

## Article

# Effects of Sampling Context on Spontaneous Expressive Language in Males With Fragile X Syndrome or Down Syndrome

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**Purpose:** In this study, the authors examined the impact of sampling context on multiple aspects of expressive language in male participants with fragile X syndrome in comparison to male participants with Down syndrome or typical development.

**Method:** Participants with fragile X syndrome ( $n = 27$ ), ages 10–17 years, were matched groupwise on nonverbal mental age to adolescents with Down syndrome ( $n = 15$ ) and typically developing 3- to 6-year-olds ( $n = 15$ ). Language sampling contexts were an interview-style conversation and narration of a wordless book, with scripted examiner behavior. Language was assessed in terms of amount of talk, mean length of communication unit (MLCU), lexical diversity, fluency, and intelligibility.

**Results:** Participants with fragile X syndrome had lower MLCU and lexical diversity than did participants with typical development.

Participants with Down syndrome produced yet lower MLCU. A differential effect of context among those with fragile X syndrome, Down syndrome, and typical development emerged for the number of attempts per minute, MLCU, and fluency. For participants with fragile X syndrome, autism symptom severity related to the number of utterances produced in conversation. Aspects of examiner behavior related to participant performance.

**Conclusion:** Sampling context characteristics should be considered when assessing expressive language in individuals with neurodevelopmental disabilities.

**Key Words:** language sampling, conversation, narrative, fragile X syndrome, mean length of utterance (MLU)

Of the methods used to assess expressive language in children and adolescents with intellectual disabilities, standardized tests and spontaneous language samples are the most often used (Abbeduto, Kover, & McDuffie, 2012). Although scores on standardized assessments and language samples tend to be correlated, each provides unique information (Condouris, Meyer, & Tager-Flusberg, 2003; Ukrainetz & Blomquist, 2002). Standardized measures of expressive language offer a relatively quick evaluation of performance relative

to age expectations. However, most standardized assessments yield a single summary score for expressive language ability, which precludes the possibility of identifying patterns of relative strength or weakness across domains (e.g., vocabulary, syntax), and can mask clinically meaningful differences among individuals. When assessing individuals with intellectual disabilities, standardized language tasks are also prone to floor effects (Mervis & Robinson, 2005). Spontaneous language samples avoid these limitations by providing contextualized data on specific aspects of ability and, in this way, are well suited to establishing expressive language profiles (Westerveld, Gillon, & Miller, 2004). The present study was designed to further explore the utility of language-sampling procedures for individuals with intellectual disabilities.

In using language samples to characterize any population with disabilities, it is important to recognize that the nature of the language-sampling context can affect the language produced by the child and, consequently, the conclusions reached by the researcher or clinician. In this study, we focused on the ways in which sampling context impacts the characterization of expressive

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language in individuals with specific neurodevelopmental disabilities associated with intellectual disability. In particular, we assessed language in two contexts, conversation and narration, in male adolescents with fragile X syndrome, male adolescents with Down syndrome, and boys with typical development at similar levels of nonverbal cognitive development.

## **Contexts for Sampling Spontaneous Language**

Research on children with typical development or language impairment suggests that sampling contexts vary in the extent to which they elicit the upper bound of an individual's linguistic ability (Southwood & Russell, 2004; Thordardottir, 2008; Wagner, Nettelblatt, Sahlen, & Nilholm, 2000). Interview-style conversations, for instance, elicit more utterances and utterances with higher mean length of utterance (MLU) relative to free-play contexts (Evans & Craig, 1992; Southwood & Russell, 2004); however, narrative contexts may be optimal for assessing syntactic ability because they elicit longer and more complex sentences than either conversation or free play, on average (MacLachlan & Chapman, 1988; Westerveld et al., 2004). In terms of disabilities, high-functioning children with autism have been shown to demonstrate a weakness in complex syntax relative to typically developing children during personal narratives but not when telling a story from a wordless book, demonstrating differences between contexts, even for children in the normal range of cognitive functioning (Losh & Capps, 2003).

Context effects also have been described for individuals with intellectual disabilities. Abbeduto, Benson, Short, and Dolish (1995), for example, examined the language of children and adolescents with intellectual disability of unspecified etiology and found more communication attempts per minute in a conversation than in narration, whereas language produced in narration had a higher mean length of communication unit (MLCU) than in conversation. Abbeduto et al. suggested that narration more fully engages the syntactic abilities of individuals with intellectual disability because narrative revolves around content more likely to be encoded in multiclausal utterances (e.g., cause and effect, psychological states). Likewise, the visual scaffolding provided by a wordless storybook in a narrative context can free up cognitive resources, such as working memory, which might also support the production of more complex sentences (Miles, Chapman, & Sindberg, 2006). Thus, variation in the characteristics of the sampling context can affect conclusions reached about an individual with intellectual disability. From a clinical perspective, such findings suggest the need for multiple sampling contexts and to understand the advantages and disadvantages of

each. In this study, we were interested in the possible differential effects of context on the language of individuals with intellectual disabilities.

## **Consistency Within Language-Sampling Contexts**

Difficulties with the interpretation of individual or diagnostic group differences in expressive language profiles can arise from inadequate consistency of procedures within a given context. In particular, performance can be affected by variations in materials, partners, and experimenter behavior (Dollaghan, Campbell, & Tomlin, 1990; Hansson, Nettelblatt, & Nilholm, 2000). In conversation, for example, the amount of talk and rate of questioning by a partner can influence both the child's amount of talk and MLU (Cowan, Weber, Hoddinott, & Klein, 1967; Evans & Craig, 1992; Johnston, 2001; Johnston, Miller, Curtiss, & Tallal, 1993). Thus, failure to consistently structure the sampling context and script examiner behavior across participants could result in uninterpretable differences across individuals, diagnostic groups, and studies because differences in language ability cannot be distinguished from variations attributable to the sampling context. In the present study, we created procedures and scripts for consistently structuring critical aspects of our two language-sampling contexts, thereby allowing unambiguous interpretation of differences in performance across diagnostic groups.

Although it is important to structure and script the language-sampling context, this "standardization" must be flexible enough to allow the examiner to adapt his or her behavior to the skill level and interaction style of the child. Consequently, even within the constraints of a structured procedure, variation in the amount and complexity of examiner talk can emerge as the examiner attempts to engage the individual in the language sampling task. For example, an examiner's syntactic complexity, as reflected in complex sentence use, has been found to be positively correlated with child MLU, lexical diversity, and morphosyntactic complexity in school-age children with language impairments (Dethorne & Channell, 2007). This correlation was found despite the fact that for each child, the language sample was 15 min long, elicited with a standard set of toys, and followed Leadholm and Miller's (1992) recommendations for language sampling (e.g., use of open-ended questions). Although it is difficult to determine causality in such a pattern of concurrent relationships, it is important to document them. In the present study, we used structured procedures that were highly prescriptive with respect to examiner behavior, but allowed for responsiveness and adaptation to the participant. We then evaluated the relationship between examiners' language and the language

produced by participants with fragile X syndrome, who were the focus of this study, and those with Down syndrome or typical development, as points of comparison.

