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# Expressive Language in Male Adolescents with Fragile X Syndrome with and without Comorbid Autism

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## Abstract

**Background—**Approximately one-quarter of individuals with fragile X syndrome(FXS) meet diagnostic criteria for autism; however, it is unclear whether individuals with comorbid FXS and autism are simply more severely affected than their peers with only FXS or whether they have qualitatively different profiles of behavioral impairments. To address this issue, variation in the FXS linguistic phenotype was examined in males with FXS with and without autism. The syndrome - specificity of the expressive language impairment of both groups of those with FXS was assessed in relation to Down syndrome. The extent to which different language sampling contexts affected expressive language in each diagnostic group was also examined.

**Method—**Spontaneous language samples were collected from male adolescents with FXS without autism ($n$ = 20), comorbid FXS and autism ($n$ = 8), and Down syndrome ($n$ = 16). Syntactic complexity (indexed by MLU), expressive vocabulary (indexed by lexical diversity), talkativeness, fluency, and intelligibility were assessed in two contexts: conversation and narration. Groups were matched on nonverbal IQ, nonverbal mental age, and chronological age to allow the assessment of relative strengths and weaknesses across language variables.

**Results—**Males with comorbid FXS and autism were less intelligible than males with only FXS; no other differences between these two groups were found. Participants' performance differed across contexts for syntactic complexity, lexical diversity, talkativeness, and fluency.

**Conclusions—**These findings contribute to existing research on the behavioral profiles of individuals with FXS or FXS with autism who have low cognitive abilities. Although individuals with comorbid FXS and autism may be, as a group, more impaired than those with only FXS, data from this small sample of males with comorbid FXS and autism with low IQs suggest that their relative strengths and weaknesses in spontaneous expressive language are largely comparable and not differentially affected by the context in which their talk occurs.

### Keywords

fragile X syndrome; Down syndrome; autism; expressive language; conversation; narrative

Fragile X syndrome (FXS) results from a mutation of the Fragile X Mental Retardation-1 (*FMR1*) gene in the form of an expansion of a CGG sequence to 200 or more repeats. The FXS phenotype is marked by cognitive functioning in the normal range for about half of females and moderate intellectual disability in most males (Keysor & Mazzocco, 2002). Along with cognitive deficits, individuals with FXS often have significant language difficulties that include syndrome-specific features and considerable within -syndrome variation (Abbeduto, Brady, &

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Kover, 2007). Autistic-like behaviors (e.g., gaze aversion, hand-flapping) are common in FXS, although the extent to which they are displayed varies widely across individuals (Feinstein & Reiss, 1998; Hagerman, 2002). Between 16% and 47% of those with FXS meet diagnostic criteria for autistic disorder (hereafter termed FXS+AUT), with a consensus estimate of 20 to 30 % (Bailey et al., 1998; Clifford et al., 2007; Demark, Feldman, & Holden, 2003; Hagerman, Jackson, Levitas, Rimland, & Braden, 1986; Hatton et al., 2006; Kau et al., 2004; Lewis et al., 2006; Rogers, Wehner, & Hagerman, 2001). In contrast to FXS, autistic disorder (AUT) is a behavioral diagnosis, defined in terms of deficits in communication and social interaction and the presence of repetitive and stereotyped behaviors (APA, 1994). Most individuals with idiopathic AUT have language impairments that continue into adolescence and adulthood, although both cognitive and linguistic abilities vary considerably in this population (Seltzer, Shattuck, Abbeduto, & Greenberg, 2004; Tager-Flusberg, Paul, & Lord, 2005). Of individuals with AUT, it is estimated that between 2% and 6% have FXS (Hagerman, 2006). In this study, we focused on understanding the linguistic phenotype associated with the comorbidity of these two disorders.

The question of how to interpret the co-occurrence of FXS and AUT has received increasing attention and generated considerable controversy. A better understanding of this issue necessitates (a) fully describing the variable FXS phenotype, with particular attention to autistic behaviors and their role as predictors of developmental outcomes, and (b) reconciling the case of FXS with our current conceptualization of AUT (Bailey et al., 2004). Examining the comorbidity of FXS and AUT thus provides a gateway through which to understand both FXS and AUT, lay the groundwork for effective interventions, and ultimately connect genetic mechanisms with behavioral outcomes for these neurodevelopmental disorders (Belmonte & Bourgeron, 2006).

Two overarching issues characterize the controversy surrounding the association between FXS and AUT (Lewis et al., 2006; c.f. Bailey et al., 2004). First, it is unclear whether the occurrence of AUT in individuals with FXS represents simply the most highly affected end of the FXS continuum or a distinct behavioral phenotype that is qualitatively different from that characteristic of individuals with FXS without AUT (hereafter, “FXS” will refer to FXS without a diagnosis of AUT). If individuals with FXS+AUT are simply on the severe end of the fragile X syndrome continuum (i.e., have increased impairments and more, or more severe, AUT symptoms), the co-occurrence of FXS and AUT should be correlated with cognitive ability (Bailey et al., 2004). In fact, there is evidence that individuals with FXS+AUT tend to have lower IQs than those with FXS (e.g., Philofsky, Hepburn, Hayes, Hagerman, & Rogers, 2004). In contrast, differing profiles of cognitive or linguistic strengths and weaknesses would suggest a qualitative distinction between FXS and FXS+AUT. Evidence for this could be gleaned, for example, from comparisons of individuals with FXS and FXS+AUT who are matched on nonverbal cognitive ability, but perform differently on measures of language. In this vein, Lewis et al. (2006) found receptive language to be a relative weakness in adolescent males with FXS+AUT compared to those with FXS, matched on nonverbal cognition. In this case, FXS+AUT would be associated with a specific linguistic profile and could be considered a distinct subgroup, qualitatively different from those with FXS.

Second, there is some doubt that a diagnosis of AUT in individuals with FXS is “true” AUT (Cohen et al., 1991; Fisch, 1993). It has been claimed, for instance, that autistic-like behaviors reflect anxiety-based social avoidance in FXS, but social indifference in idiopathic AUT (Budimirovic et al., 2006; Feinstein & Reiss, 1998). However, similarity in cognitive or linguistic profiles in FXS+AUT and idiopathic AUT would suggest that AUT in FXS is not distinguished from AUT of other etiologies on the basis of underlying psychological mechanisms (Bailey et al., 2004). Thus, the comparison of FXS+AUT to the typical AUT profile will be invaluable for understanding the association between FXS and AUT.

The purpose of the present study was to expand upon existing evidence of the relationship between FXS and AUT and to provide a better understanding of the variability associated with the FXS phenotype. Because language difficulties are hallmark symptoms of individuals with FXS+AUT but have yet to be completely characterized, the linguistic profile associated with FXS+AUT was the focus. In particular, we compared the expressive language abilities of adolescents with FXS+AUT to those of adolescents with FXS matched on nonverbal cognitive ability. To assist in interpreting the findings, individuals with Down syndrome (DS) were included as a comparison group, allowing us to differentiate expressive language deficits specific to FXS or FXS+AUT from those associated with general cognitive disability. Although we did not include an idiopathic AUT group, the question of the relation between AUT in FXS and idiopathic AUT was indirectly addressed by considering the linguistic profile of individuals with AUT already documented in the literature.

## Methodological Issues in Understanding Language Development in Fragile X Syndrome and Autism

Questions about FXS+AUT, especially concerning language development, have gone unanswered in part due to challenges and traditions in research on intellectual disabilities. First, individuals with comorbid AUT have been excluded from many studies examining FXS, particularly those addressing the linguistic phenotype (Abbeduto & Murphy, 2004). Although this practice removes the potential confound of AUT status and increases the homogeneity of the sample, it has also contributed to the paucity of published data on language abilities in individuals with FXS+AUT (Abbeduto, McDuffie, Brady, & Kover, in press). In line with Tager-Flusberg's (2004) call for within-group comparisons across the range of AUT, within-syndrome comparisons of FXS will further elucidate the extent, nature, and causes of within-syndrome variability and the possible existence of qualitatively different FXS subgroups.

Second, research on expressive language in FXS has generally underutilized spontaneous language samples. Despite the potential for extracting highly specific and clinically relevant variables, the analysis of language samples from individuals with FXS has largely been limited to gross measures of performance that ignore distinctions across domains or features of language (Abbeduto & Hagerman, 1997). Moreover, language samples have often been drawn from poorly standardized conversational contexts, even though variations in experimenter behavior or materials are known to affect child language output (Dollaghan, Campbell, & Tomlin, 1990). In addition, sampling contexts vary in the effectiveness with which they assess different dimensions of linguistic ability. Abbeduto, Benson, Short and Dolish (1995) found that children and adolescents with intellectual disability of mixed etiology produced more syntactically complex language in narration than conversation, whereas the reverse was true for talkativeness. This suggests that contexts may also differ in their utility for uncovering differences among diagnostic groups (Chapman, Seung, Schwartz, & Bird, 1998; Sudhalter, Maranion, & Brooks, 1992). Furthermore, there is evidence that diagnostic groups can be distinguished by differences in their expressive language profiles across contexts (Miles, Chapman, & Sindberg, 2006). Thus, differences in syntactic complexity between FXS, FXS +AUT, and DS might only be detectable in contexts that elicit more complexity than others. The present study, therefore, was designed to describe spontaneous language in individuals with FXS+AUT using variables relevant to specific dimensions of expressive language (e.g., syntactic complexity, lexical diversity), based on two sampling contexts characterized by well standardized procedures; namely, conversation and narration. It was of interest whether the effect of diagnostic group on expressive language would differ between sampling contexts given their characteristics.

