Syntactic Complexity During Conversation of Boys With Fragile X Syndrome and Down Syndrome

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Purpose: This study compared the syntax of boys who have fragile X syndrome (FXS) with and without autism spectrum disorder (ASD) with that of (a) boys who have Down syndrome (DS) and (b) typically developing (TD) boys.
Method: Thirty-five boys with FXS only, 36 boys with FXS with ASD, 31 boys with DS, and 46 TD boys participated. Conversational language samples were evaluated for utterance length and syntactic complexity (i.e., Index of Productive Syntax; H. S. Scarborough, 1990).
Results: After controlling for nonverbal mental age and maternal education levels, the 2 FXS groups did not differ in utterance length or syntactic complexity. The FXS groups and the DS group produced shorter, less complex utterances overall and less complex noun phrases, verb phrases, and sentence structures than did the TD boys. The FXS with ASD group and the DS group, but not the FXS-only group, produced less complex questions/negations than did the TD group. Compared with the DS group, both FXS groups produced longer, more complex utterances overall, but on the specific complexity measures, they scored higher only on questions/negations.
Conclusion: Boys with FXS and DS have distinctive language profiles. Although both groups demonstrated syntactic delays, boys with DS showed greater delays.

KEY WORDS: fragile X syndrome, Down syndrome, syntax, X-linked

For many children with developmental language disorders, syntax appears to be a particularly vulnerable domain. For example, children with specific language impairment (SLI; Hadley, 1998; Rice, Tomblin, Hoffman, Richman, & Marquis, 2004; Schuele & Dykes, 2005) and subgroups of children with high-functioning autism (Kjelgaard & Tager-Flusberg, 2001; Landa & Goldberg, 2005) demonstrate weaknesses in productive syntax despite nonverbal cognitive skills within the normal range. Children with *Down syndrome* (DS), the most common genetic cause of intellectual disability, demonstrate syntactic deficits beyond their general cognitive and language delays (Abbeduto & Chapman, 2005; Chapman & Hesketh, 2000; Chapman, Schwartz, & Kay-Raining Bird, 1991). However, few investigations have explored the syntactic skills of children with *fragile X syndrome* (FXS), the most common inherited form of intellectual disability (Rice, Warren, & Betz, 2005), and additional research in this area is needed. Moreover, a major goal of current research in developmental disabilities is to compare language phenotypes across syndromes to determine whether each syndrome is characterized by a distinct language profile or if language characteristics can be explained more generally by the presence of intellectual disability (Rice et al., 2005; Tager-Flusberg, 2005). In this study, we compared the syntactic skills of boys with FXS with and without autism spectrum disorder (ASD) to those

of boys with DS and younger boys who are typically developing (TD) in order to further characterize the language profiles of each group and to determine whether syntactic skills differ according to diagnosis.

Genotype and Phenotype of FXS

FXS is the most common inherited cause of intellectual disability, occurring in 1 of every 4,000 boys and 1 of every 8,000 girls (Crawford, Acuna, & Sherman, 2001; Turner, Webb, Wake, & Robinson, 1996). For a discussion of the causes of FXS, see Devys, Lutz, Rouyer, Bellocq, and Mandel (1993) and Jin and Warren (2003). Because FXS is an X-linked disorder, more boys than girls have the syndrome, and the development of boys is more severely affected than that of girls (Hagerman & Hagerman, 2002; Loesch et al., 2003; Reiss & Dant, 2003). The presence of a second, unaffected X chromosome in girls with FXS moderates the effects of the affected X chromosome (Hagerman, 2002). Boys with FXS generally have mental retardation and language deficits, but only a subgroup of girls with FXS has mental retardation, and girls have less severe language difficulties than boys (Abbeduto & Chapman, 2005; Keysor & Mazzocco, 2002). In addition, the considerable task of recruiting girls with FXS with mental retardation, which occurs with a relatively low incidence, was beyond the scope of the present study. Therefore, boys are the focus of this study.

Most boys with FXS have moderate to severe levels of intellectual disability (Abbeduto & Chapman, 2005). Boys with FXS typically have moderate to severe delays in communication, with greater delays in the expressive than the receptive modality (Abbeduto & Hagerman, 1997; Bennetto & Pennington, 2002; Roberts, Mirrett, & Burchinal, 2001; Philofsky, Hepburn, Hayes, Hagerman, & Rogers, 2004), although there is wide variability among individuals (Abbeduto & Hagerman, 1997; Roberts et al., 2001). Studies of adolescents and adults with FXS have reported difficulties in many aspects of communication, including grammar, vocabulary, pragmatics, and speech development (Fryns, Jacobs, Kleczkowska, & Van den Berghe, 1984; Madison, George, & Moeschler, 1986; Newell, Sanborn, & Hagerman, 1983; Palmer, Gordon, Coston, & Stevenson, 1988; Sudhalter, Scarborough, & Cohen, 1991).