## **Fragile X Syndrome**

Fragile X syndrome is the leading cause of inherited intellectual disability. It is caused by a cytosine-guanine-guanine (CGG) trinucleotide expansion beyond 200 repeats on the *FMR1* gene of the X chromosome (Verkerk et al., 1991). In addition to intellectual disabilities, approximately one third of males with fragile X syndrome meet diagnostic criteria for autism, with many of the remainder displaying symptoms of autism, including stereotyped language (Bailey et al., 1998; Hagerman, 2008; Hagerman, Jackson, Levitas, Rimland, & Braden, 1986; Rogers, Wehner, & Hagerman, 2001).

Delays in vocabulary and syntax are present in both receptive and expressive language for the majority of males with fragile X syndrome regardless of the extent of autism symptoms (Abbeduto, Brady, & Kover, 2007). Expressive language is usually delayed relative to chronological age in males with fragile X syndrome, although it may be commensurate with nonverbal cognition when assessed with standardized measures (Finestack & Abbeduto, 2010; Roberts, Price, et al., 2007). Relatively few studies, however, have thoroughly described the spontaneous expressive language abilities of individuals with fragile X syndrome.

*Spontaneous expressive language in fragile X syndrome.* Spontaneous expressive language in males with fragile X syndrome generally has been characterized as rapid, repetitive, disfluent, and unintelligible (Belser & Sudhalter, 2001; Ferrier, Bashir, Meryash, Johnston, & Wolff, 1991; Hanson, Jackson, & Hagerman, 1986; Paul, Cohen, Breg, Watson, & Herman 1984; Van Borsel, Dor, & Rondal, 2008); however, early studies have produced some conflicting results. For example, some studies of males with fragile X syndrome pointed to a specific delay in MLU relative to nonverbal mental age (e.g., Paul et al., 1984), whereas others failed to find weaknesses in MLU compared with cognitive-level expectations (e.g., Ferrier et al., 1991).

In a recent series of studies, Roberts and colleagues (Roberts, Hennon, et al., 2007; Roberts, Martin, et al., 2007; Price et al., 2008; Barnes et al., 2009) analyzed language samples collected during the Autism Diagnostic Observation Schedule (ADOS; Lord, Rutter, DiLavore, & Risi, 1999). Participants included boys between the ages of 3 and 16 years who were categorized as having either only fragile X syndrome or comorbid fragile X syndrome and autism spectrum disorder (ASD), as determined by the ADOS. In one study, boys with only fragile X syndrome were found to perform more poorly on lexical

diversity, MLU, and the Index of Productive Syntax (IPSyn; Scarborough, 1990) relative to typically developing boys after controlling for nonverbal mental age, intelligibility, and maternal education (Roberts, Hennon, et al., 2007). In a follow-up study, Price et al. (2008) found that boys with fragile X syndrome with and without comorbid ASD and boys with Down syndrome had lower MLU and IPSyn scores than typically developing boys after controlling for nonverbal mental age and maternal education.

Additional analyses by Roberts and colleagues focused on other aspects of spoken language using these same language samples. Boys with fragile X syndrome, with and without comorbid ASD, produced fewer intelligible words in connected speech than typically developing boys and did not differ from those with Down syndrome in this regard, controlling for nonverbal mental age (Barnes et al., 2009). In terms of pragmatics, boys with comorbid fragile X syndrome and ASD displayed more noncontingent talk than boys with fragile X syndrome only, Down syndrome, or typical development in utterances that were collected equally from free-play versus other ADOS activities. In addition, boys with fragile X syndrome produced more perseverative language than boys with Down syndrome or typical development regardless of ASD status (Roberts, Martin, et al., 2007).

*Limitations of previous research.* Although previous studies in which spontaneous expressive language in fragile X syndrome has been examined are informative, most have been based on small samples, poorly structured sampling contexts, or language samples collected in a single context. The studies of Roberts and colleagues, for example, included a wide age range of males and incorporated a protocol that was not designed specifically for the elicitation of language samples. Although the ADOS provides a standard set of activities, the nature of the activities, the amount of time spent on each activity, and the level of examiner prompting might vary within and across modules as well as across participants. For example, conversation and telling a story from a book are probed in Modules 2 and 3, but not Module 1; free play appears in Modules 1 and 2, but not Module 3. Due to potentially important variations in materials and examiner behavior, the ADOS may not provide the ideal sampling context from which to draw conclusions about language profiles within and across groups of individuals with neurodevelopmental disabilities, although it is considered the gold standard for assessing autism symptoms.

In addition, most research has addressed the potential impact of autism symptoms on language performance in individuals with fragile X syndrome from a categorical perspective by either (a) comparing participants with

or without a comorbid autism or ASD diagnosis (e.g., Price et al., 2008) or (b) excluding participants with comorbid autism from analyses (e.g., Roberts, Hennon, et al., 2007). Despite the contributions of this research, there is a need to move beyond dichotomizing the fragile X syndrome phenotype according to cut offs for diagnosing individuals with idiopathic autism. Such cut offs, although displaying adequate levels of sensitivity and specificity for individuals with idiopathic autism, might not be suited to characterizing variation among individuals with neurodevelopmental disabilities other than autism, particularly given that autism symptoms are present to some degree in most males with fragile X syndrome (Hall, Lightbody, Hirt, Rezvani, & Reiss, 2010; Moss & Howlin, 2009). Instead, including participants with all levels of autism symptom severity and considering the incremental impact of increased autism symptomology on language performance is likely to be more informative with respect to the full range of the phenotype in fragile X syndrome (McDuffie, Kover, Abbeduto, Lewis, & Brown, 2012; Moss & Howlin, 2009). Thus, we included all participants with fragile X syndrome in group comparisons regardless of their potential autism-diagnostic status and examined variability within fragile X syndrome by evaluating the relationship between language performance and a continuous scale of autism symptom severity derived from the ADOS.

## Down Syndrome

Down syndrome is the leading genetic cause of intellectual disability and, in most cases, results from a third copy of chromosome 21 (Chapman & Hesketh, 2000). Autism can occur in individuals with Down syndrome, but it is less common than in fragile X syndrome, with prevalence estimates of approximately 10% (Hepburn, Philofsky, Fidler, & Rogers, 2008; Kent, Evans, Paul, & Sharp, 1999). Language is significantly impaired in Down syndrome compared with nonverbal cognition, with expressive language more delayed than receptive language, and syntax delayed beyond vocabulary (Chapman & Hesketh, 2001). Individuals with Down syndrome provide a useful comparison to those with fragile X syndrome because their distinct impairments may differentially affect their ability to meet the demands of any given language-sampling task, thereby yielding information about etiological differences in the effects of sampling context on expressive language.

*Spontaneous expressive language in Down syndrome.* Research on expressive language in Down syndrome has yielded more consistent results than studies on fragile X syndrome, particularly in terms of a relative delay in syntactic complexity (Laws & Bishop, 2003).

Roberts and colleagues, as described above, found that males with Down syndrome had lower MLU and IPSyn scores when controlling for nonverbal cognition and maternal education than males with fragile X syndrome or typical development (Price et al., 2008).