Third, cross-syndrome comparisons are infrequently made in studies of language development in FXS or FXS+AUT. The DS phenotype is a valuable contrast to FXS because many

individuals with FXS and DS share characteristics (e.g., cognitive, speech, and language impairments relative to chronological age), but strengths and weaknesses in expressive language across contexts may be specific to each phenotype (Abbeduto & Chapman, 2005; Roberts et al., 2007). Although the profile of abilities in DS is not "flat," (i.e., expressive language abilities tend to be impaired beyond cognitive level-expectations), a comparison to DS can help distinguish aspects of the linguistic phenotype of FXS associated with the diagnosis rather than simply the presence of intellectual disability. Thus, including adolescents with DS in the current study added the benefit of identifying the syndrome specificity of the profiles of the FXS and FXS+AUT subgroups. Both within-and cross -syndrome comparisons are key to understanding the association between FXS and AUT.

## Linguistic Profiles in Fragile X Syndrome, Autism, and Down Syndrome

### Fragile X syndrome

Delays in cognition and language in individuals with FXS (without comorbid AUT) increase relative to typically developing peers during late childhood and adolescence due to a generally slowed rate of development (Murphy & Abbeduto, 2003). For adolescents with FXS, receptive vocabulary and receptive syntax seem to keep pace with cognitive abilities (Abbeduto & Hagerman, 1997). In line with this, Abbeduto et al. (2003) found that male and female adolescents with FXS displayed relatively synchronized performance across nonverbal cognition and the receptive vocabulary and receptive syntax subtests of the *Test for Auditory Comprehension of Language-Revised* (TACL; Carrow-Woolfolk, 1985).

Expressive language in FXS is often characterized by rapid rate, poor intelligibility, and perseveration on words, sentences, and topics (Belser & Sudhalter, 2001; Ferrier, Bashir, Meryash, & Johnston, 1991; Madison, George, & Moeschler, 1986; Paul et al., 1984; Sudhalter, Cohen, Silverman, & Wolf-Schein, 1990). Vocabulary and syntax in the expressive domain may be commensurate with nonverbal mental age (NVMA), although the evidence is not consistent on this point (Madison, George, & Moeschler, 1986; Paul et al., 1987; Sudhalter et al., 1992). For instance, some research has pointed to a specific expressive syntax delay based on mean length of utterance(MLU; Paul et al., 1984; Paul et al., 1987; Sudhalter, Scarborough, & Cohen, 1991)or an uneven profile with relative strengths and weaknesses across different syntactic abilities in boys with FXS (Levy, Gottesman, Borochowitz, Frydman, & Sagi, 2006). In contrast, Madison et al. (1986) concluded that MLU was consistent with cognitive ability levels in males with FXS. More recently, Roberts et al. (2007) compared MLU and number of different words in language samples collected during the Autism Diagnostic Observation Schedule (ADOS; Lord, Rutter, DiLavore, & Risi, 2001) from 35 boys with FXS between the ages of 2 and 14 years. On average, the boys with FXS produced language with lower MLU and fewer different words than NVMA-matched typically developing boys, reflecting an expressive language deficit beyond NVMA and extending to multiple domains of language. These studies are challenging to compare, however, because of the use of inadequately described or standardized contexts across participants in some cases (Abbeduto & Hagerman, 1997).

In adolescent males with FXS, receptive vocabulary and syntax seem to be on par with nonverbal cognitive abilities. It is less clear whether this is true of expressive vocabulary and syntax. Clarifying this picture will require collecting spontaneous language samples in multiple, standardized contexts.

### Autism

The profile of language development in individuals with AUT includes impairments in receptive and expressive language relative to chronological age expectations, especially among

those with lower IQs (Tager-Flusberg, 2001). On average, receptive language is often impaired beyond developmental-level expectations and even expressive language (Gillum & Camarata, 2004; Paul & Sutherland, 2005). There are areas of substantial impairment in expressive language as well, however, including echolalia (Tager-Flusberg & Calkins, 1990), delays and unusual errors in the use of deictic terms, such as personal pronouns (Lee, Hobson, & Chiat, 1994), and difficulty with the acquisition of mental state terms (Baron-Cohen et al., 1994). When measured with standardized tests, expressive vocabulary can be a relative strength, but individuals with AUT do not always display their abilities in a normative way (e.g., they produce stereotyped phrases, use words with idiosyncratic meanings) in spontaneous language production (Tager-Flusberg et al., 2005).

A handful of studies on children with AUT have pointed to a relative deficit in syntax. Kjelgaard and Tager-Flusberg (2001) divided children with AUT into three groups based on language ability. The children in the lowest performing language groups showed relative difficulty with syntax as opposed to vocabulary. In the same language-impaired sample of children with AUT, morphosyntactic production (e.g., tense marking) was an area of particular impairment (Roberts, Rice, & Tager-Flusberg, 2004). Along these lines, Landa and Goldberg (2005) found relative impairment in sentence formulation by high-functioning children and adolescents with AUT compared to IQ-matched typically developing children.

In summary, there are differences between the linguistic profiles of individuals with AUT and those with FXS. Whereas receptive language might be a relative deficit in AUT, receptive language abilities in individuals with FXS tend to be similar to nonverbal cognitive abilities. The evidence for a relative deficit in expressive syntax for a subgroup of individuals with AUT suggests this is an important domain for comparison with FXS.

## Fragile X syndrome and comorbid autism

The few studies addressing language abilities in individuals with FXS+AUT have generally employed summary scores that fail to distinguish particular dimensions of language to the degree necessary for creating a profile of abilities. Nonetheless, a general finding is that children with FXS+AUT have especially impaired receptive language relative to comparison groups. For example, Philofsky et al. (2004) suggested that poor receptive language may be a clinical marker for AUT in individuals with FXS; however, nonverbal cognition and expressive language were also depressed in the FXS+AUT group relative to both FXS and AUT comparison groups in this study, making the findings difficult to interpret. Examining domain-specific aspects of receptive language using the TACL, Lewis et al. (2006) found that receptive language skills in adolescents with FXS+AUT were more impaired than in their NVMA- and age-matched peers with FXS, with similar performance across vocabulary, grammatical morphology, and syntax.

Findings on expressive language abilities in FXS+AUT relative to FXS are less clear. Some investigators have reported particularly impaired expressive language in FXS+AUT (Roberts, Mirrett, & Burchinal, 2001; Rogers et al., 2001). In contrast, spontaneous syntactic complexity in conversational language samples was not found to be different between boys with FXS and boys with FXS and a co morbid autism spectrum disorder (FXS+ASD; i.e., not distinguishing between those with AUT or an ASD), after controlling for NVMA and maternal education (Price et al., 2008). Likewise, adolescent males with FXS+AUT, based on the *Oral Expression Scales* of the *Oral and Written Language Scales* (Carrow -Woolfolk, 1995), were not significantly different from NVMA-matched adolescents with FXS (Lewis et al., 2006). In the Lewis et al. study, however, expressive language was examined in a global manner, with no distinction among different dimensions of language.

In summary, a relative deficit in receptive language in FXS+AUT compared to FXS has been identified in a few studies. For expressive language, previous studies have yielded inconsistent results across different periods of development, measurement strategies, and aspects of language, highlighting the importance of assessing multiple aspects of expressive language skills in FXS+AUT, using multiple well-defined contexts over a well -defined age range.

### Down syndrome

Widely studied as the leading genetic cause of intellectual disability, DS results from a triplicate of chromosome 21 and the over-expression of its genes, although there does not appear to be a critical set of genes(Abbeduto & McDuffie, in press; Chapman & Hesketh, 2000). Compared to FXS, it is thought that the prevalence of AUT in individuals with DS is lower, estimated at 5 to 10% (Capone, Grados, Kaufmann, Bernad-Ripoll, & Jewell, 2005; Kent, Evans, Paul, & Sharp, 1999). Individuals with DS have a linguistic profile characterized by delays beyond nonverbal cognitive deficits, with expressive language more impaired than receptive language (Chapman, 2003; Chapman, Schwartz, & Kay-Raining Bird, 1991). There is also evidence of a specific syntactic deficit (Abbeduto et al., 2003; Chapman et al. 1991), which is especially pronounced in the expressive domain (Chapman et al., 1998; Laws& Bishop, 2003). High rates of unintelligible speech characterize the expressive language of individuals with DS (Abbeduto & Chapman, 2005; Kumin, 2004) and struggles with fluency are anecdotally noted to be particularly problematic as individuals with DS age into adolescence (Kumin, 1994). Disfluency in expressive language can reflect problems with planning and utterance-formulation in children with language disorders, which are to be expected in the face of severe limitations in expressive syntax, phonology, or other aspects of language (Miller, Leddy, & Leavitt, 1999). In the current study, areas of specific impairment for individuals with DS (e.g., intelligibility) as well as areas of relative strength (e.g., vocabulary) were included among the variables of interest. For each expressive language variable, differential performance between either of the FXS groups and a NVMA -matched DS comparison would suggest syndrome-specific linguistic profiles.