Among individuals with FXS, 15%–25% are diagnosed with autism (Bailey, Hatton, & Skinner, 1998; Dykens & Volkmar, 1997; Hagerman, 2002), and approximately 5.5% of males with autism test positive for FXS (Dykens & Volkmar, 1997; Hagerman, 2002). Communication deficits are a defining feature of autism (American Psychiatric Association, 2000), and language impairments vary widely and include both receptive and expressive modalities (Joseph, Tager-Flusberg, & Lord, 2002; Kjelgaard & Tager-Flusberg, 2001). Even though there is a high comorbidity rate between FXS and autism, only a few studies to date have investigated the relationship between autism and language skills in children with FXS, and these findings have not reached consensus. This may be for a number of reasons—FXS samples' inclusion of both boys and girls (who have different levels of cognitive and linguistic functioning), small sample size, different age groups, different criteria for autism or autism spectrum diagnosis, different control variables, and different measures of language abilities.

In general, receptive language appears to be lower or the same in individuals with FXS and autism compared with individuals with FXS only. Some studies have found lower receptive language skills in FXS with autism than in FXS only (Lewis et al., 2006; Rogers, Wehner, & Hagerman, 2001), and some have found no differences between groups in receptive language ability (Price, Roberts, Vandergrift, & Martin, 2007; Roberts, Price, et al., 2007). This pattern of findings may be influenced by methodological differences among the studies. Lewis et al. (2006) and Rogers et al. (2001) each used different criteria to establish autism status that may have been more stringent than that used by Price et al. (2007) and Roberts, Price, et al. (2007), whose samples overlapped. In addition, the Lewis et al. (2006) and Rogers et al. (2001) studies included both girls and boys with FXS, whereas the Price et al. (2007) and Roberts, Price, et al. (2007) studies included only boys. The instruments used to assess receptive language also varied across the studies, with only Lewis et al. (2006) and Price et al. (2007) using the same instrument (i.e., Test for Auditory Comprehension of Language; Carrow-Woolfolk 1985, 1999). The ages of the samples also varied considerably: Rogers et al. (2001) studied toddlers, Lewis et al. (2006) studied adolescents and young adults, and Price et al. (2007) and Roberts, Price, et al. (2007) studied school-aged children.

Expressive language abilities may be more stable across groups. Although Philofsky et al. (2004) found that expressive language was lower in FXS with autism than in FXS only, other studies found that there was no difference (Lewis et al., 2006; Roberts, Price, et al., 2007). In the Philofsky et al. (2004) study, developmental level was not controlled, and participants were much younger than in the Lewis et al. (2006) and Roberts, Hennon, et al. (2007) studies. In the Philofsky et al. (2004) and Lewis et al. (2006) studies, standardized instruments were used to measure expressive language, whereas Roberts, Price, et al. (2007) used naturalistic language samples. Again, the measure of expressive language, the criteria for autism, and whether girls were included in the sample varied for each study. In order for research findings on language abilities in individuals with FXS with and without autism to reach consensus, the above studies need to be replicated, with attention paid to increasing sample size and increasing consistency of age

groups, criteria for autism or autism spectrum diagnosis, control variables, and measures of language abilities. To our knowledge, no studies have specifically examined whether syntactic skills differ for boys with FXS with and without autism. The current study includes both of these FXS groups in order to make this comparison.

Expressive Syntax in FXS

Using a variety of measures, Sudhalter and colleagues (1991, 1992) have reported that the expressive syntactic skills of individuals with FXS without autism are similar to those of younger TD children. In their first study (Sudhalter et al., 1991), two measures of overall syntactic development—mean length of utterance (MLU) and Index of Productive Syntax (IPSyn; Scarborough, 1990) scores—were calculated from language samples of 19 males with FXS without autism (ages 5–36 years; mean Communication age equivalent on the Vineland Adaptive Behavior Scales [VABS; Sparrow, Balla, & Cicchetti, 1984] = 4.89, *SD* = 1.73). Although they did not use a comparison group of TD children in their study, the authors concluded that the MLU values and IPSyn scores of the males with FXS were similar to those of TD preschoolers previously reported by Scarborough (1990). In the second study (Sudhalter, Maranion, & Brooks, 1992), the number of syntactic errors on a sentence completion task produced by 11 males with FXS without autism (ages 6–41 years; mean Communication age equivalent on the VABS = 4 years, 4 months) did not differ from that of 11 TD 4-year-olds. Although the FXS and TD groups were presumably at similar language levels, the two groups were not statistically matched on either developmental or language level. In a similar vein, Madison and colleagues (1986) reported that the levels of expressive syntax of 5 males with FXS within one family (ages 4–64 years; autism status not reported) were judged commensurate with their cognitive levels. Paul and colleagues (1987) reported that the MLU and overall expressive language scores of 12 adult males with FXS (2 of whom also were diagnosed with autism) did not differ significantly from those of males with nonspecific forms of intellectual disability or males with autism matched on age and cognitive level. However, the authors suggested that a nonsignificant trend indicated possible syntactic deficits.