Even for individuals with Down syndrome, however, language-sampling contexts can have differential effects on expressive language. Miles and colleagues (2006) assessed interview-style conversation and narrative performance in 14 adolescents with Down syndrome, 12–21 years of age, relative to 14 typically developing children who were matched on receptive language ability. Although the MLU of the typically developing children did not differ across the conversation and narration contexts, the narrative context yielded significantly higher MLU for participants with Down syndrome. In fact, MLU did not differ between participants with Down syndrome and typically developing children in the narrative context. This work illustrates the nuanced characterization of expressive language ability that can emerge from the comparison of language elicited in multiple, carefully structured language-sampling contexts. Whether similar effects of sampling context extend to other neurodevelopmental disabilities, such as fragile X syndrome, remains to be determined.

## Comparisons Between Fragile X Syndrome and Down Syndrome

Little research has compared the expressive language profiles of adolescents with fragile X syndrome and Down syndrome using structured sampling contexts designed to elicit spontaneous language, with the exception of a series of studies by Abbeduto and colleagues. Two studies examined performance only in a narrative context and demonstrated that adolescent and young adult males and females with fragile X syndrome used more grammatically correct and complex utterances than did participants with Down syndrome (Finestack & Abbeduto, 2010; Keller-Bell & Abbeduto, 2007). Examining both a conversation and a narrative context, Kover and Abbeduto (2010) reported several effects of context (e.g., greater MLCU and fluency, but poorer lexical diversity, in narration than in conversation) for older adolescents with fragile X syndrome or with Down syndrome, but lacked a comparison group of typically developing participants, making the expressive language profiles more difficult to interpret. We extend the findings of these studies in the present study by evaluating the effect of context (i.e., conversation vs. narration) on multiple aspects of expressive language ability, including the extent to which younger male adolescents with fragile X syndrome or Down syndrome successfully completed the language-sampling tasks.



## Research Aims

The present study was designed to examine the effects of language-sampling context on the expressive language profiles of males with fragile X syndrome using structured conversation and narration sampling contexts. The performance of males with fragile X syndrome was compared with that of adolescents with Down syndrome and typically developing boys at similar levels of nonverbal cognitive development to assess the possibility that sampling context differentially affects individuals with different neurodevelopmental disabilities. For participants with fragile X syndrome, we also explored the impact of autism symptom severity on performance. Finally, the relationship of examiner behavior to the language produced was examined to probe the potential benefits and limitations of these sampling contexts.

## Method

### Participants

Participants ( $N = 57$ ) were boys drawn from a large longitudinal project on language development. Youth with fragile X syndrome were recruited nationally (see McDuffie et al., 2010); those with Down syndrome or typical development were recruited primarily locally. Participants with typical development were not receiving special education services and had no significant sensory or motor impairments according to parent report. All participants were native English speakers and were reported by a parent to regularly use three-word phrases. Approximately 95% of the participants were Caucasian. Approximately 85% of typically developing boys had mothers with a college degree or higher compared with approximately 50% of participants with fragile X syndrome or Down syndrome. Although several sibling pairs participated, only one child per family was included in the analyses reported here. This sample overlaps with that of McDuffie et al. (2010) and Pierpont et al. (2011), although the primary measures differ. The project was Institutional Review Board approved.

Each participant contributed one conversation sample and one narration sample from a single time point. Exclusions of participants from the present analyses were made on the basis of incomplete conversations or narrations or level of nonverbal cognitive ability. Three participants with Down syndrome from the larger project were not included in the present sample because they did not complete one or both language-sampling tasks at any visit. One participant with fragile X syndrome from the larger project was not included in the sample because his conversation was only 5 min in duration. Participants were included in the present analyses only if

they completed the narrative task *in a meaningful way*—that is, the child produced an utterance relevant to the storybook for at least 12 of the 16 pages. Three participants with fragile X syndrome and two participants with Down syndrome were not included in the present analyses because they failed to meet this criterion. The analyzed language samples were collected during the first annual assessment for all but five participants with fragile X syndrome and three participants with Down syndrome, who failed to engage in the language-sampling tasks at previous annual visits. Because these participants had not successfully engaged in the tasks during prior visits, data for all participants reflect their first completion of the language-sampling activities. Finally, four participants with typical development and six participants with fragile X syndrome were excluded because their nonverbal mental ages were higher than would allow valid comparisons across all three groups, as described below. The foregoing exclusions resulted in the following samples for the present analyses: 10- to 17-year-olds with fragile X syndrome ( $n = 27$ ) or Down syndrome ( $n = 15$ ) and 3- to 6-year-old boys with typical development ( $n = 15$ ).

Participants with fragile X syndrome or Down syndrome had a confirmed genetic diagnosis. Of those with fragile X syndrome, all had molecular genetic test results indicating the full mutation or mosaicism (i.e., full and premutation cells;  $n = 7$ ). Karyotypes confirming trisomy 21 were available to the project for 13 of the 15 participants with Down syndrome; however, for the remaining two participants, Down syndrome was listed as the diagnosis in educational or medical reports and corroborated by parent report.

We administered the Leiter-R Brief IQ Screener (Roid & Miller, 1997), which yields both a standard score (nonverbal IQ) and an age-equivalent score (nonverbal mental age). Nonverbal mental age was obtained by averaging the subtest age equivalents associated with the earned raw scores over all completed subtests. To ensure that group comparisons were not confounded by differences in nonverbal mental age, participants were selected such that there were no significant group differences in nonverbal mental age. To achieve this groupwise matching while also maximizing sample sizes, comparisons of the participants with fragile X syndrome to those with Down syndrome or typical development were limited to participants whose nonverbal mental age scores ranged between 3.1 and 6.54, yielding the 57 participants included in the analyses described here. Participant characteristics are shown in Table 1. Participants with fragile X syndrome had Leiter Brief IQs between 36 and 62; participants with Down syndrome had Leiter Brief IQs between 36 and 54, and typically developing boys had Leiter Brief IQs between 87 and 127.

**Table 1.** Participant characteristics.

Participant group	<i>n</i>	Chronological age	Nonverbal IQ <sup>a</sup>	Nonverbal age equivalent
		<i>M (SD)</i>	<i>M (SD)</i>	<i>M (SD)</i>
Fragile X syndrome	27	12.70 (1.84)	44.15 (6.72)	5.08 (0.73)
Down syndrome	15	12.99 (2.02)	41.33 (6.03)	4.73 (0.75)
Typical development	15	4.67 (1.12)	107.80 (12.25)	4.95 (1.03)

*Note.* The sample of boys with fragile X syndrome (*n* = 27) includes participants regardless of autism symptom severity.  
<sup>a</sup>Due to a missing subtest, one participant with fragile X syndrome lacked a nonverbal IQ score.

All but one participant with fragile X syndrome received the ADOS (*n* = 26), usually at the first annual assessment. Examiners were trained to research reliability. Because variability in the Down syndrome phenotype due to autism symptomology was not the focus of the present study, individuals with Down syndrome were not screened for autism. Autism severity scores, ranging from 1 (*nonspectrum*) to 10 (*autism, highest possible severity score*), were assigned to participants with fragile X syndrome according to the chronological age, module administered, and total algorithm score from the ADOS (Gotham, Pickles, & Lord, 2009). Of the 26 participants with fragile X syndrome who received the ADOS, 10 fell in the nonspectrum range (i.e., severity scores ranging from 1 to 3), one fell in the ASD range (i.e., severity scores of 4 or 5), and 15 fell in the autism range (i.e., severity scores ranging from 6 to 10). The mean severity score was 5.81, with a standard deviation of 3.19.

