44) 60–171	128.56 (27.32) 82–180	124.94 (29.98) 78–170	128.75 (31.42) 60–181
Expressive vocabulary age†	5.39 (1.44) 3.58–8.92	4.92 (1.06) 2.67–7.08	5.18 (0.97) 3.83–6.91	5.67 (1.60) 3.58–8.58	5.35 (1.38) 2.92–7.75
Receptive vocabulary age†	5.48 (1.70) 3.17–10.00	5.72 (1.62) 2.42–8.83	5.88 (1.37) 3.42–8.67	5.26 (1.46) 2.42–7.50	5.74 (1.47) 2.17–8.17
Nonverbal mental age†	5.74 (1.00) 4.08–7.25	5.03 (0.57) 3.5–6.00	5.16 (0.55) 4.42–6.33	5.38 (0.90) 4.33–8.25	5.18 (1.16) 3.58–7.50
Nonverbal IQ	69.65_{a} (15.37) 40–102	54.04_{b} (11.80) 40 – 79	$58.89_{a,b}$ (14.58) 38–89	53.12_{b} (9.96) 38–73	114.37_{c} (11.87) 100–139
ADOS severity	7.55_{a} (1.96) 4–10	6.61_{a} (1.44) 4–9	2.11_{b} (0.90) 1–3	1.41_{b} (0.80) 1–3	1.44_{b} (0.63) 1–3
Chronological age	9.04_{a} (2.29) 4.15–12.73	10.28_{a} (2.62) 6.58–15.07	9.73_{a} (2.50) 6.06–14.98	11.01_{a} (2.18) 6.81–14.86	4.55_{b} (0.92) 3.21–6.15

†Means do not differ significantly (ps > .05).

Note. EVT = Expressive Vocabulary Test; PPVT = Peabody Picture Vocabulary Test—Third Edition; ADOS = Autism Diagnostic Observation Schedule. Means in the same row with different subscripts differ at $p < .05$, Bonferroni adjusted.

Table 2

Genetic Characteristics of Children with Fragile X Syndrome

Variable	Fragile X syndrome with autism	Fragile X syndrome without autism
	M (*SD*) Range *n*	
CGG Repeats[a]	842.38 (333.36)	551.58 (255.62)
	201–1363	201–863
	13	12
Percentage of methylation	94.83 (8.55)	87.78 (27.66)
	76–100	18.4–100
	16	9
Percentage of lymphocytes producing FMRP	12.00 (10.6)	8.63 (7.93)
	1.00–38.00	1.00–25.00
	12	12

[a]Means differ at $p < .05$.