In contrast, Roberts, Hennon, and colleagues (2007), in a study of syntactic complexity and vocabulary diversity during conversation of boys with FXS without autism and younger TD boys, found that MLUs and IPSyn scores of boys with FXS without autism were lower than those of younger TD boys, after controlling for nonverbal cognitive skills, speech intelligibility levels, and maternal education. The participants in Roberts, Hennon, et al.'s (2007) study overlap with those in the FXS without autism group and TD group investigated in the current study. Methodological differences between the work of Roberts, Hennon, et al. (2007) and Sudhalter et al. (1991, 1992) may account for the differences in their findings. Roberts, Hennon, et al.'s (2007) sample of 35 boys (mean age = 9.1 years) was larger and considerably younger than those in Sudhalter et al.'s (1991, 1992) studies. Roberts, Hennon, et al. (2007) also included a TD control group and controlled for nonverbal cognitive level, speech intelligibility, and maternal education level. Paul, Cohen, Breg, Watson, and Herman (1984) reported that expressive syntactic skills were lower than nonverbal cognition in 3 boys with FXS (ages 10–13 years), echoing the findings of Roberts, Hennon, and colleagues (2007).

Particular areas of strength and weakness within the domain of syntax remain relatively unexplored. However, Roberts, Hennon, et al. (2007) reported children's scores on subscales of the IPSyn. The noun phrases, verb phrases, and sentence structures, but not questions and negations, produced by boys with FXS without autism were less complex than those of younger TD boys, after controlling for nonverbal mental age, intelligibility levels, and maternal education.

Genotype and Phenotype of DS

DS is a genetic disorder in which there is a third chromosome 21. It is the most common known cause of intellectual disability and occurs in approximately 13.65 of 10,000 live births or in approximately 1 of 730 births (Carothers, Hecht, & Hook, 1999; Centers for Disease Control and Prevention, 2006). Unlike FXS, DS affects boys and girls similarly. For children with DS, language skills are more severely affected than nonverbal cognition (Abbeduto et al., 2003; Fowler, 1990; Miller, 1988; Yoder & Warren, 2004). Expressive language skills are poorer than receptive language skills (Abbeduto et al., 2003; Chapman, Seung, Schwartz, & Kay-Raining Bird, 1998; Sigman & Ruskin, 1999), and syntax tends to be considerably delayed (Chapman et al., 1998; Eadie, Fey, Douglas, & Parsons, 2002; Fowler, Gelman, & Gleitman, 1994; Laws & Gunn, 2004).

Robust research findings indicate that expressive syntax is an area of particular weakness for individuals with DS. Children with DS have lower MLUs than younger TD nonverbal mental age matches (Chapman et al., 1998; Miller, 1988; Rosin, Swift, Bless, & Vetter, 1988) and mental age matches with intellectual disability of unknown etiology (Rosin et al., 1988). When compared with younger MLU-matched children, children with DS omit more grammatical function words (such as copula and auxiliary *be,* articles, and prepositions; Chapman et al., 1998) and more tense and non-tense bound morphemes (Chapman et al., 1998; Eadie et al., 2002). Children with DS have also been reported to use fewer grammatical verbs (auxiliary and copula *do, be,* and *have*) and fewer

lexical verbs (main verbs that do not include *do, be,* or *have*) per utterance than MLU-matched controls (Hesketh & Chapman, 1988).

Only a few studies have compared expressive language in FXS and DS, and only one has compared expressive syntax in particular. In the only study of syntax, Ferrier, Bashir, Meryash, Johnston, & Wolff (1991) found that 18 male children and adults with FXS without autism and 18 children and adults with DS, matched on chronological age and overall cognitive level, did not differ on MLU or on a broad-based measure of semantics and syntax derived from conversational language samples. However, Abbeduto and colleagues (2001) found that individuals with FXS (autism status not specified) outperformed individuals with DS on a standardized measure of overall expressive language skills.