## Summary

In the current study, spontaneous expressive language was examined in male adolescents with FXS+AUT through a comparison with cognitive level-matched groups of adolescent males with FXS or DS. It was reasoned that differences in the profile of relative strengths and weaknesses of expressive language abilities between FXS and FXS+AUT would be consistent with claims of qualitative differences between them. We focused on expressive language variables thought to be particularly impaired in idiopathic AUT and DS so as to be maximally informative as to the interpretation of the linguistic profile of FXS+AUT and its syndrome specificity. By assessing expressive language in two distinct sampling contexts, conversation and narration, the extent to which effects of contexts differed among groups was considered.

Two sets of hypotheses were the focus of this study. First, we expected participants with FXS and FXS+AUT to perform as well as or better than their peers with DS on most measures of expressive language (i.e., syntactic complexity, disfluency, and unintelligibility), but not all (i.e., lexical diversity, talkativeness); we also expected participants with FXS+AUT to demonstrate less syntactic complexity compared to their peers with FXS. Second, it was thought that syntactic complexity would be greater in narration than conversation, whereas talkativeness would be greater in conversation than narration (Abbeduto, Benson, Short, & Dolish, 1995). More generally, the effect of context was expected to vary across measures of language and diagnostic groups.

# Method

## Participants

Participants for the present study were males drawn from those tested in a larger program of research on the language and communication skills of adolescents and young adults with FXS or DS (references deleted for anonymous review). Participants were only recruited into the larger study if the parent reported that their son could use at least a three -word phrase to describe a picture. Individuals with FXS were recruited nationally through postings to Internet listservs, advertisements in newspapers in large urban areas of the United States, nationwide newsletters and websites of organizations on developmental disabilities, and through a university registry of families interested in research participation. Individuals with DS were recruited locally for the most part through newspaper advertisements, mailings to special educators, and a university research registry. The participants in the FXS+AUT group were included in the sample studied by (reference deleted for anonymous review) and many participants with FXS or DS were included in other previous reports (references deleted for anonymous review). Although several participating families in the larger project contained multiple siblings with FXS, only one sibling pair was included in the present analyses (one sibling each in the FXS and FXS+AUT group). The parent or legal guardian of each participant provided informed written consent. This research was reviewed and approved by the (deleted for anonymous review) IRB.

Reports of DNA analyses confirming the diagnosis of full mutation were available for all but two participants: only cytogenetic results were available for one male with FXS and the blood test results were not available for an additional participant in the FXS group, but parent report and medical records verified the diagnosis. Six participants with FXS or FXS+AUT were mosaic (i.e., had both the full mutation and the premutation). For most participants with DS, parents reported the etiology of the disability to be trisomy 21 and in all but four cases, confirmatory reports of the karyotype analysis were obtained. One participant with DS had a translocation.

The hearing of each participant was evaluated at the time of participation and no participant was included if he had more than a mild hearing loss, defined as an average pure tone threshold of 30 dB or worse in the better ear over the “speech frequencies” of 500 Hz, 1000 Hz, and 2000 Hz. Pure tone threshold results concurrent with the measures of interest, however, were unavailable for one participant in the FXS+AUT group who was noncompliant during the hearing evaluation. The parent reported no history of hearing loss, and audiological screenings from visits in subsequent years supported this. Given the high rate of hearing problems in DS, the groups significantly differed on average pure tone threshold (i.e., thresholds averaged across both ears and across all three speech frequencies), $F(2, 40) = 5.57$, $p = .007$, with the participants with DS having significantly higher average thresholds than participants in the FXS and FXS+AUT groups, $t(34) = 3.19$, $p =. 003$ and $t (21) =2. 21$, $p = .033$, respectively. Average threshold did not differ statistically between the FXS and FXS+AUT groups, $t(25) = .16$, $p = .87$.

Participants with FXS or DS were identified as having AUT for the purpose of this study by way of a multi-step process, which at the time the data were collected reflected the practices of the clinic at our research center. Participants were first screened for behavioral manifestations of autism symptoms using the Autism Behavior Checklist (ABC; Krug, Arick, &Almond, 1980). This informant-completed screener of 57 items was filled out by each participant’s teacher, mother, and, in the case of two-parent households, father. Higher scores on the ABC indicate greater presence of autistic-like behaviors and a cut-off score of 44 was set to flag the possibility of an appropriate AUT diagnosis (Volkmar et al., 1998). Receiving this or a higher score from at least two of three informants for participants in two-parent

households or from at least one of two informants for single-parent households led to a further clinical evaluation. Correlations among ratings across informants were positive and moderate to large in size (see Table 1).

Participants who met the screening criteria on the ABC were referred to a licensed clinical psychologist, with many years of experience in evaluating and diagnosing AUT. Her clinical assessment was composed of three parts: observations of the participant interacting with his parent(s) for about 10 minutes, direct interaction with the participant for about 10 minutes, and an interview with the parent(s) to establish a development al history. The observations and interviews were structured to allow evaluation against DSM-IV (APA, 1994) criteria for autistic disorder (see reference deleted for anonymous review for details). Based on this evaluation, the psychologist assigned a diagnosis of AUT when appropriate; categorization of participants for the present study was based on her diagnosis. Diagnoses of PDD -NOS were not considered.

Only participants who met criteria for AUT in a strict sense (i.e., autistic disorder) were included in the FXS+AUT group. All other participants were assigned to the FXS group, including those who might be considered to be on the autism spectrum (e.g., PPD-NOS) but failed to meet criteria for AUT as specified for the current study. This approach to defining the groups might have led to a conservative analysis of the impact of AUT symptomology on expressive language, as the FXS group could include individuals with high, but sub-diagnostic-threshold, symptom levels.

The method of AUT diagnosis used here, based on direct observation of, and interaction with, the participant in addition to a structured parent interview based on DSM-IV criteria, yielded group assignments that are in line with the rate of diagnoses made with other methods. In the larger project, ten male participants out of a total of 54 males and females with FXS met diagnostic criteria for an AUT diagnosis (FXS+AUT). A single participant with DS from the larger study met criteria for AUT and was subsequently excluded from all analyses. The rate of AUT in this sample of individuals with FXS (approximately 19%) is in the range of other published reports (e.g., Bailey et al., 1998; Hatton et al., 2006; Kaufmann et al., 2004).

Furthermore, differentiating males with FXS and FXS+AUT with this method has yielded data in our previous research suggesting that that receptive language and theory of mind are areas of weakness for males with FXS+AUT compared to those with FXS (reference deleted for anonymous review). These results are in line with what is known about the behavioral profile of individuals with idiopathic AUT, thereby providing further support for this method of AUT diagnosis in males with FXS. Although not utilizing the current gold-standard measures of AUT diagnosis, such as the ADOS, we believe these expert clinical diagnoses yielded valid diagnostic decisions.

One of the participants with FXS+AUT from the larger project failed to engage in a conversation with the examiner and another failed to complete the narrative task (i.e., produced only utterances that were not relevant to the narrative materials). As such, these two participants with FXS+AUT were excluded from the present study because they failed to complete one of the two expressive language activities that are the main focus of this research. One participant with DS similarly failed to complete the narrative task in a meaningful way and was thus excluded.

Participants with FXS, FXS+AUT, and DS were selected from the pool available from the larger project on the basis of nonverbal IQ (NVIQ), and then matched group-wise on NVMA and chronological age. Given that expressive language ability was the cognitive domain of interest, groups were matched on nonverbal cognition as opposed to a measure of language ability in order to assess relative strengths and weaknesses across multiple language variables

without confounding differences in intellectual ability. The two measures of nonverbal cognitive ability (NVIQ and NVMA) were based on performance on three subtests of the Stanford-Binet, 4th edition, described below (Thorndike, Hagen, & Sattler, 1986). For the Stanford-Binet subtests, all participants with FXS+AUT received a standard score of 36, which is the lowest possible standard score for this measure. However, a standard score of 36 does not preclude differing raw scores or NVMAs, which ranged from 3.19 to 4.25 for participants with FXS+AUT (*M*= 3.85). Participants with FXS and DS were selected such that they had all received standard scores of 36 on the Stanford-Binet also. This led to the inclusion of 20 of the participants with FXS (*M*= 3.74, range = 2.86 to 4.39), 16 of the participants with DS(*M*= 3. 78, range = 2.56 to 4.94), and all eight of the participants with FXS+AUT who completed the tasks of interest. The resulting groups did not differ based on separate one-way ANOVAs with respect to NVMA, $F(2, 41) = .15$, $p = .86$, or chronological age, $F(2, 41) = .21$, $p = .82$ (see Table 2), and met the criterion for being well-matched proposed by Mervis & Robinson (1999).

In addition to the matching variables described above, potential differences on relevant demographic variables between groups were investigated in a pair-wise fashion with Fisher's exact tests for the distribution of race (Caucasian vs. non-Caucasian) and level of maternal education (college degree attained vs. no college degree), using the Holm (1979) method to control Type I error rate. There were no significant differences for either race or level of maternal education, *p*s > .10 and *p*s > .67, respectively (see Table 2). There was also no difference among groups in family income level (measured with a rating scale based on ten-thousand dollar intervals), $F(2, 39) = .12$, $p = .89$.

In line with the procedures used to establish the groups, the average ABC rating of each participant's three informants (or the two available informants)differed significantly among groups, $F(2, 41) = 28.11$, $p <. 0005$. Based on directional pair-wise contrasts, participants with FXS+AUT (*M*= 58.35) showed more autistic-like symptoms than participants with FXS (*M*= 35.37), $t(26)= 3.89$, $p = < .0005$, and participants with DS(*M*= 13. 53), $t(22) = 7.32$, $p <. 0005$. Participants with FXS were rated higher on the ABC than those with DS, $t(34) = 4.61$, $p <. 0005$.