### Measurement of Expressive Language Abilities

Expressive language abilities were assessed by eliciting spontaneous speech in two distinct sampling contexts, as described by Abbeduto et al. (1995): an interview-style conversation and a narration of a wordless picture book.

In the conversation, each participant talked with one of several female examiners for a target time of 10 min, during which the examiner’s goal was to keep her talk to a minimum. The examiner said that she would like to get to know the participant better and asked open-ended questions, while avoiding yes/no questions as much as possible. Consistency across participants was ensured with a scripted order of topics and follow-up questions; however, the amount of time spent on a topic was based on the participant’s interests. The topics included school, teachers, pets, and so forth, and were introduced in a broad manner, such as “Do you have any pets? Tell me about them.” Follow-up probes were also broad (e.g., “Tell me what you like about your pet”). All but two conversations elicited from participants

with fragile X syndrome and two from participants with typical development reached the 10-min target. Because of logistical constraints or participant engagement, these four conversations were nearly 8 min.

In the narration activity, participants were shown one of two wordless picture books, *Frog Goes to Dinner* (Mayer, 1974) or *Frog on His Own* (Mayer, 1973), alternating between participants in the larger study. No significant differences in performance between books was found for any of the seven dependent variables of interest described below (all *ps* > .18). In this sample of 57 participants, *Frog Goes to Dinner* was told by 59% of participants with fragile X syndrome, 53% of participants with Down syndrome, and 47% of participants with typical development. The participant was told that he would look at the book and then be asked to tell the story. The examiner turned the pages of the book one by one, allowing the participant to look at each for about 10 s on the initial viewing. The participant then was asked to tell everything about the story for each page. This time through, the experimenter turned to the next page 5 s after the participant had finished narrating a page. The examiner was restricted to scripted prompts if the participant did not respond to the first page (e.g., “What about the boy? What’s he doing, thinking, and feeling?”). If the participant did not talk on subsequent pages, the examiner’s scripted prompts were more limited (e.g., “What’s happening in this part of the story?”). Narratives ranged from 1 to 7 min for the participants with fragile X syndrome, from 3 to 8 min for participants with typical development, and from 3 to 11 min for participants with Down syndrome.

The language samples were recorded onto audio tapes and transcribed using Systematic Analysis of Language Transcripts software (Miller & Iglesias, 2006) according to research conventions. An experienced primary transcriber first transcribed each sample. A second trained transcriber then listened to the tape and marked suggested changes or perceived discrepancies on every transcript. The primary transcriber then verified or updated the transcript for each language sample before the data were analyzed. The first 10 min

of each conversation were transcribed, whereas each narrative was transcribed in its entirety. For 17 participants from the larger project, conversation and/or narration samples were independently transcribed again in the manner described to assess interrater agreement. Intertranscriber agreement including utterance segmentation and morpheme-level variables averaged 90% across contexts and groups (see Table 2).

*Expressive language variables.* All speech was segmented into *communication units* (C-units). A C-unit is defined as an independent clause and any of its modifiers, including dependent clauses (Loban, 1976). Segmenting speech into C-units, as opposed to utterances, avoids overestimating language abilities for long but simple utterances combined with coordinating conjunctions (Abbeduto et al., 1995; Scarborough, Rescorla, Tager-Flusberg, Fowler, & Sudhalter, 1991). The C-unit is the unit of analysis in the present study.

The amount of talk was assessed in terms of (a) the total number of C-units produced, (b) the number of complete and intelligible C-units produced, and (c) the number of C-units attempted per minute (including incomplete C-units). Language ability was assessed in terms of MLCU, lexical diversity, fluency, and intelligibility. *MLCU* was defined as the mean number of morphemes per complete and intelligible C-unit. *Lexical diversity* was defined as the total number of lexical word roots in the language sample, reflecting range of vocabulary use. Fluency was calculated as the proportion of C-units containing mazes, such as filled pauses and partial or full repetitions. *Intelligibility* was defined as the proportion of C-units partially or fully unintelligible during transcription.

## Results

### Comparison of Fragile X Syndrome, Down Syndrome, and Typical Development

We assessed profiles of expressive language abilities in a comparison of all participants with fragile X syndrome

( $n = 27$ ), Down syndrome ( $n = 15$ ), and typical development ( $n = 15$ ). Participants with fragile X syndrome and those with Down syndrome did not differ in terms of chronological age,  $t(40) = 0.53, p = .600$ , or nonverbal IQ,  $t(40) = 1.04, p = .305$ . The three groups did not differ in nonverbal mental age,  $F(2, 54) = 0.91, p = .410$ ; however, because the  $p \geq .50$  matching criteria suggested by Mervis and Robinson (1999) was not reached, nonverbal mental age was included as a covariate in these analyses. As such, we conducted repeated measures analyses of covariance to test the effects of context, group, and their interaction. We conducted planned comparisons following omnibus tests with Shaffer's (1986) postomnibus procedure.

*Amount of talk.* The unadjusted scores for the amount of language produced are presented in Table 3. The total number of C-units produced was higher in conversation than in narration,  $F(1, 53) = 5.44, p = .024, \eta_p^2 = .09$ . There was neither an effect of group,  $F(2, 53) = 2.09, p = .133$ , nor an interaction between context and group,  $F(2, 53) = 1.82, p = .173$ , for the number of C-units produced. The number of complete and intelligible utterances was higher in conversation than in narration, but just failed to reach significance,  $F(1, 53) = 3.88, p = .054$ . There was also no effect of group,  $F(2, 53) = 0.58, p = .562$ , or Context  $\times$  Group interaction,  $F(2, 53) = 0.50, p = .612$ , for complete and intelligible utterances.

For the number of C-units attempted per minute, the interaction between context and group was significant,  $F(2, 53) = 5.99, p = .005, \eta_p^2 = .18$ . The differential effect of context across groups was significant for fragile X syndrome versus Down syndrome,  $t(53) = 3.41, p = .001$ , and Down syndrome versus typical development,  $t(53) = 2.43, p = .018$ , but not fragile X syndrome versus typical development,  $t(53) = 0.69, p = .492$ . Participants with Down syndrome showed the largest effect of context with few C-units attempted during narration compared with conversation relative to participants with typical development and participants with fragile X syndrome. Neither the main effect of context,  $F(1, 53) = 3.37, p = .072$ , nor the main effect of group was significant,  $F(2, 53) = 2.83, p = .068$ .

*Language ability.* Results for expressive language ability are presented in Table 4. For MLCU, the main effect of context was not significant,  $F(1, 53) = 1.26, p = .267$ , but there was an effect of group with the participants with typical development outperforming those with fragile X syndrome, who in turn scored higher than those with Down syndrome,  $F(2, 53) = 17.55, p < .001, \eta_p^2 = .40$ , one-tailed  $ps < .05$ . There was also an interaction between context and group for MLCU,  $F(2, 53) = 4.20, p = .020, \eta_p^2 = .14$ . The differential effect of context across groups was significant for fragile X syndrome versus typical development,  $t(53) = 2.87, p = .006$ , and just failed to reach significance for Down syndrome

**Table 2.** Transcription reliability.

Participant group	Conversation		Narration	
	<i>n</i>	% agreement	<i>n</i>	% agreement
Fragile X syndrome	6	91	6	90
Down syndrome	5	87	5	86
Typical development	5	92	5	94

Note. Percentage (%) agreement is averaged over utterance segmentation, intelligibility, mazes, overlaps, pauses, abandonment, word identification, number of morphemes and words, and ending punctuation.