Use of MLU and IPSyn

Language samples are often used to analyze the syntactic production of both young children and individuals with intellectual disability (Condouris, Meyer, & Tager-Flusberg, 2003; Hadley, 1998; Hewitt, Hammer, Yont, & Tomblin, 2005; Scarborough, Rescorla, Tager-Flusberg, Fowler, & Sudhalter, 1991; Sudhalter et al., 1991). Language samples are especially useful with individuals who may have difficulty attending to standardized tests, including young children and those with intellectual disability (Scarborough et al., 1991). They also provide naturalistic samples of syntax produced by children in spontaneous conversation.

MLU is a well-established measure used to quantify utterance length in language samples and has been shown to relate to grammatical complexity, especially at lower MLU levels (Brown, 1973; Paul, 2007; Scarborough et al., 1991). However, although MLU has often been used to assign stages of development (Brown, 1973; Hadley, 1998) and for identifying language impairment (Eisenberg, Fersko, & Lundgren, 2001), it does not specifically quantify emerging syntax (Leonard & Finneran, 2003). To do this, the IPSyn (Scarborough, 1990) has been used. The IPSyn was developed to measure syntactic and morphological complexity in the language samples of preschoolers. It was also designed for efficient analysis of language samples in large-scale research studies (Scarborough, 1990). It has been used to measure emerging syntax in late talkers (Rescorla, Bascome, Lampard, & Feeny, 2001), in children with SLI (Hadley, 1998; Hewitt et al., 2005; Oetting, Cantrell, & Horohov, 1999), in preterm children at risk for language delays (Holdgrafer, 1995), and also in individuals with FXS (Roberts, Hennon, et al., 2007; Scarborough et al., 1991; Sudhalter et al., 1991), DS (Scarborough et al., 1991), and autism (Condouris et al., 2003; Scarborough et al., 1991). It has also been shown to be an effective measure of syntax in studies of typically developing children up to 6 years of age (Hewitt et al., 2005; Oetting et al., 1999). Studies have shown that MLU and IPSyn scores are closely related in typically developing children (Roberts, Hennon, et al., 2007; Scarborough, 1990), individuals with FXS (Roberts, Hennon, et al., 2007; Scarborough et al., 1991; Sudhalter et al., 1991), individuals with DS (Scarborough et al., 1991), and individuals with autism (Tager-Flusberg & Calkins, 1990), although this relationship may weaken as MLU increases.

Current Study

In this investigation, we described and compared the syntactic skills of children with FXS with and without autism, children with DS, and TD children. The existing literature on syntax in FXS reveals inconsistent findings and fails to examine the impact of autism on syntactic skills in FXS. Children with DS and boys with FXS have similar degrees of cognitive impairment, yet they appear to have some differences in language skills, forming ideal comparison samples. Previous work has shown that receptive and expressive language is correlated with nonverbal cognitive ability for individuals with DS and individuals with FXS (Abbeduto et al., 2003; Price et al., 2007; Roberts et al., 2001), but the relationship specifically between syntax and nonverbal cognition has not been explored for these populations. This study differs from our previous work that investigated the syntactic production and lexical diversity of only two of these groups (children with FXS without autism and TD children; Roberts, Hennon, et al., 2007) and the receptive and expressive vocabulary and speech skills, but not syntax, of all four groups (Roberts, Price, et al., 2007). Our research questions were:

1. Do children with FXS without ASD, children with FXS with ASD, children with DS, and younger TD children differ on measures of expressive syntax?
2. For each of the four groups, are syntactic levels correlated with nonverbal cognitive levels?

We hypothesized that, after controlling for nonverbal developmental and maternal education levels, boys with FXS (regardless of autism status) would score lower on expressive syntactic skills than TD boys; boys with FXS without ASD would demonstrate more advanced syntactic skills than boys with FXS with ASD; and the DS group would have lower expressive syntactic skills than all of the other groups. We further hypothesized that syntactic and nonverbal cognitive levels would be correlated for all groups.