## Materials and Procedure

**Measurement of nonverbal cognition—**Nonverbal cognitive ability was assessed using three subtests from the Stanford-Binet, 4th edition (Thorndike et al., 1986). Both NVMA and NVIQ were estimated based on participants' performance on Bead Memory, Pattern Analysis, and Copying. These subtests assess, respectively, visuospatial abilities relating to skills in short-term memory, analyzing patterns, and reproducing patterns. For each of the subtests, responses are nonverbal actions, and verbal instructions during administration are minimal. Following the procedure outlined by Chapman et al. (1991), NVMA was calculated as the mean of the age-equivalents for the three subtests. NVIQ was calculated using the standard score of each subtest according to the partial composite procedure described by Thorndike et al., and as indicated previously, all participants received the lowest NVIQ possible.

**Measurement of expressive language abilities—**Expressive language abilities were assessed through a conversation activity and a narration activity, each designed to elicit spontaneous speech, based on procedures described by Abbeduto et al. (1995). During the conversation, each participant took part in an interview-style conversation with a single examiner with a target of 10 to 12 minutes of interaction. The activity was introduced by saying that the examiner and participant would sit and talk for about 10 minutes to get to know each other a little better. The examiner attempted to use open -ended prompts (e.g., "Tell me everything you did in school yesterday,") and to limit her own speech, thereby eliciting as

much talk from the participant as possible. Reasonable standardization was ensured across examiners and participants by the use of a script of topics (e.g., school, after-school activities) and of follow-up questions and prompts (e.g., "What's your favorite part of school? Tell me all about that."). The examiner followed the same order of topics for each participant, with the amount of time spent on a particular topic varying based on participant interest. Due to logistical issues and variation in participants' compliance, three participants with FXS talked for just over nine minutes, two participants with FXS+AUT and three with DS talked for just over eight minutes, and one participant with FXS had a conversation just over seven minutes long.

In the narration activity, participants were shown a wordless picture book, *Frog Goes to Dinner* (Mayer, 1974). The examiner introduced the book and turned from one page to the next after approximately 10 seconds of viewing, allowing the participant to look through the entire story before giving a narrative. The participant was then asked to tell the story to the examiner and to say what everyone is doing, thinking, and feeling on each page. This second time through the book, the experimenter did not turn the page until five seconds after the participant had finished narrating the page. To ensure standardization, the experimenter was restricted to scripted prompts if the participant did not respond to the first page (e.g. "What is everyone doing, thinking, and feeling in this part of the story?") and to more limited prompts for a lack of response on subsequent pages (i.e., "What's happening in this part of the story?"). The mean duration for the FXS and FXS+AUT groups was just over four minutes and was about six and a half minutes for the DS group. Although the duration of narratives differed significantly among the groups, $F(2, 41) = 9.48$, $p < .0005$, with the participants with DS taking more time to complete the narrative than either the FXS participants, $t(34) = 4.15$, $p < .0005$, or the participants with FXS+AUT, $t(22) = 2.99$, $p = .005$, the total number of C-units (defined below) produced during the narrative did not differ statistically among the groups, $F(2, 41) = .02$, $p = .98$. Narrative time for participants with FXS and FXS+AUT did not significantly differ, $t(26) = .23$, $p = .82$.

Descriptive statistics for the total number of C-units produced by participants in each group are presented in Table 3. Although some participants produced fewer C-units than others and not every participant achieved 50-C-unit samples in both contexts, we favored a standardized method of data collection rather than utilizing disproportionately intense strategies across participants to collect a predetermined number of C-units. On average, the number of total C-units did not significantly differ among groups in conversation $F(2, 41) = 1.20$, $p = .31$, or in narration, as described above. The number of complete and intelligible C-units produced also did not differ statistically among groups for conversation, $F(2, 41) = 2.19$, $p = .13$, or narration, $F(2,41) = .17$, $p = .85$. Ideally, each participant would contribute a large language sample from each context. However, the amount of language produced by an adolescent with FXS, FXS +AUT, or DS in a conversation or narration is an interesting source of information in and of itself.

Conversations and narrations were audio -tape recorded and later transcribed and analyzed using the Systematic Analysis of Language Transcripts (SALT; Miller & Chapman, 1999). This computer program supports the transcription of standard English speech samples according to child language research conventions. After initial transcription, an independent transcriber listened to the audio-tape while marking the transcript with suggested changes or perceived discrepancies. The original transcriber listened to the audiotape again and accepted or rejected the suggestions before data analysis. For standardization purposes, the first 10 minutes of each participant's conversation(or the entire conversation if less than 10 minutes) and the entire narrative were transcribed for analysis.

A small percentage (≈ 10%) of narrative and conversation audio recordings from the larger program of research was independently transcribed again. Inter-rater agreement, averaged over

contexts and groups, had an overall rate of over 90% across aspects of transcription that could affect the variables of interest: utterance segmentation, judgment of the presence or absence of an unintelligible segment, number of morphemes, number of words, presence of mazes, judgment of whether an utterance is complete, presence of a maze, and word identity. For participants with DS, agreement averaged over those dimensions of transcription was 91%, with the highest agreement for number of morphemes and utterance completion (99%) and the lowest agreement for word identity (71%). For participants with FXS, agreement averaged 92%, with the highest agreement also for number of morphemes and utterance completion (99%) and the lowest agreement for word identity (80%).

**Expressive language variables—**All speech was segmented into communication units (C-units), with a C-unit defined as an independent clause and any of its modifiers, including dependent clauses (Loban, 1976). Analyzing C-units rather than "utterances" avoids over estimating syntactic complexity in cases where long utterances are achieved by combining simple clauses with coordinating conjunctions (e.g., "He likes school and he rides bikes and he has recess and he likes TV. "). Because of the standardization of the activities, all C-units were included in analyses with no attempt to distinguish between spontaneous and prompted speech.

Five dependent variables were created, each reflecting a specific aspect of expressive language ability. These variables were derived with the purpose of gaining a broad characterization across the various dimensions of language (for further descriptions see references deleted for anonymous review). Syntactic complexity was indexed by the mean length of C-unit in number of morphemes, a general measure of syntactic ability. This variable was based only on complete and intelligible C-units. A higher score reflects mastery of greater syntactic complexity. Lexical diversity was defined as the number of different lexical word roots in a sample of 50 complete and intelligible C-units (or the total number of complete and intelligible C-units if a particular individual did not produce 50 C-units), reflecting the size of expressive vocabulary. A higher lexical diversity score indicates the use of a greater number of different lexical items. Talkativeness was defined as the number of C -unit attempts made per minute, including incomplete C -units. This variable reflects expressive productivity. Disfluency was calculated as the proportion of C-units containing mazes (i.e., verbal disfluencies, such as filled pauses and partial or full repetitions). Disfluency is a measure of planning, with lower rates of disfluency reflecting better planning. Unintelligibility was defined as the proportion of C -units partially or fully unintelligible to the transcriber. A lower unintelligibility score reflects better intelligibility and better articulation abilities.

**Procedure—**Participants were tested individually on a larger battery of assessments in several sessions over one or two days. A single examiner administered the entire battery to any given participant. Sessions lasted between roughly 90 minutes and two hours, with breaks as needed. The conversation and narration tasks took place during the second session of each participant's visit. The order of these two language tasks was randomized. Of participants in the present study, 45% completed the conversation activity before the narrative activity.

## Results

The five linguistic variables of interest were analyzed with a mixed-design in order to examine possible main effects of participant group, main effects of context of the language sample, and Group × Context interactions. In both typical and atypical development, MLU and lexical diversity are thought to be intimately related to each other and are highly correlated in samples of conversation and narration (Dethorne, Johnson, & Loeb, 2005; Miller, 1991). In the current sample of 44 participants, syntactic complexity and lexical diversity were significantly correlated in both conversation, $r(42) = .93$, $p < .0005$, and narration, $r(42) = .85$, $p < .0005$.

Because of their close relationship, syntactic complexity and lexical diversity were analyzed together in a repeated-measures MANOVA. As a way of addressing hypotheses that were specific to each variable, the Holm (1979) sequential procedure was used for the univariate results following a significant omnibus multivariate finding to control Type I error. Because the three remaining variables are thought to represent at least partly distinct aspects of expressive language ability, separate 3 (Group) × 2 (Context) ANOVAs with repeated measures on the second factor were conducted for talkativeness, disfluency, and unintelligibility. To control Type I error in these analyses, Shaffer's (1986) planned post-omnibus multiple comparison procedure was used for pair-wise comparisons, in which the p-value for each of the three pair-wise contrasts is sequentially compared to the alpha assigned to the corresponding significant omnibus F-test (i.e., $\alpha = .05$). A significant interaction was followed by tests of the simple effects and Type I error was controlled using Holm (1979) for the dependent variables, followed by Shaffer's (1986) planned post-omnibus procedure for any necessary pair-wise comparisons. Of the five variables of interest, two of these, disfluency and unintelligibility, were calculated as proportions. An arcsine transformation was performed on these proportions prior to further analysis; however, the descriptive statistics in Table 4 reflect the untransformed means and standard deviations.