**Table 3.** Amount of talk produced in conversation and narration.

Participant group	n	Total C-units			Complete and intelligible			C-units attempted per minute		
		M	SD	Range	M	SD	Range	M	SD	Range
Conversation										
Fragile X syndrome	27	126.04	39.22	65–202	107.07	36.93	42–187	12.79	3.85	6.50–20.20
Down syndrome	15	131.73	33.17	70–174	100.87	28.23	38–143	13.17	3.32	7.00–17.40
Typical development	15	107.33	25.64	70–148	95.60	23.25	61–140	11.03	2.86	7.50–16.80
Narration										
Fragile X syndrome	27	43.56	16.89	21–76	36.41	13.09	19–60	11.32	4.27	4.30–22.14
Down syndrome	15	41.33	14.22	18–67	31.13	10.79	10–50	7.30	3.04	3.17–14.11
Typical development	15	39.87	10.07	20–56	34.93	10.41	17–52	8.65	2.61	4.48–12.81

Note. C-units = communication units.

versus typical development,  $t(53) = 1.91, p = .061$ . The effect of context on MLCU did not differ between participants with fragile X syndrome and Down syndrome,  $t(53) = 0.69, p = .496$ . Boys with typical development showed little effect of context on MLCU compared with the other groups, with those with fragile X syndrome producing higher MLCU in narration than in conversation.

For lexical diversity, range of vocabulary use differed among groups,  $F(2, 53) = 4.32, p = .018, \eta_p^2 = .14$ , with better performance by the typically developing boys than participants with fragile X syndrome or Down syndrome ( $ps < .05$ ), who did not differ ( $p = .211$ ). There was no effect of context,  $F(1, 53) = 0.10, p = .757$ , or interaction,  $F(2, 53) = 2.54, p = .089$ .

Fluency differed among groups,  $F(2, 53) = 3.90, p = .026, \eta_p^2 = .13$ , and was a strength of the participants with fragile X syndrome relative to those with Down syndrome ( $p = .008$ ). There was also an interaction between

group and context,  $F(2, 53) = 7.03, p = .002, \eta_p^2 = .21$ . The effect of context differed across all groups: fragile X syndrome versus Down syndrome,  $t(53) = 2.04, p = .046$ ; fragile X syndrome versus typical development,  $t(53) = 2.19, p = .033$ ; and Down syndrome versus typical development,  $t(53) = 3.75, p < .001$ . Participants with fragile X syndrome showed similar fluency in both contexts; participants with Down syndrome were more fluent (i.e., produced a smaller proportion of C-units with mazes) during conversation than narration, whereas participants with typical development were more fluent during narration. The main effect of context was not significant,  $F(1, 53) = 0.41, p = .526$ .

Intelligibility differed among groups,  $F(2, 53) = 9.10, p < .001, \eta_p^2 = .26$ , such that it was a weakness of participants with Down syndrome relative to those with fragile X syndrome and those with typical development ( $ps < .002$ ). Participants with fragile X syndrome were less intelligible than those with typical development,

**Table 4.** Language performance in conversation and narration.

Participant group	n	MLCU			Lexical diversity <sup>a</sup>			Fluency <sup>b</sup>			Intelligibility <sup>c</sup>		
		M	SD	Range	M	SD	Range	M	SD	Range	M	SD	Range
Conversation													
Fragile X syndrome	27	3.75	1.45	1.32–7.05	143.85	70.45	38–313	18	13	0–42	12	8	2–37
Down syndrome	15	3.06	0.80	1.84–4.32	115.00	30.69	59–171	23	17	1–66	20	12	9–57
Typical development	15	5.52	1.26	3.30–7.05	173.73	50.80	72–236	27	10	11–48	6	4	1–13
Narration													
Fragile X syndrome	27	4.85	1.36	2.17–7.37	62.26	22.34	28–127	17	10	2–34	10	8	0–28
Down syndrome	15	3.94	1.23	1.40–6.00	50.27	21.63	14–79	31	19	4–68	20	18	3–67
Typical development	15	5.59	0.93	4.07–7.11	67.20	18.35	38–100	18	10	3–38	8	5	2–18

Note. MLCU = mean length of communication unit.

<sup>a</sup>Lexical diversity reflects the number of different word roots produced in the language sample. <sup>b</sup>Fluency reflects the percentage of C-units with mazes. <sup>c</sup>Intelligibility reflects the percentage of unintelligible C-units.



but not significantly so ( $p = .151$ ). Neither the effect of context,  $F(1, 53) = 0.02, p = .900$ , nor the interaction,  $F(2, 53) = 0.71, p = .495$ , was significant for intelligibility.

## Variability Within Fragile X Syndrome

We examined the impact of autism symptom severity on the amount of talk and language performance of participants with fragile X syndrome ( $n = 26$ ), controlling for nonverbal mental age. For each sampling context, we entered autism severity and nonverbal mental age into separate regressions predicting (a) the number of complete and intelligible C-units, (b) the number of C-unit attempts per minute, and (c) MLCU. We predicted that autism symptom severity would negatively relate to language production and, therefore, used one-tailed  $p$  values for testing the regression coefficients in this exploratory analysis; nondirectional  $p$  values were used to test overall models. Unstandardized coefficients are reported (see Table 5).

In conversation, autism severity scores were negatively related to the number of complete and intelligible C-units produced, controlling for nonverbal cognitive ability ( $b = -4.36, t(23) = -2.02, p = .028$ , one-tailed, semipartial  $r = -.37$ ). The overall model predicting the number of attempts per minute in conversation was not significant ( $p = .079$ ); however, severity scores did relate negatively to the number of attempts per minute, controlling for nonverbal cognitive ability ( $b = -0.44, t(23) = -1.91, p = .034$ , one-tailed, semipartial  $r = -.36$ ). Nonverbal cognitive ability was positively related to MLCU in conversation ( $b = 0.92, t(23) = 2.53, p = .010$ , one-tailed, semipartial  $r = .46$ ); autism severity was not ( $b = -0.07, t(23) = -0.81, p = .212$ , one-tailed).

In the narrative context, only the regression predicting MLCU was significant ( $p = .007$ ). As in conversation, nonverbal mental age was positively related to MLCU ( $b = 0.99, t(23) = 3.06, p = .003$ , one-tailed, semipartial  $r = .52$ ), whereas autism severity was not ( $b = -0.10, t(23) = -1.30, p = .103$ , one-tailed). The overall models predicting the number of complete and intelligible C-units or the number of C-units attempted failed to reach significance for narration ( $ps > .18$ ).

## Examiner Behavior

We examined the extent to which examiner behavior was related to the language performance of participants with fragile X syndrome using separate regressions for conversation and narration. As a point of comparison, we repeated these regressions for the adolescents with Down syndrome and the boys with typical development. Of course, these analyses cannot tease apart the direction of causation in the relationship between examiner and child language, but they do provide guidance for future research. Number of C-unit attempts per minute, MLCU, and proportion of questions by the examiner were all expected to be negatively related to amount of participant language and performance (Kover, Abbeduto, Schroeder, Giles, & Richmond, 2008). We tested these three aspects of examiner behavior as simultaneous predictors of participant language output and performance using one-tailed  $p$  values. We chose participant number of C-unit attempts per minute as a dependent variable to index language output because it reflects the clinical utility and efficiency of language samples. We chose participant MLCU as a dependent variable to index language performance because it is widely used as an estimate of spoken language ability.