Method

Participants

Four groups of children (boys with FXS with ASD, boys with FXS without ASD, boys with DS, and TD boys)

participated in this study as part of a larger longitudinal investigation of the speech and language skills of boys with FXS, DS, or typical development (Roberts, Price, et al., 2007). To be enrolled in the study, boys with FXS and boys with DS were 16 years of age or younger, had an expressive vocabulary of at least 40 words, and were combining at least two words (i.e., MLU > 1.1). The TD boys were between 2 and 6 years of age and, compared with the boys with FXS and the boys with DS, showed a similar distribution of developmental ages for nonverbal cognitive abilities on the Brief IQ composite of the Leiter International Performance Scale–Revised (Leiter-R; Roid & Miller, 1997). Leiter-R Brief IQ age-equivalent scores of the four groups did not differ, $F(3, 144) = 1.52$, $p = .2115$. Further, the three disorder groups (boys with FXS with ASD, boys with FXS without ASD, and boys with DS) did not differ significantly on either chronological age, $F(2, 99) = 2.49$, $p = .0885$, or Leiter-R IQ scores, $F(2, 99) = .26$, $p = .7680$. All children's hearing thresholds were below 30 dB in the better ear, as determined by screening at 500, 1000, 2000, and 4000 Hz using a Grason Stadler GSI 16, Grason Stadler GSI 17, or MAICO MA 40 audiometer. In addition, all participants used spoken English, rather than sign, as their primary mode of communication, and English was the primary language spoken in their homes. The School of Medicine Institutional Review Board at the University of North Carolina at Chapel Hill reviewed and approved study protocols annually. The parent or guardian provided informed consent at study entry.

Seventy-one boys with FXS participated in the study. All boys with FXS had been diagnosed with full mutation FXS. They were recruited from ongoing longitudinal studies of children with FXS, genetics and developmental clinics, physicians' offices, and parent support groups in the Eastern United States. Boys with FXS were divided into two groups based on their autism status, as determined by the Autism Diagnostic Observation Schedule (ADOS; Lord, Rutter, DiLavore, & Risi, 2001). See *Measures* section for a description of the ADOS and how boys with FXS were grouped.

Boys with FXS only (FXS-O). Thirty-six boys with FXS did not have ASD. Their chronological ages ranged from 2.9 to 14.4 years ($M = 9.9$ years, $SD = 2.0$), and their nonverbal mental ages ranged from 2.2 to 6.7 years ($M = 5.0$ years, $SD = 0.9$), with IQs from 37 to 108 ($M = 54.4$, $SD = 15.0$) as measured by the Leiter-R Brief IQ. (See *Measures* section for a description of the Leiter-R Brief IQ.) Maternal education levels ranged from 12 to 20 years ($M = 14.1$ years, $SD = 2.3$).

Boys with FXS and autism spectrum disorder (FXS-ASD). Thirty-five boys with FXS also had ASD. Their chronological ages ranged from 3.5 to 14.0 years ($M = 8.4$ years, $SD = 2.9$), with nonverbal mental ages ranging from 2.1 to 5.9 years ($M = 4.5$ years, $SD = 1.0$) and IQs ranging from 38 to 87 ($M = 56.9$, $SD = 13.1$) on the Leiter-R Brief IQ. Maternal education levels ranged from 12 to 20 years ($M = 15.2$ years, $SD = 2.2$).

Boys with DS. Thirty-one boys with DS participated in the study, ranging in chronological age from 4.3 to 16.0 years ($M = 9.2$ years, $SD = 2.9$), with nonverbal mental ages from 2.3 to 8.2 years ($M = 4.8$ years, $SD = 1.0$) and IQs from 37 to 83 ($M = 55.8$, $SD = 10.6$) on the Leiter-R Brief IQ. They were recruited from genetics and developmental clinics, physicians' offices, schools, and parent support groups in North Carolina. Children with DS who also had a diagnosis of ASD, according to parent report, were excluded from the study. In addition, boys with DS who received a score of "autism" or "spectrum" on the ADOS were excluded from data analyses. Maternal education levels ranged from 12 to 20 years ($M = 16$ years, $SD = 2.3$).

TD boys. Forty-six TD boys participated in the study. They were recruited from child care centers, physicians' offices, and schools in North Carolina and, according to parent report, did not have a history of speech, language, or developmental delays; ASD; or hearing loss. Additionally, any TD boy who received a score of "autism" or "spectrum" on the ADOS was excluded from analyses. We also excluded any TD boy who scored more than 1.5 *SD*s below the mean on any standardized speech or language test given as part of the larger assessment battery. Chronological ages ranged from 2.1 to 6.6 years ($M = 4.5$ years, $SD = 1.1$), with Leiter-R Brief IQ nonverbal mental ages ranging from 2.8 to 7.1 years ($M = 4.8$ years, $SD = 1.0$). Maternal education levels ranged from 12 to 20 years ($M = 16.4$ years, $SD = 2.0$).

Procedures for Data Collection

Each child was tested in his home, at his school, or at a university research center, depending on parental preference. All sessions were audiotaped using a portable Digital Auditory Tape TASCAM (DA-P1) recorder with a Shure WBH headset microphone system and were videotaped using a Sony Digital8 video camera (Model DCR-TVR27).