For syntactic complexity and lexical diversity, the MANOVA revealed no significant effect of group, Wilks' Lambda $F(4, 80) = .66$, $p = .62$, partial $\eta^2 = .03$. There was, however, a significant effect of context, Wilks' Lambda $F(2, 40) = 43.21$, $p < .0005$. The effect of context was significant for both syntactic complexity, $F(1, 41) = 24.28$, $p < .0005$, partial $\eta^2 = .37$, and for lexical diversity, $F(1, 41) = 21.84$, $p < .0005$, partial $\eta^2 = .35$. Participants displayed higher syntactic complexity in their expressive language in narration than conversation. The reverse was true for lexical diversity: participants used a wider range of vocabulary in conversation than narration. The interaction between group and context was not significant, Wilks' Lambda $F(4, 80) = 1.71$, $p = .16$.

A significant main effect of group was found for talkativeness, $F(2, 41) = 3.70$, $p = .033$, partial $\eta^2 = .15$. Pair-wise comparisons revealed that participants with FXS attempted significantly more C-units per minute than the participants with DS, $t(34) = 2.63$, $p = .012$. Participants with FXS+AUT did not differ from those with FXS, $t(26) = .27$, $p = .79$, or those with DS, $t(22) = 1.77$, $p = .08$. There was also a significant main effect of context for C-unit attempts per minute, $F(1, 41) = 18.59$, $p < .0005$, partial $\eta^2 = .31$, such that participants attempted more C-units per minute during the conversation activity than during the narration activity. The interaction reached significance for talkativeness, $F(2, 41) = 3.69$, $p = .034$, partial $\eta^2 = .15$. In the conversation context, groups did not differ, $F(2,41) = .96$, $p = .39$. In the narrative context, talkativeness differed among groups, $F(2, 41) = 5.22$, $p = .010$, such that participants with DS were less talkative than both those with FXS, $t(34) = 2.93$, $p = .005$, and those with FXS+AUT, $t(22) = 2.48$, $p = .017$. Talkativeness did not differ significantly for participants with FXS and FXS+AUT during narration, $t(26) = .22$, $p = .83$.

For disfluency, the only significant effect was of context, $F(1, 41) = 10.77$, $p = .002$, partial $\eta^2 = .21$, with participants producing proportionally more C-units with mazes during conversation than during narration. There was a significant effect of group for unintelligibility, $F(2, 41) = 3.54$, $p = .038$, partial $\eta^2 = .15$. Participants with FXS+AUT and participants with DS produced more utterances judged to be unintelligible by the transcriber than did participants with FXS, $t(26) = 2.15$, $p = .038$ and $t(34) = 2.22$, $p = .031$, respectively. Participants with FXS+AUT and DS did not differ in intelligibility, $t(22) = .35$, $p = .73$.

## Discussion

The primary goal of the present study was to determine whether qualitative differences – differences in profiles of relative strengths and weaknesses – exist between male adolescents with FXS and those with FXS+AUT, with the interpretive aid of a comparison with males with DS. In addition to characterizing the expressive language abilities of male adolescents with FXS with and without comorbid AUT, this study examined the effects of the context from which language samples were drawn, overcoming some methodological limitations of prior studies. Insight into the linguistic abilities of adolescents with these diagnoses contributes to theories on behavioral phenotypes and to the development of appropriate and, when necessary, syndrome-tailored interventions.

### Comparison of Expressive Language in Fragile X Syndrome and Down Syndrome

We predicted group differences on the expressive language variables of interest such that, regardless of AUT status, participants with FXS with or without AUT would perform as well as those with DS on lexical diversity and talkativeness but better than those with DS on syntactic complexity, disfluency, and unintelligibility. However, significant advantages over the DS group were present only for talkativeness and intelligibility. These results suggest that the spontaneous expressive language abilities of male adolescents with FXS (with or without AUT) and DS may be similar when groups are matched on NVMA, at least with respect to some broad dimensions, such as lexical diversity and disfluency. Despite the profound difficulties with expressive language experienced by individuals with DS, they showed only a significant disadvantage to males with FXS and FXS+AUT in the number of C-unit attempts per minute during narration and a higher proportion of unintelligible C-units compared to males with FXS. It is surprising that participants with FXS did not differ from participants with DS on syntactic complexity because expressive syntax is known to be a pervasive deficit and relative weakness in individuals with DS (Chapman et al., 1998). This may have been due to the fact that the current samples of participants represented only a subset of individuals with FXS or DS because of the need to match them to those with FXS+AUT. The documented expressive language advantage for FXS over DS (e.g., Keller-Bell & Abbeduto, 2007; Price et al., 2008) might be limited to the higher IQ range of these syndromes.

### Comparison of Expressive Language in Fragile X Syndrome with and without Autism

It was hypothesized that the profiles of language abilities associated with FXS and FXS+AUT would be distinguished from each other, with syntactic complexity being an area of relative weakness for those with comorbid AUT. However, this difference in profiles was not found. Unlike some individuals with idiopathic AUT who have syntactic impairments beyond cognitive level expectations, individuals with FXS+AUT, as a group, may have a profile of expressive language abilities more similar to individuals with FXS (Kjelgaard & Tager - Flusberg, 2001; Landa & Goldberg, 2005). Syntactic complexity may be comparable in NVMA-matched adolescents with FXS, FXS+AUT, and DS. Alternatively, group differences may not have been found for syntactic complexity because of the small sample sizes. Further research with an idiopathic AUT comparison is warranted.

In the present study, adolescents with FXS+AUT were not distinguished from their NVMA-matched peers with FXS on four of the five expressive language measures (syntactic complexity, lexical diversity, talkativeness, and disfluency). Consequently, the current study fails to provide strong evidence for a qualitative difference in profiles for the areas of expressive language examined for FXS and FXS+AUT. Other investigations of expressive language in individuals with FXS+AUT that included multiple comparison groups have sometimes revealed quantitative and sometimes qualitative differences between FXS+AUT and FXS or idiopathic AUT (Philofsky et al., 2004; Price et al., 2008; Roberts et al., 2001; Rogers et al.,

2001). The inconsistent results could be due to generally small sample sizes, differing age ranges, and variability in measures, accompanied by the wide individual variability in each syndrome. Nevertheless, the notion that adolescents with FXS or FXS+AUT do not differ qualitatively in the expressive language domain is in agreement with some recent studies. Lewis et al. (2006) also failed to find differences in expressive language between NVMA-matched adolescents with FXS or FXS+AUT. The Lewis et al. study did reveal relative weaknesses in receptive language and theory of mind in FXS+AUT compared to FXS (i.e., a qualitatively different profile of abilities). Accordingly, the idea that individuals with FXS +AUT may be a distinct subgroup with an accompanying profile of strengths and weaknesses should not be discounted.

The elevated rate of unintelligibility in participants with FXS+AUT relative to FXS found here could be related to oral-motor difficulties, in line with findings of delayed or impaired oral-motor skills in children with AUT (Gernsbacher, Sauer, Geye, Schweigert, & Goldsmith, 2008). Articulation errors of adolescents and adults with AUT- even those who are high-functioning- are increased relative to typically developing individuals (Shriberg, Paul, McSweeny, Klin, Cohen, & Volkmar, 2001).

Alternatively, decreased intelligibility in FXS+AUT relative to FXS could be an artifact of the content of the language samples from which this measure was drawn. Participants with FXS +AUT might have been challenged in meeting the pragmatic demands of the language sampling tasks, thereby contributing more noncontingent, tangential talk than those with FXS. This could have limited the extent to which intelligibility was aided by the immediate context of the talk surrounding the participants' utterances compared to the participants with FXS. Roberts et al. (2007) recently addressed the issue of noncontingent talk in young males with FXS+ASD using conversational language samples taken from the ADOS and found that the FXS+ASD group was distinguished from the FXS group on the basis of increased rates of semantically noncontingent conversational turns. These conversational turns might be harder for a listener to interpret than turns that are contingent on the discourse or relevant to the narrative.

Barnes and colleagues (2009) examined intelligibility in 3 -to 15 -year-old boys with FXS during language samples collected in the context of the ADOS. They failed to find differences in phonological accuracy between those with FXS and those with FXS+ASD. In that study, the percent of intelligible words produced by the boys with FXS and FXS+ASD was significantly lower than for the TD boys, although they did not differ from the boys with DS. The results from Barnes et al. could support the argument that the difference in unintelligibility between participants with FXS+AUT and FXS in the present study may not result from true differences in phonological accuracy, but rather from content -based differences in the language samples obtained from participants. Alternatively, it is possible that the most severe problems in intelligibility within FXS are limited to those with full-blown AUT, a fact which would have been obscured by the inclusion of the full spectrum in the FXS+ASD group by Barnes and colleagues. It is also possible, given the older ages of our participants, that the problems associated with AUT symptoms become more pronounced with age. In any event, the results from the present study do suggest that intelligibility is an appropriate area of further research in boys with FXS.

It is important to keep in mind that here, as in the Lewis et al. (2006) report, the groups were matched on NVIQ and so the present study included only participants who received standard scores at the floor level on the measure of non-verbal cognitive ability administered. As such, the FXS comparison group was not a representative sample of all male adolescents with FXS, but rather only those functioning at the lower end of the FXS spectrum. Although only one weakness in the profile of expressive language was found for FXS+AUT relative to FXS in

the present study, those with FXS+AUT are more impaired, on average, than those with only FXS across multiple domains, including expressive language.