**Table 5.** Results of regressions for variability among participants with fragile X syndrome ( $n = 26$ ).

Dependent variable	Full model		Individual predictors			
	F	R <sup>2</sup>	NVMA		Autism severity	
			b	t	b	t
Conversation						
Complete and intelligible C-units	3.48*	.23	13.49	1.44	-4.36	-2.02 <sup>†</sup>
C-units attempted per minute	2.84	.20	1.16	1.18	-0.44	-1.91 <sup>†</sup>
MLCU	3.82*	.25	0.92	2.53 <sup>†</sup>	-0.07	-0.81
Narration						
Complete and intelligible C-units	1.85	.14	6.46	1.86 <sup>†</sup>	0.57	0.71
C-units attempted per minute	1.25	.10	1.77	1.51	0.18	0.65
MLCU	6.11*	.35	0.99	3.06 <sup>†</sup>	-0.10	-1.30

Note. NVMA = nonverbal mental age.

<sup>†</sup> $p < .05$ , one-tailed. \* $p < .05$ .

For conversation, the overall model predicting the number of C-units attempted per minute was not significant for participants with fragile X syndrome ( $p = .080$ ) or Down syndrome ( $p = .151$ ). The model was significant for participants with typical development,  $F(3, 11) = 3.69$ ,  $p = .047$ , for whom examiner MLCU was significantly negatively related to C-units per minute ( $b = -2.25$ ),  $t(11) = -2.46$ ,  $p = .016$ , one-tailed, semipartial  $r = -.52$  (see Table 6). When predicting participant MLCU, the overall model was significant for participants with fragile X syndrome,  $F(3, 23) = 11.67$ ,  $p < .001$ , with examiner number of C-units per minute ( $b = -0.24$ ),  $t(23) = -4.88$ ,  $p < .001$ , one-tailed, semipartial  $r = -.64$ , and examiner MLCU ( $b = -0.66$ ),  $t(23) = -3.01$ ,  $p = .003$ , one-tailed, semipartial  $r = -.40$ , emerging as negative predictors (see Table 7). The model predicting MLCU was also significant for typically developing participants,  $F(3, 11) = 5.67$ ,  $p = .014$ , with only examiners' number of C-unit attempts per minute negatively relating to MLCU ( $b = -0.26$ ),  $t(11) = -1.83$ ,  $p = .047$ , one-tailed, semipartial  $r = -.35$ . The overall model predicting MLCU was not significant for participants with Down syndrome ( $p = .065$ ).

For narration, the model predicting the number of C-units per minute was significant for participants with fragile X syndrome,  $F(3, 23) = 5.64$ ,  $p = .005$ . A higher proportion of questions asked by the examiner was associated with fewer C-units per minute ( $b = -0.26$ ),  $t(23) = -4.10$ ,  $p < .001$ , one-tailed, semipartial  $r = -.65$ . The models were not significant for participants with Down syndrome or typical development ( $ps > .18$ ). The model predicting MLCU was not significant for participants with fragile X syndrome ( $p = .238$ ), but it was for participants with Down syndrome and typical development,  $F(3, 11) = 6.99$ ,  $p = .007$ ; and,  $F(3, 11) = 3.77$ ,  $p = .044$ , respectively. For participants with Down syndrome, the number of attempts per minute by the

examiner was significantly associated with lower MLCU ( $b = -0.50$ ),  $t(11) = -3.69$ ,  $p = .002$ , one-tailed, semipartial  $r = -.65$ . For participants with typical development, examiner MLCU was negatively related to MLCU ( $b = -0.44$ ),  $t(11) = -1.89$ ,  $p = .043$ , one-tailed, semipartial  $r = -.40$ .

## Discussion

In this study, we sought to examine the differential effects of language-sampling context on the spontaneous expressive language profiles of male adolescents with fragile X syndrome and Down syndrome. The inclusion of a comparison group of typically developing boys also allowed conclusions to be drawn about the extent of delay in each neurodevelopmental disability relative to nonverbal cognitive ability.

### Comparison of Fragile X Syndrome, Down Syndrome, and Typical Development

*Effects of context among groups.* Performance on the conversation and narration language-sampling tasks was assessed in terms of the amount of talk produced and in terms of language ability. Within both of these areas, differential effects of context on performance were found among participants with fragile X syndrome (regardless of autism symptom severity), Down syndrome, and typical development, controlling for nonverbal mental age.

In terms of the amount of language produced, a significant Group  $\times$  Context interaction revealed that adolescents with Down syndrome differed from both adolescents with fragile X syndrome and boys with typical development in the number of C-units attempted per minute in conversation relative to narration. Although all

**Table 6.** Aspects of examiner behavior as predictors of participant C-unit attempts per minute.

Participant group	n	Individual predictors							
		Full model		Ex. attempts		Ex. MLCU		Ex. questions	
		F	R <sup>2</sup>	b	t	b	t	b	t
Conversation									
Fragile X syndrome	27	2.56	.25	-0.09	-0.52	-1.75	-2.20 <sup>†</sup>	0.03	0.34
Down syndrome	15	2.16	.37	0.56	2.10	0.08	0.06	-0.13	-1.44
Typical development	15	3.69*	.50	-0.45	-1.23	-2.25	-2.46 <sup>†</sup>	0.18	2.11
Narration									
Fragile X syndrome	27	5.64*	.42	0.08	0.26	0.65	1.14	-0.26	-4.10 <sup>†</sup>
Down syndrome	15	0.77	.17	-0.29	-0.55	0.06	0.06	-0.10	-1.05
Typical development	15	1.94	.35	-0.01	-0.01	-0.75	-0.99	-0.07	-0.83

Note. Ex. = Examiner.

<sup>†</sup> $p < .05$ , one-tailed. \* $p < .05$ .

**Table 7.** Aspects of examiner behavior as predictors of participant MLCU.

Participant group	n	Individual predictors							
		Full model		Ex. attempts		Ex. MLCU		Ex. questions	
		F	R <sup>2</sup>	b	t	b	t	b	t
Conversation									
Fragile X syndrome	27	11.67*	.60	-0.24	-4.88 <sup>†</sup>	-0.66	-3.01 <sup>†</sup>	0.01	0.29
Down syndrome	15	3.22	.47	-0.07	-1.12	0.19	0.62	-0.04	-1.98 <sup>†</sup>
Typical development	15	5.67*	.61	-0.26	-1.83 <sup>†</sup>	-0.16	-0.45	-0.02	-0.71
Narration									
Fragile X syndrome	27	1.51	.17	-0.12	-1.04	-0.20	-0.90	-0.02	-0.65
Down syndrome	15	6.99*	.66	-0.50	-3.69 <sup>†</sup>	-0.20	-0.81	-0.02	-0.84
Typical development	15	3.77*	.51	-0.26	-1.69	-0.44	-1.89 <sup>†</sup>	0.01	0.46

<sup>†</sup>p < .05, one-tailed. \*p < .05.

groups tended to have higher rates of C-unit attempts in conversation than narration, adolescents with Down syndrome had particularly low rates of C-units attempted per minute in the narrative context relative to the conversation context. Adolescents with fragile X syndrome showed the smallest difference in number of attempts per minute across contexts, but this effect did not differ from the boys with typical development. Miller (1996) has suggested that the amount of talking during a language sample is an important quantifiable aspect of performance that might reflect an individual's linguistic competence. Narration may be a particularly challenging task for adolescents with Down syndrome relative to their nonverbal cognitive abilities and relative to those with other sources of intellectual disability, such as fragile X syndrome. Given the constraints and demands of the content of the wordless book on the language produced, we speculate that participants with Down syndrome may have required additional processing time in formulating their expressive responses, leading to fewer C-units per minute. In contrast, the demands of reciprocal social interaction in conversation might be more of a challenge for adolescents with fragile X syndrome, resulting in a smaller difference between conversation and narration language production for this population.