Measures

Nonverbal cognition. The Brief IQ composite of the Leiter-R was used as a measure of nonverbal cognition. Four subtests were administered: Figure Ground, Form Completion, Sequential Order, and Repeated Patterns. In these subtests, the individual is asked to find an item in a picture, choose the next item in a sequence, or arrange items in a pattern. The Leiter-R is standardized on 1,719 individuals aged 2–20 years. It has high reliability levels, with a test–retest coefficient of .96 for the Brief IQ composite and alpha reliability coefficients ranging

from .75 to .88 for the four subtests. The Leiter-R has adequate validity, correlating (.85 to .86) with other widely used IQ tests. An age-equivalent score was computed for each child using published norms.

Maternal education. Maternal education was measured by total years of education at time of enrollment in the study. It was included as a proxy for the effects of environmental factors on the relationship between verbal and nonverbal skills. A number of studies have demonstrated that children with higher levels of maternal education have more sophisticated speech and language development (Campbell et al., 2003; Dollaghan et al., 1999; Fewell & Deutscher, 2003; Rice, Spitz, & O'Brien, 1999).

Autism status in FXS. The boys with FXS were classified according to autism status using the ADOS. The ADOS is a standardized observation of children's communicative and social behavior that discriminates autistic disorder from other developmental disorders and normal behavior and yields categorical scores of "no autism," "spectrum," and "autism." The examiner interacted with the child for approximately 45 minutes in a series of structured and semistructured activities in which the child was given opportunities to exhibit behaviors indicative of autism. Trained examiners scored videotapes of ADOS interactions, and reliability computed on 16% of the boys was .89 for the individual items (range: .83–.96) and .93 on diagnosis (range: .81–1.00). Thirty-six boys with FXS received an ADOS score of "no autism," whereas 23 boys received a score of "spectrum" and 12 boys received a score of "autism." In our analyses, boys with FXS with scores of "spectrum" or "autism" were combined into a single group (FXS-ASD).

Language samples. The ADOS was administered to all study participants, and the ADOS interactions were used as language samples. During the ADOS, the examiner elicited language from the child by engaging him in play-based activities using developmentally appropriate materials (e.g., balloons, blocks, family figurines and miniature furniture, action figures) and social situations (e.g., giving a pretend birthday party, requesting a snack). The examiner encouraged the child to interact with the examiner. One hundred utterances are required to complete the IPSyn analysis (Scarborough, 1990); however, a few children with DS or FXS produced fewer than 100 useable utterances during the ADOS. One child with DS (3% of the DS group) produced 93 utterances during the ADOS, 3 children with FXS-O (9% of the FXS-O group) produced 81, 79, and 62 utterances each, and 4 children with FXS-ASD (11% of the FXS-ASD group) produced 99, 91, 68, and 63 utterances each. For these 8 children, transcribers reviewed tapes from other language assessments administered during the same visit as the ADOS and transcribed consecutive spontaneous utterances produced by the child until the 100-utterance requirement was met. Child responses elicited by test questions or prompts were not used. Most of these additional, spontaneous utterances were unsolicited comments about test pictures or stimulus materials or utterances produced during breaks between administration of tests or test items. For example, during administration of picture-pointing tasks, children sometimes produced spontaneous comments relating the picture to their personal experiences (e.g., "Mine"; "I have one like that").

Transcription. Trained research assistants transcribed and coded at least 100 useable child utterances from videotapes using the Child Language Data Exchange System (CHILDES; MacWhinney, 1995). Following the guidelines specified by Scarborough (1990) for use of the IPSyn, an utterance was excluded if it was partially or fully unintelligible, part of a routine (e.g., reciting the alphabet), an imitation of an examiner utterance, an exact self-repetition (e.g., "Let's play soccer." "Let's play soccer."), or a yes/no response to a question. The first 100 useable utterances for each child were used for all analyses.

Measures of Syntax

MLU. MLU is a ratio of the total number of morphemes divided by the total number of utterances, as described by Brown (1973). MLU was computed using CHILDES and was used to measure the length of children's utterances.

IPSyn. A trained research assistant scored all transcripts using the IPSyn (Scarborough, 1990), which identifies 56 syntactic and morphological forms, plus four items labeled "other." The first two occurrences of each form are scored, for a maximum of 2 points for each form. Zero occurrences indicate that the child does not yet produce this form, one occurrence reflects emerging abilities with the form, and two occurrences indicate productive use of the form (Scarborough, 1990). Total points, as well as points for four subscales (Noun Phrases, Verb Phrases, Questions and Negations, and Sentence Structure), are calculated. The maximum possible scores are 120 for the IPSyn Total, 24 for Noun Phrases, 34 for Verb Phrases, 22 for Questions and Negations, and 40 for Sentence Structure. Good reliability and age sensitivity have been reported for children between 2 and 4 years of age (Scarborough, 1990), and the IPSyn has been used effectively with TD children up to 6 years of age and with a variety of clinical populations, as discussed in the literature review. The participants in the current study were roughly within this developmental range, with mean nonverbal mental ages on the Leiter-R ranging from 4.5 to 5 years for each of the four groups studied.