### Effects of Sampling Context on Expressive Language

Hypotheses related to contextual variations in performance were partially supported. The effects of context across participants were generally expected and in line with findings from Abbeduto et al.'s (1995) study of individuals with intellectual disability of unspecified etiology and typically developing children. As in that study, adolescents were more talkative (made more C-unit attempts per minute) in conversation than narration, but used more syntactically complex language in narration than conversation. Increased rate of talk during conversation could be due to the interactive nature of the activity, in which participants were asked open-ended and follow-up questions, encouraging participation. During the narration, they were minimally prompted and could say as much or as little as they liked for each page of the book.

More sophisticated syntactic abilities were engaged by narration than by interview -style conversation. This may be due in part to the fact that successful and complete narration of the book involves detailing a story line through a number of related episodes. This feat necessitates cause-and-effect statements with subordinate clauses and the mental states of multiple characters, which requires the use of sentences with complement clauses. In contrast, the conversation revolved around describing routine aspects of daily life rather than a well-developed series of events, which did not require, although it allowed, the use of complex syntactic structures. The conversation also lacked any visual support, whereas the picture book provided visual scaffolding throughout the narration, which might have supported syntactic complexity.

Unlike the study by Abbeduto et al. (1995), lexical diversity and rates of disfluency were significantly higher in conversation than in narration in the present study. The number of different words used by participants was greater in conversation perhaps because of the opportunity to display strengths in expressive vocabulary on a range of topics, whereas the narrative context was constrained by the content of the story book. Participants were likely more disfluent (i.e., produced more mazes, self-repetitions, and filled pauses, reflecting poor planning) during conversation due to the increased opportunity for planning in advance of speaking during narration. Participants looked through the entire picture book before beginning their narration and again while telling the story, affording time to "brainstorm" lexical items or phrases. This procedure also removed potential memory demands because the support of the storybook pictures was present throughout. In contrast, the interview-style conversation required responding to the examiner's questions and follow-up prompts, which were not introduced ahead of time. This finding is in accordance with that of Miles et al. (2006), who found that adolescents with DS and typically developing children were more disfluent in conversation than narration.

Contrary to expectation, group differences did not vary by context (i.e., Group × Context interactions were not significant) for any variables except talkativeness. Talkativeness did not differ among groups in conversation; however, participants with DS produced a particularly low rate of talk in the narrative context compared to participants with FXS or FXS+AUT. This finding is consistent with literature that suggests some aspects of narration may be especially challenging to adolescents with DS, even relative to those with FXS, in light of their expressive language deficits (Keller-Bell & Abbeduto, 2007). Despite the fact that conversation and narration varied in the ways they elicited language, in their task demands, and in the level of social interaction required, group differences on other dimensions of language did not differ by context.

## Clinical and Research Implications

The findings reported here have implications for assessment of and intervention for expressive language in differing contexts in FXS, FXS+AUT, and DS. Expressive language intervention that targets advanced syntactic constructions, vocabulary usage, frequent communication attempts, and planning across contexts would be appropriate. Adolescents with FXS+AUT were less intelligible than their peers with FXS, but did not differ significantly from participants with DS. Because DS is characterized by poor articulation and unintelligibility, speech-related processes may also be an important point of intervention for males with FXS and comorbid AUT. Likewise, increasing communication attempts may be a syndrome-specific deficit for individuals with DS worth particular attention in behavioral therapy in the face of low rates of talk– especially during narration – and difficulty with articulation. Further research on the speech and language abilities in adolescents with these syndromes will provide the foundation for relevant evidence-based practice.

As Abbeduto et al. (1995) suggested, narrative samples should be utilized to assess the limit, or upper bound, of expressive syntactic skills because they more fully engage syntactic abilities, whereas conversation samples may be optimal for attaining large samples of utterances or for assessing lexical diversity. Narratives and interview-style conversations differ not only in opportunities for complex syntax (e.g., occasions to talk about relationships among events and internal states), but also on the level of scaffolding and contextual support (Miles et al., 2006). For these reasons, assessing expressive linguistic abilities in multiple contexts is crucial to fully understanding the expressive language of individuals with intellectual disabilities.

## Limitations and Future Research

The present study was limited in several respects. First, although we did rely on expert clinical judgment, we did not diagnose AUT with the Autism Diagnostic Interview-Revised (ADI-R; Lord, Rutter, & Le Couteur, 1994) and the ADOS, which together are the gold standard diagnostic methods of AUT research. Ideally, AUT diagnosis in individuals with FXS would be based upon the combined use of ADI-R, ADOS, DSM-IV criteria, and clinical judgment. Such an approach would have the advantage of aligning with the current approach to diagnosing idiopathic AUT. It is quite challenging, however, to identify ASDs in children and adolescents with FXS. Diagnoses and estimated rates of prevalence of AUT in FXS can vary widely depending upon the chosen criteria (e.g., Clifford et al., 2007; Harris et al., 2008). Because the true prevalence of AUT in FXS is unknown, it is unclear that methods other than the ADI-R and the ADOS cannot be used to identify individuals with FXS+AUT in a reliable way or that the ADI-R and ADOS are actually appropriate for the FXS population. The method of AUT diagnosis utilized here has led to findings about adolescents with FXS+AUT in line with expectations about rates of comorbid AUT in FXS and expectations about the profile of language abilities of those with FXS+AUT as compared to what is known about idiopathic AUT (reference deleted for anonymous review). Although not ideal, the method of assessment used in the present study is useful at this point in our understanding of the comorbidity of FXS and AUT. Future research should address the relative merits of different approaches to the challenging task of diagnosing individuals with FXS+AUT.

Second, participants included only a small number of males with FXS+AUT. As such, the small sample size could have partially accounted for failure to find group differences on some of our variables and may limit the generalizability of the results to the FXS population. Results should not be generalized to females with FXS+AUT without further investigation. Furthermore, it is important to keep in mind that the FXS and DS comparison groups were not comprised of individuals drawn from the full range of functioning in the FXS or DS phenotype. Because these groups were matched on NVIQ to adolescents with FXS+AUT, participants with FXS and DS were from the lower end of the continuum of males for the syndrome.

However, only participants with phrase speech were recruited into the study. Results of the current study might have differed if participants with less well-developed expressive language capabilities or with higher cognitive abilities had been included. Thus, the results from the current study may not be generalizable to other portions of these populations. Finally, the five language variables of interest were broad in nature. Although they each addressed separate aspects of expressive language, more fine-grained measures of spontaneous expressive language abilities might bring to light areas of particular difficulty for individuals FXS, FXS +AUT, and DS (e.g., particular grammatical constructions). Levy et al. (2006), for instance, found that relying on MLU may mask variations across subdomains of syntax in boys with FXS.

Future studies on the linguistic profile of individuals with FXS+AUT should carefully address other areas of language development. Receptive language may be a key domain for exploration because previous work has revealed relative weaknesses (i.e., a qualitative difference in profile) in performance in FXS+AUT compared to NVMA-matched groups of individuals with FXS on standardized measures of receptive language (Lewis et al., 2006). Furthermore, research addressing pragmatic abilities in these populations will be important because difficulties in social interaction, particularly in conversation and other discourse tasks, are so salient in individuals with AUT (Tager-Flusberg et al., 2005). Research assessing language abilities of adolescents with FXS+AUT relative to individuals with idiopathic AUT and typical development would be particularly helpful. Studies in which within-syndrome and cross-syndrome comparisons are made (i.e., using multiple, carefully specified groups) will continue to be important for understanding the full range of variability associated with the FXS phenotype (Burack, Iarocci, Flanagan, & Bowler, 2004).

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## References

Abbeduto L, Benson G, Short K, Dolish J. Effects of sampling context on the expressive language of children and adolescents with mental retardation. Mental Retardation 1995;33(5):279–288. [PubMed: 7476250]

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, Hagerman RJ. Language and communication in fragile X syndrome. Mental Retardation and Developmental Disabilities Research Reviews 1997;3(4):313–322.

Abbeduto, L.; McDuffie, A. Genetic syndromes associated with intellectual disability. In: Armstrong, C., editor. Handbook of Medical Neuropsychology: Applications of Cognitive Neuroscience. Springer; in press

Abbeduto, L.; McDuffie, A.; Brady, N.; Kover, ST. Language development in fragile X syndrome: Syndrome-specific features, within-syndrome variation, and contributing factors. In: Burack, J.; Hodapp, R.; Zigler, E., editors. Handbook of mental retardation and development. 2. New York: Oxford University Press; in press

Abbeduto, L.; Murphy, MM. Language, social cognition, maladaptive behavior, and communication in Down syndrome and fragile X syndrome. In: Rice, ML.; Warren, SF., editors. Developmental language disorders: From phenotypes to etiologies. Lawrence: Erlbaum Associates, Publishers; 2004. p. 77-97.

Abbeduto L, Murphy MM, Cawthon SW, Richmond EK, Weissman MD, Karadottir S, et al. 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]

American Psychiatric Association. Diagnostic and Statistical Manual of Mental Disorders: DSM-IV. 4. Washington, DC: American Psychiatric Association; 1994.

Bailey DB Jr, Mesibov GB, Hatton DD, Clark RD, Roberts JE, Mayhew L. Autistic behavior in young boys with fragile X syndrome. Journal of Autism and Developmental Disorders 1998;28(6):499–508. [PubMed: 9932236]

Bailey, DB., Jr; Roberts, JE.; Hooper, SR.; Hatton, DD.; Mirrett, PL.; Roberts, JE., et al. Developmental language disorders: From phenotypes to etiologies. Lawrence: Erlbaum Associates, Publishers; 2004. Research on fragile X syndrome and autism: Implications for the study of genes, environments, and developmental language disorders; p. 121-150.