In terms of expressive language ability, the effect of context on MLCU differed across groups. Relative to the typically developing boys, for whom MLCU was similar in conversation and narration, adolescents with fragile X syndrome demonstrated higher MLCU in narration than in conversation. Adolescents with Down syndrome also produced higher MLCU in narration than conversation, although the comparison with boys with typical development for the effect of context just failed to reach significance. Previous studies have also suggested that MLU elicited during narration tends to be higher than

conversation for adolescents and young adults with neurodevelopmental disabilities, including fragile X syndrome, Down syndrome, and unspecified etiology (Abbeduto et al., 1995; Kover & Abbeduto, 2010; Levy, Gottesman, Borochowitz, Frydman, & Sagi, 2006; Miles et al., 2006). The narrative task provides opportunities to describe the actions and mental states of a protagonist in relation to other characters as well as to linguistically encode event sequences, all of which are best accomplished using multiclausal constructions (Heilmann, Nockerts, & Miller, 2010). The visual support of narrating a book also may allow production of more advanced syntax (Levy et al., 2006). Miles and colleagues (2006), for example, found that MLCU for participants with Down syndrome was higher in narratives obtained using wordless picture books compared with narratives embedded within an interview-style conversation. Thus, our findings reinforce that narration is particularly well suited to eliciting the upper bounds of syntactic ability in individuals with neurodevelopmental disabilities and that weaknesses might be more likely identified in conversation samples, in which MLCU was lower for participants with fragile X syndrome and Down syndrome, than in narration, in which the discrepancy between the groups with intellectual disability and typical development was minimized (Abbeduto et al., 1995).

There was also a differential effect of context across groups for fluency, which was indexed by the proportion of C-units with mazes (i.e., filled pauses and repetitions). Fluency was found to be better (i.e., fewer mazes) in narration than conversation for typically developing boys, relative to participants with Down syndrome who were more fluent in conversation than narration. Adolescents with fragile X syndrome showed comparable fluency across contexts relative to the other groups. Research on adolescents with Down syndrome and children with typical development has shown poorer fluency in conversation

than narration (Miles et al., 2006), although some research on children with language problems has indicated the opposite (MacLachlan & Chapman, 1988; Wagner et al., 2000). Becoming familiar with a story before narrating likely leads to mental rehearsal, and thus a fluent narrative, whereas the dynamic demands of conversation decrease the opportunities to plan words and phrases in advance (Kover & Abbeduto, 2010). The demands of the narrative task relative to nonverbal cognitive ability for adolescents with particular neurodevelopmental disabilities, however, might outweigh the benefits of previewing a story, leading to particular difficulty with fluency during narration.

The finding that fluency differed less between conversation and narration for participants with fragile X syndrome relative to those with Down syndrome or typical development is surprising, as one might expect that the well-documented social anxiety associated with fragile X syndrome would lead to poorer fluency in conversation. However, relative to the boys with typical development, the adolescents with fragile X syndrome had increased age and life experiences that may have supported performance in the conversation task. It is also possible, however, that individuals with fragile X syndrome relied more on rote or repetitive (i.e., highly rehearsed) linguistic contributions, thereby reducing the processing demands of the task. In fact, Roberts, Price, et al. (2007) found that boys with fragile X syndrome had more perseverative talk than typically developing children and that boys with fragile X syndrome with comorbid ASD had more noncontingent talk than those with fragile X syndrome only. Future research will need to analyze the content of utterances produced by individuals with fragile X syndrome in relation to the other aspects of their language performance. Although the content and form of communicative attempts might be appropriate targets for intervention, producing those attempts fluently appears to be a strength for adolescents with fragile X syndrome.

Of the dimensions of performance examined, there was one significant main effect of context. Participants produced more C-units in conversation than in narration. These results are consistent with those of Abbeduto et al. (1995), who studied youths with intellectual disability of unspecified etiology. The context effect for the absolute number of C-units might be accounted for simply by the fact that the conversation sample was structured to last at least 10 min, whereas the child had the option to say as much or as little as he chose for each page of the storybook in the narrative context. Because the number of C-units attempted per minute also tended to be greater in conversation than narration, these results suggest that conversation is ideal for assessing some aspects of expressive language because of the increased likelihood of efficiently eliciting a larger sample of utterances.

Taken together, the effects of context observed in the present study for participants with fragile X syndrome and Down syndrome highlight the need for language-sampling procedures that include multiple structured contexts to fully understand the profile of language abilities in individuals or groups of individuals with developmental disabilities (Abbeduto et al., 2012). Although the ADOS has been recommended as one structured context in which language can be sampled (Tager-Flusberg et al., 2009), it is unknown whether the ADOS provides a sufficiently consistent context with stable expectations of language performance, even for typically developing individuals. Given the differential effects of context across diagnostic groups, one might expect different conclusions to be drawn about language abilities in neurodevelopmental disabilities depending on the properties of the language-sampling task.

*Extent of delay in fragile X syndrome and Down syndrome.* Several differences in performance emerged between adolescents with fragile X syndrome (irrespective of autism symptoms) or Down syndrome and boys with typical development, controlling for nonverbal mental age.

Adolescents with fragile X syndrome demonstrated lower MLCU than typically developing boys, indicating that syntactic complexity is an area of particular weakness, as shown in previous studies (Roberts, Hennon, et al., 2007). Participants with Down syndrome produced lower MLCU than both the boys with typical development and the adolescents with fragile X syndrome. Price et al. (2008) found the same pattern of results in a younger sample of boys using the ADOS as a language-sampling context: Participants with typical development outperformed those with fragile X syndrome, who outperformed those with Down syndrome. In a narrative context, Finestack and Abbeduto (2010) found that participants with fragile X syndrome without comorbid autism also outperformed participants with Down syndrome on a measure of overall grammatical ability. Children with other neurodevelopmental disabilities, including idiopathic autism, also have deficits in MLU beyond nonverbal expectations (e.g., IQ; Eigsti, Bennetto, & Dadlani, 2007), reinforcing the notion that syntax should be a focus of intervention for a wide range of children with neurodevelopmental disabilities. However, in agreement with findings of previous research, the present study demonstrates that syntactic complexity is a syndrome-specific area of weakness in need of particular attention in adolescents with Down syndrome.