Reliability

Transcription. All original transcripts were verified and corrected by a second researcher via audiotape.

Thirteen percent (n = 19) of the original transcripts were randomly selected and then independently verified and corrected via audiotape by a third researcher for reliability. The verified transcripts of the second and third researchers were then compared, and morpheme-to-morpheme agreement was calculated. Overall agreement was 91%. Agreement was 85% for the transcripts of boys with FXS-O (n = 4, 11% of FXS-O group); 85% for boys with FXS-ASD (n = 5, 14% of FXS-ASD group); 90% for boys with DS (n = 5, 16% of DS group); and 95% for TD boys (n = 5, 11% of TD group).

IPSyn. Fourteen percent of transcripts were independently scored by a second researcher. Inter-rater reliability, as reported in a kappa value, for IPSyn Total scores was .79, indicating excellent reliability (Landis & Koch, 1977; Rosner, 2000).

Data Analysis Strategy

Analyses of covariance (ANCOVAs) tested both the two overall measures of syntax (MLU and IPSyn Total) and the four measures of specific aspects of syntax (Noun Phrases, Verb Phrases, Questions and Negations, and Sentence Structure). In all models, the primary predictor variable was diagnostic group. This was a categorical variable with four levels: boys with FXS-O, boys with FXS-ASD, boys with DS, and TD boys. In all analyses, the primary predictor variable was diagnostic group, with maternal education as measured in years and nonverbal mental level as measured by Leiter-R age-equivalent score treated as covariates. In addition to these main effects, the interactions of the covariates with diagnostic group were included to test whether each might have an impact on the degree of differences between the groups. Subsequent to each ANCOVA, pairwise tests comparing all diagnosis groups were conducted to explore any indicated group differences. Where significant between groups differences were detected, Cohen's d was calculated as a measure of effect size. Effect sizes were computed as the difference between the adjusted means between the two groups divided by the model root-mean-square error from the corresponding analysis. Cohen (1988) designated an effect size of .2 as small, .5 as medium, and .8 as large. We also calculated Spearman correlations among three variables: Leiter-R nonverbal mental age, MLU, and IPSyn Total.

Results

Descriptive Analyses

Means and standard deviations for MLU, IPSyn Total, Verb Phrases, Noun Phrases, Questions and Negations, and Sentence Structure scores are shown in Table 1. Correlations among MLU, IPSyn Total scores, and Leiter-R nonverbal mental ages for each of the four groups are reported in Table 2. All of the correlations were significant for all groups. MLU and IPSyn Total scores were equally highly correlated for all of the groups (r = .78–.93, $p < .001$). Although MLU and IPSyn Total scores were also correlated in all the groups, these correlations were stronger for the DS and TD groups (r = .48–.67, $p < .001$) than for the FXS-O and FXS-ASD groups (r = .39–.48, $p < .05$).

Group Comparisons

Measures of overall syntax. The models for MLU and IPSyn Total were statistically significant, $F(12, 136) = 274.86$, $p < .0001$, and $F(12, 136) = 274.86$, $p < .0001$, respectively, after controlling for nonverbal mental age and maternal education levels. Further, the results indicated significant tests for the effect of diagnostic group on MLU, $F(3, 136) = 32.68$, $p < .0001$, and IPSyn

Table 1. Means and standard deviations for syntactic measures.

	FXS-O		FXS-ASD		DS		TD	
Variable	*M*	*SD*	*M*	*SD*	*M*	*SD*	*M*	*SD*
Overall syntax								
MLU	3.6	1.1	3.3	0.8	2.8	0.8	4.7	1.0
IPSyn total	70.0	12.2	66.2	10.6	60.7	11.6	83.3	10.8
IPSyn subscale								
Verb Phrases	21.7	4.3	20.0	3.6	18.2	3.9	25.1	3.6
Noun Phrases	17.6	2.3	17.5	1.7	16.8	2.2	19.7	1.6
Questions/Negations	13.2	3.5	12.3	3.8	10.3	4.0	14.7	2.9
Sentence Structure	17.4	4.8	16.4	4.2	15.4	4.1	23.7	5.5

Note. FXS-O = Fragile X syndrome only group; FXS-ASD = Fragile X syndrome–autism spectrum disorder group; DS = Down syndrome group; TD = typically developing group; MLU = mean length of utterance; IPSyn = Index of Productive Syntax.