Barnes E, Roberts J, Long SH, Martin GE, Berni MC, Mandulak KC, Sideris J. Phonological accuracy and intelligibility in connected speech of boys with fragile X syndrome or Down syndrome. Journal of Speech, Language, and Hearing Research 2009;52:1048–1061.

Baron-Cohen S, Ring H, Moriarty J, Schmitz B, Costa D, Ell P. Recognition of mental state terms: Clinical findings in children with autism and a functional neuroimaging study of normal adults. British Journal of Psychiatry 1994;165(5):640–649. [PubMed: 7866679]

Belmonte MK, Bourgeron T. Fragile X syndrome and autism at the intersection of genetic and neural networks. Nature Neuroscience 2006;9(10):1221–1225.

Belser RC, Sudhalter V. Conversational characteristics of children with fragile X syndrome: Repetitive speech. American Journal on Mental Retardation 2001;106(1):28–38. [PubMed: 11246710]

Budimirovic DB, Bukelis I, Cox C, Gray RM, Tierney E, Kaufmann WE. Autism spectrum disorder in Fragile X syndrome: Differential contribution of adaptive socialization and social withdrawal. American Journal of Medical Genetics 2006;140(17):1814–1826. [PubMed: 16906564]

Burack JA, Iarocci G, Flanagan TD, Bowler DM. On mosaics and melting pots: Conceptual considerations of comparison and matching strategies. Journal of Autism and Developmental Disorders 2004;34(1):65–73. [PubMed: 15098959]

Capone GT, Grados MA, Kaufmann WE, Bernad-Ripoll S, Jewell A. Down syndrome and comorbid autism-spectrum disorder: characterization using the aberrant behavior checklist. American Journal of Medical Genetics Part A 2005;134(4):373–380. [PubMed: 15759262]

Carrow-Woolfolk, E. Test for Auditory Comprehension of Language -Revised. Austin, TX: Pro-Ed; 1985.

Chapman, RS. Language and communication in individuals with Down syndrome. In: Abbeduto, L., editor. International Review of Research in Mental Retardation. Vol. 27. New York: Academic Press; 2003. p. 1-34.

Chapman RS, Schwartz SE, Kay-Raining Bird E. Language skills of children and adolescents with Down syndrome: I. Comprehension. Journal of Speech & Hearing Research 1991;34(5):1106–1120. [PubMed: 1836243]

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.

Clifford S, Dissanayake C, Bui QM, Huggins R, Taylor AK, Loesch DZ. Autism spectrum phenotype in males and females with fragile X full mutation and permutation. Journal of Autism and Developmental Disorders 2007;37(4):738–747. [PubMed: 17031449]

Cohen IL, Sudhalter V, Pfadt A, Jenkins EC, Brown WT, Vietze PM. Why are autism and the fragile-X syndrome associated? Conceptual and methodological issues. American Journal of Human Genetics 1991;48(2):195–202. [PubMed: 1990832]

Demark JL, Feldman MA, Holden JJ. Behavioral relationship between autism and fragile X syndrome. American Journal on Mental Retardation 2003;108(5):314–326. [PubMed: 12901707]

Dethorne LS, Johnson BW, Loeb JW. A closer look at MLU: What does it really measure? Clinical Linguistics & Phonetics 2005;19(8):635–648. [PubMed: 16147407]

Dollaghan CA, Campbell TF, Tomlin R. Video narration as a language sampling context. Journal of Speech & Hearing Disorders 1990;55(3):582–590. [PubMed: 2381199]

Feinstein C, Reiss AL. Autism: The point of view from fragile X studies. Journal of Autism and Developmental Disorders 1998;28(5):393–405. [PubMed: 9813775]

Ferrier LJ, Bashir AS, Meryash DL, Johnston J. Conversational skills of individuals with fragile-X syndrome: A comparison with autism and Down syndrome. Developmental Medicine & Child Neurology 1991;33(9):776–788. [PubMed: 1834506]

Fisch GS. What is associated with the fragile X syndrome? American Journal of Medical Genetics 1993;48(2):112–121. [PubMed: 8362927]

Gernsbacher MA, Sauer EA, Geye JM, Schweigert EK, Goldsmith HH. Infant and toddler oral- and manual-motor skills predict later speech fluency in autism. The Journal of Child Psychology and Psychiatry 2008;49(1):43–50.

Gillum H, Camarata S. Importance of treatment efficacy research on language comprehension in MR/DD research. Mental Retardation and Developmental Disabilities Research Reviews 2004;10(3):201–207. [PubMed: 15611987]

Hagerman, RJ. The physical and behavioral phenotype. In: Hagerman, RJ.; Hagerman, PJ., editors. Fragile X syndrome: Diagnosis, treatment, and research. 3. Baltimore: The Johns Hopkins University Press; 2002. p. 3-109.

Hagerman RJ. Lessons from fragile X regarding neurobiology, autism, and neurodegeneration. Journal of Developmental & Behavioral Pediatrics 2006;27(1):63–74. [PubMed: 16511373]

Hagerman RJ, Jackson AW 3rd, Levitas A, Rimland B, Braden M. An analysis of autism in fifty males with the fragile X syndrome. American Journal of Medical Genetics 1986;23(1–2):359–374. [PubMed: 3953654]

Harris SW, Goodlin-Jones B, Ferranti J, Bacalman S, Barbato I, Tassone F, et al. Autism profiles of males with fragile X syndrome. American Journal on Mental Retardation 2008;113(6):427–438. [PubMed: 19127654]

Hatton DD, Sideris J, Skinner M, Mankowski J, Bailey DB Jr, Roberts J, et al. Autistic behavior in children with fragile X syndrome: Prevalence, stability, and the impact of FMRP. American Journal of Medical Genetics Part A 2006;140(17):1804–1813. [PubMed: 16700053]

Holm S. A simple sequentially rejective multiple test procedure. Scandinavian Journal of Statistics 1979;6:65–70.

Kau AS, Tierney E, Bukelis I, Stump MH, Kates WR, Trescher WH, et al. Social behavior profile in young males with fragile X syndrome: Characteristics and specificity. American Journal of Medical Genetics Part A 2004;126(1):9–17. [PubMed: 15039968]

Kaufmann WE, Cortell R, Kau AS, Bukelis I, Tierney E, Gray RM, et al. Autism spectrum disorder in fragile X syndrome: Communication, social interaction, and specific behaviors. American Journal of Medical Genetics Part A 2004;129(3):225–234. [PubMed: 15326621]

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]

Kent L, Evans J, Paul M, Sharp M. Comorbidity of autistic spectrum disorders in children with Down syndrome. Developmental Medicine and Child Neurology 1999;41(3):153–158. [PubMed: 10210247]

Keysor CS, Mazzocco MM. A developmental approach to understanding fragile X syndrome in females. Microscopy Research Technology 2002;57(3):179–186.

Kjelgaard MM, Tager-Flusberg H. An investigation of language impairment in autism: Implications of genetic subgroups. Language and Cognitive Processes 2001;16(2):287–308. [PubMed: 16703115]

Krug, DA.; Arick, JR.; Almond, PJ. Autism Screening Instrument for Educational Planning. Examiner's manual. Portland, OR: ASIEP Education; 1980.

Kumin, L. Communication skills in children with Down syndrome: A guide for parents. Rockville, MD: Woodbine House, Inc; 1994.

Kumin L. Speech intelligibility and childhood verbal apraxia in children with Down syndrome. Down Syndrome Research and Practice 2004;10(1):10–22.

Landa RJ, Goldberg MC. Language, social, and executive functions in high functioning autism: A continuum of performance. Journal of Autism and Developmental Disorders 2005;35(5):557–573. [PubMed: 16211332]

Laws G, Bishop DV. A comparison of language abilities in adolescents with Down syndrome and children with specific language impairment. Journal of Speech, Language, and Hearing Research 2003;46(6): 1324–1339.

Lee A, Hobson RP, Chiat S. I, you, me, and autism: An experimental study. Journal of Autism and Developmental Disorders 1994;24(2):155–176. [PubMed: 8040159]

Levy Y, Gottesman R, Borochowitz Z, Frydman M, Sagi M. Language in boys with fragile X syndrome. Journal of Child Language 2006;33(1):125–144. [PubMed: 16566323]

Lewis P, Abbeduto L, Murphy M, Richmond E, Giles N, Bruno L, et al. 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, Ill: National Council of Teachers of English; 1976.

Lord, C.; Rutter, M.; DiLavore, PC.; Risi, S. Autism Diagnostic Observation Schedule. Los Angeles: Western Psychological Services; 2001.

Lord C, Rutter M, Le Couteur A. Autism Diagnostic Interview--Revised: A revised version of a diagnostic interview for caregivers of individuals with possible pervasive developmental disorders. Journal of Autism and Developmental Disorders 1994;24(5):659–685. [PubMed: 7814313]

Madison LS, George C, Moeschler JB. Cognitive functioning in the fragile-X syndrome: A study of intellectual, memory and communication skills. Journal of Mental Deficiency Research 1986;30(2): 129–148. [PubMed: 3735410]

Mayer, M. Frog Goes to Dinner. NY: Dial Books; 1974.