Lexical diversity also was found to be an area of weakness for adolescents with fragile X syndrome or Down syndrome. Both groups of participants with intellectual disability used a smaller range of vocabulary than the boys with typical development, although participants with fragile X syndrome and Down syndrome did not



differ. Although the interaction between context and group failed to reach significance in the present study, the impact of sampling context on lexical diversity should continue to be considered in future research on neurodevelopmental disabilities. Descriptively, participants with Down syndrome seem to use a particularly restricted range of vocabulary during narration, and previous studies have shown that conversation is more likely than narration to assess the scope of vocabulary available to a participant (Kover & Abbeduto, 2010).

Adolescents with fragile X syndrome or Down syndrome did not differ from boys with typical development in fluency; however, those with fragile X syndrome produced a smaller proportion of C-units with mazes (i.e., were more fluent) compared with participants with Down syndrome. In general, participants with fragile X syndrome were the most fluent, which might be expected if these participants were the most likely to use repetitive (i.e., well-practiced) language. The overall group difference between fragile X syndrome and Down syndrome is likely driven by the particularly poor fluency of the adolescents with Down syndrome during the narrative task, suggesting a syndrome- and, to some extent, context-specific area of weakness.

Participants with Down syndrome were less intelligible than both participants with fragile X syndrome and those with typical development. Problems with intelligibility for individuals with Down syndrome have been well documented in previous research, in which even single-word production was impaired relative to typically developing boys and boys with fragile X syndrome with similar nonverbal cognitive abilities (Roberts, Price, et al., 2007). The present results are also generally in line with Barnes et al. (2009), who found that boys with Down syndrome produced a smaller percentage of intelligible words than those with typical development; however, boys with fragile X syndrome did not differ from those with Down syndrome in the Barnes et al. study. In a sample of older adolescents, Kover and Abbeduto (2010) also failed to find differences between participants with Down syndrome and those with fragile X syndrome with or without autism. The inconsistent findings for differences in intelligibility between individuals with fragile X syndrome and Down syndrome may be attributable to varying age ranges of participants, procedures for assessing intelligibility, or language-sampling contexts.

In summary, combining words into developmentally progressive grammatical utterances and expanding range of vocabulary are likely to be important intervention targets for adolescents with fragile X syndrome. For adolescents with Down syndrome, extending syntactic complexity and vocabulary, in addition to enhancing fluency and intelligibility during connected speech, are likely to be areas in need of remediation.

## **Autism Symptom Severity**

We examined the relationship between a continuous metric of autism symptom severity in males with fragile X syndrome and their language production in the conversation and narrative contexts, controlling for nonverbal mental age. Our results suggest that individuals with more severe autism symptoms are less able to engage with the demands of conversation, as reflected in the production of fewer complete and intelligible utterances. We found no relationship between autism symptom severity and language production in narration; however, nonverbal cognitive level accounted for variability in MLCU in both narration and conversation. It seems reasonable that males with fragile X syndrome who display more symptoms of autism could find the social demands of conversation more challenging given the pressure to engage in a social give-and-take, whereas engaging in the narrative task—and perhaps producing grammatically complex utterances in general—is more dependent on nonverbal cognitive skills. Producing contingent talk in conversational contexts has been identified as an area of weakness for children with idiopathic autism as well (Tager-Flusberg & Anderson, 1991).

## **Examiner Behavior**

To our knowledge, no previous studies have examined the ways in which examiner behavior relates to spontaneous language in youths with fragile X syndrome or Down syndrome. Even within the constraints of our structured procedures, relationships were found between variability in examiners' talk and the language performance of participants. In conversation, participants' MLCU was negatively related to both examiner MLCU and number of C-unit attempts per minute for participants with fragile X syndrome and was related to C-unit attempts per minute for participants with typical development. Examiner behavior was not related to the performance of participants with Down syndrome in conversation. In the narrative, number of C-unit attempts per minute by participants with fragile X syndrome was negatively related to the examiner's proportion of questions, whereas MLCU for participants with Down syndrome or typical development was negatively predicted by examiners' C-unit attempts per minute and MLCU, respectively.

For a group of language-impaired children, Dethorne and Channell (2007) interpreted correlations between clinician behavior and child language as resulting from the clinician adjusting her talk in relation to the talk produced by the children. It would be expected that a child who is reticent might elicit more coaxing from an examiner, and the structure provided by the examiner as an interlocutor should not be discounted. Similarly,

the most likely explanation of our results is that the relationships observed reflect variability in response to the language-sampling tasks by participants and the efforts of the examiner to maintain participant engagement; however, it is noteworthy that the pattern of relationships varied across participants with fragile X syndrome, Down syndrome, and typical development. Such relationships should be taken into account in designing language-sampling protocols that are flexible, yet structured, to meet the needs of particular individuals and diagnostic groups. Future research should probe the direction of causality between examiner and child language.

## Limitations

The results presented here are based on language samples of relatively short duration and a relatively small number of utterances. Although longer language samples are in some ways ideal (e.g., 175 utterances; Rice et al., 2010), there is evidence that shorter (e.g., 3-min) language samples can provide reliable estimates of language ability (Heilmann et al., 2010). In this sense, the results are optimally interpretable from a clinical perspective, given that it might be unrealistic to collect larger language samples to assess youths with neurodevelopmental disabilities in a clinical setting. This is particularly true for those with fragile X syndrome, for whom inattentiveness or hyperactivity could impact participation in assessment procedures. Nonetheless, the fact that the language samples analyzed here contained a limited number of utterances, particularly for the narrative samples, should be considered in terms of the generalizability of the findings. Only nine participants with fragile X syndrome, four participants with Down syndrome, and three participants with typical development produced 50 or more C-units during narration. A potential advantage of the ADOS as a language-sampling context is the likelihood of obtaining a larger sample during the 30- to 60-min assessment.

The participants in the present study were limited to those who scored within a restricted range of nonverbal cognitive ability, in order to allow simultaneous comparison of those with fragile X syndrome and typical development, who tended to have higher developmental levels, to those with Down syndrome, who tended to have slightly lower developmental levels in the larger sample from which these data are drawn. Having controlled for nonverbal mental age, this analysis strategy allowed for unambiguous interpretation of group differences within this developmental range but limits the generalizability of the findings to other individuals along the range of the highly variable phenotypes of fragile X syndrome and Down syndrome. Investigation of the impact of sampling contexts on the language of individuals with neurodevelopmental disabilities

across a range of nonverbal cognitive ability levels is warranted.

Finally, the present study did not address the content of the utterances produced by participants in terms of repetitiveness, perseveration, or contingency. This will be an important area for future research, especially with respect to the impact of autism symptoms on spontaneous expressive language in adolescents with fragile X syndrome. Indeed, behavioral and pharmaceutical interventions that seek to improve expressive language in fragile X syndrome are likely to target not only the amount of talk and the grammatical complexity of that talk but also its communicative function by assessing and documenting reduction in perseverative or non-contingent utterances.

## Conclusions

We have reported findings that draw attention to the importance of considering the context in which language samples are elicited for individuals with neurodevelopmental disabilities. These results suggest that the demands of the language-sampling context could have implications for targets selected for intervention and conclusions drawn about language profiles.

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**Effects of Sampling Context on Spontaneous Expressive Language in Males With Fragile X Syndrome or Down Syndrome**

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