Mervis CB, Robinson BF. Methodological issues in cross-syndrome comparisons: Matching procedures, sensitivity (*Se*), and specificity (*Sp*)Commentary on M. Sigman & E. Ruskin, Continuity and change in the social competence of children with autism, Down syndrome, and developmental delays. Monographs of the Society for Research in Child Development 1999;64:115–130. (Serial No. 256). [PubMed: 10412223]

Miles S, Chapman R, Sindberg H. Sampling context affects MLU in the language of adolescents with Down syndrome. Journal of Speech, Language, and Hearing Research 2006;49(2):325–337.

Miller, JF. Research on child language disorders: A decade of progress. Austin, Tex: Pro-Ed; 1991.

Miller, JF.; Chapman, RS. Systematic Analysis of Language Transcripts (Version 6.0). Madison, WI: Language Analysis Laboratory, University of Wisconsin; 1999.

Miller, JF.; Leddy, M.; Leavitt, LA. Improving the communication of people with Down syndrome. Baltimore, MD: Paul H. Brookes Publishing Co; 1999.

Murphy, MM.; Abbeduto, L. Language and communication in fragile X syndrome. In: Abbeduto, L., editor. International Review of Research in Mental Retardation. Vol. 27. New York: Academic Press; 2003. p. 83-119.

Paul R, Cohen DJ, Breg WR, Watson M, Herman S. Fragile X syndrome: Its relations to speech and language disorders. Journal of Speech & Hearing Disorders 1984;49(3):328–332. [PubMed: 6540336]

Paul R, Dykens E, Leckman JF, Watson M, Breg WR, Cohen DJ. A comparison of language characteristics of mentally retarded adults with fragile X syndrome and those with nonspecific mental retardation and autism. Journal of Autism and Developmental Disorders 1987;17(4):457–468. [PubMed: 3479423]

Paul, R.; Sutherland, D. Enhancing early language in children with autism spectrum disorders. In: Volkmar, FR.; Paul, R.; Klin, A.; Cohen, D., editors. Handbook of autism and pervasive developmental disorders. 3. Vol. 1. Hoboken, New Jersey: John Wiley & Sons, Inc; 2005. Diagnosis, development, neurobiology, and behavior

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 JR, Roberts JE, 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;51:3–15.

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 JA, Rice ML, Tager-Flusberg H. Tense marking in children with autism. Applied Psycholinguistics 2004;25(3):429–448.

Roberts JE, 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]

Rogers SJ, Wehner EA, Hagerman R. The behavioral phenotype in fragile X: Symptoms of autism in very young children with fragile X syndrome, idiopathic autism, and other developmental disorders. Journal of Developmental & Behavioral Pediatrics 2001;22(6):409–417. [PubMed: 11773805]

Seltzer MM, Shattuck P, Abbeduto L, Greenberg JS. Trajectory of development in adolescents and adults with autism. Mental Retardation and Developmental Disabilities Research Reviews 2004;10(4):234–247. [PubMed: 15666341]

Shaffer JP. Modified Sequentially Rejective Multiple Test Procedures. Journal of the American Statistical Association 1986;81(395):826–831.

Shriberg LD, Paul R, McSweeny JL, Klin A, Cohen DJ, Volkmar FR. Speech and prosody characteristics of adolescents and adults with high-functioning autism and Asperger syndrome. Journal of Speech, Language, and Hearing Research 2001;44:1097–1115.

Sudhalter V, Cohen IL, Silverman W, Wolf-Schein EG. Conversational analyses of males with fragile X, Down syndrome, and autism: Comparison of the emergence of deviant language. American Journal on Mental Retardation 1990;94(4):431–441. [PubMed: 2137003]

Sudhalter V, Maranion M, Brooks P. Expressive semantic deficit in the productive language of males with fragile X syndrome. American Journal of Medical Genetics 1992;43(1–2):65–71. [PubMed: 1605237]

Sudhalter V, Scarborough HS, Cohen IL. Syntactic delay and pragmatic deviance in the language of fragile X males. American Journal of Medical Genetics 1991;38(2–3):493–497. [PubMed: 2018092]

Tager-Flusberg H. Understanding the language and communicative impairments in autism. International Review of Research in Mental Retardation 2001;23:185–205.

Tager-Flusberg H. Strategies for Conducting Research on Language in Autism. Journal of Autism and Developmental Disorders 2004;34(1):75–80. [PubMed: 15098960]

Tager-Flusberg H, Calkins S. Does imitation facilitate the acquisition of grammar? Evidence from a study of autistic, Down's syndrome and normal children. Journal of Child Language 1990;17(3):591–606. [PubMed: 2148571]

Tager-Flusberg, H.; Paul, R.; Lord, C. Language and communication in autism. In: Volkmar, FR.; Paul, R.; Klin, A.; Cohen, D., editors. Handbook of autism and pervasive developmental disorders, Vol. 1: Diagnosis, development, neurobiology, and behavior. 3. New York: John Wiley & Sons, Inc; 2005. p. 335-364.

Thorndike, RL.; Hagen, EP.; Sattler, JM. Stanford-Binet Intelligence Scale. 4. Chicago: Riverside; 1986.

**Table 1**

Correlations of Autism Behavior Checklist (ABC) Scores among Informants

| Informant | 1. | 2. | 3. |
|---|---|---|---|
| 1. Mother | - | .70*[a] | .59*[b] |
| 2. Father | | - | .54*[c] |
| 3. Teacher | | | - |

[a] $N = 41$.

[b] $N = 39$.

[c] $N = 38$.

* $p < .01$, one-tailed.

**Table 2**

Participant Characteristics

| | Participant Group | | | | | |
|---|---|---|---|---|---|---|
| | FXS *n* = 20 | | FXS + AUT *n* = 8 | | DS *n* = 16 | |
| **Variable** | **M** | **SD** | **M** | **SD** | **M** | **SD** |
| Chronological age | 15.61 | (2.88) | 16.38 | (2.67) | 16.05 | (3.39) |
| NVMA[a] | 3.74 | (.44) | 3.85 | (.35) | 3.78 | (.60) |
| Family income[b] | 7.84 | (3.30) | 7.38 | (3.25) | 8.00 | (2.42) |
| No. of Caucasians | 19 | | 6 | | 16 | |
| No. of mothers with college degrees | 10 | | 3 | | 7 | |

[a] Assessed with Stanford-Binet, 4th Edition subtests: Bead Memory, Copying, and Pattern Analysis (Thorndike, Hagen, & Sattler, 1986).

[b] Self-reported bracket of family income level in tens of thousands of dollars (the bracket level “6” reflects earning $50, 000 to $60,000 and level “7” reflects earning up to $70,000).

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**Table 3**

C-units Produced During Conversation and Narration

| | Participant Group | | | | | | | | |
|---|---|---|---|---|---|---|---|---|---|
| | FXS *n* = 20 | | | FXS + AUT *n* = 8 | | | DS *n* = 16 | | |
| **C-units** | **M** | **SD** | **Range** | **M** | **SD** | **Range** | **M** | **SD** | **Range** |
| Conversation | | | | | | | | | |
| Total | 141.35 | (37.02) | 77 – 202 | 126.50 | (34.17) | 82 – 181 | 126.69 | (20.50) | 96 – 162 |
| Complete & Intelligible | 122.00 | (27.87) | 69 – 171 | 104.38 | (35.45) | 65 – 167 | 105.88 | (17.81) | 74 – 129 |
| Narration | | | | | | | | | |
| Total | 50.75 | (19.91) | 26 – 91 | 52.00 | (29.66) | 26 – 111 | 49.81 | (23.42) | 21 – 96 |
| Complete & Intelligible | 45.20 | (17.84) | 18 – 83 | 43.63 | (27.48) | 16 – 97 | 41.19 | (20.45) | 18 – 92 |

**Table 4**

Expressive Language Results by Context

| | Participant Group | | | | | | | | | | | |
|---|---|---|---|---|---|---|---|---|---|---|---|---|
| | FXS *n* = 20 | | | | FXS + AUT *n* = 8 | | | | DS *n* = 16 | | | |
| | Conversation | | Narration | | Conversation | | Narration | | Conversation | | Narration | |
| **Variable** | **M** | **SD** | **M** | **SD** | **M** | **SD** | **M** | **SD** | **M** | **SD** | **M** | **SD** |
| Syntactic complexity[a] | 4.09 | (1.55) | 4.90 | (1.72) | 3.97 | (.69) | 5.14 | (1.26) | 3.59 | (2.15) | 4.80 | (2.77) |
| Lexical diversity[b] | 89.90 | (33.60) | 67.60 | (32.75) | 87.13 | (14.55) | 59.25 | (13.35) | 76.19 | (38.25) | 70.44 | (46.48) |
| Talkativeness[c] | 14.51 | (3.60) | 12.05 | (3.52) | 13.35 | (4.27) | 12.45 | (7.89) | 13.04 | (2.14) | 7.71 | (2.90) |
| Disfluency[d] | .22 | (.13) | .15 | (.10) | .22 | (.19) | .14 | (.11) | .22 | (.13) | .20 | (.19) |
| Unintelligibility[e] | .07 | (.07) | .07 | (.09) | .15 | (.10) | .13 | (.12) | .13 | (.09) | .14 | (.13) |

[a] Mean length of C-unit in morphemes.

[b] Number of different words in 50 C-units.

[c] Number of C-unit attempts per minute.

[d] Proportion of C-units containing mazes.

[e] Proportion of C-units judged unintelligible by the transcriber.