Table 2. Correlations among overall syntactic measures and nonverbal mental age.

Group	Variable	MLU	IPSyn Total	Leiter-R MA
FXS-O	MLU	–		
	IPSyn Total	.887***	–	
	Leiter-R MA	.426**	.473**	–
FXS-ASD	MLU	–		
	IPSyn Total	.783***	–	
	Leiter-R MA	.394*	.480**	–
DS	MLU	–		
	IPSyn Total	.929***	–	
	Leiter-R MA	.666***	.612***	–
TD	MLU	–		
	IPSyn Total	.835***	–	
	Leiter-R MA	.484***	.550***	–

Note. Leiter-R MA = Leiter International Performance Scale–Revised, Mental Age.

*$p < .05$. **$p < .01$. ***$p < .001$.

Total, $F(3, 136) = 29.49$, $p < .0001$. No interactions were significant.

The between-group differences, as well as adjusted means, for MLU and IPSyn Total are presented in Table 3. Post hoc comparisons indicated no differences between the FXS-O and FXS-ASD groups. Across both measures, however, TD boys scored higher than the FXS-O (d = 1.15 for MLU and 1.17 for IPSyn Total), FXS-ASD (d = 1.45 for MLU and 1.14 for IPSyn Total), and the DS (d = 2.15 for MLU and 1.91 for IPSyn Total) groups. The effect sizes for these differences are very large, all surpassing Cohen's recommendation of .8 for a large effect. Further, both the FXS-O (d = .75 for MLU and .67 for IPSyn Total) and FXS-ASD (d = .78 for MLU and .56 for IPSyn Total) groups scored significantly higher than the DS boys. Although these effects were smaller than for the comparisons involving the TD boys, all were moderate in size.

Table 3. Means adjusted for the Leiter-R mental age and maternal education.

Variable	FXS-O	FXS-ASD	DS	TD
Overall syntax				
MLU	3.5^{b}	3.4^{b}	2.8^{a}	4.7^{c}
IPSyn total	69.0^{b}	67.2^{b}	61.0^{a}	82.4^{c}
IPSyn subscales				
Verb Phrases	21.2^{b}	20.4^{b}	18.2^{b}	24.7^{a}
Noun Phrases	17.7^{b}	17.6^{b}	16.8^{b}	19.7^{a}
Questions/Negations	$13.2^{a,b}$	12.5^{b}	10.5^{c}	14.5^{a}
Sentence Structure	17.0^{b}	16.8^{b}	15.4^{b}	23.5^{a}

Note. Different superscripts in each row indicate significantly different means. If groups have the same letters, then differences were not significant.

*$p < .05$. **$p < .01$. ***$p < .001$.

Measures of specific aspects of syntax. As with the more general measures presented above, the models for all four subscale outcomes were significant after controlling for nonverbal mental age and maternal education levels: Verb Phrases, $F(12, 136) = 484.48$, $p < .0001$; Noun Phrases, $F(12, 136) = 1264.76$, $p < .0001$; Questions and Negations, $F(12, 136) = 183.81$, $p < .0001$; Sentence Structure, $F(12, 136) = 20.38$, $p < .0001$. Diagnostic group was significant in all four models: Verb Phrases, $F(3, 136) = 20.38$, $p < .0001$; Noun Phrases, $F(3, 136) = 16.13$, $p < .0001$; Questions and Negations, $F(3, 136) = 8.05$, $p < .0001$; Sentence Structure, $F(3, 136) = 26.50$, $p < .0001$.

The adjusted group means and the between-group differences for the four subscales are presented in Table 3. The pattern of results was more complex than what was seen with the overall measures of syntax. With one exception, TD boys demonstrated higher performance than all other groups on all four subscales. That exception was with the FXS-O group on the Questions and Negations subscale, where the difference was nonsignificant. The TD boys scored higher than the boys with FXS-O on Verb Phrases (d = 1.09), Noun Phrases (d = 1.01), and Sentence Structure (d = 1.26). The TD boys scored higher than the boys with FXS-ASD on all four subscales: Verb Phrases (d = 1.20), Noun Phrases (d = 1.24), Questions and Negations (d = .60), and Sentence Structure (d = 1.38). The TD boys also scored higher than the boys with DS on all of the subscales: Verb Phrases (d = 1.73), Noun Phrases (d = 1.50), Questions and Negations (d = 1.15), and Sentence Structure (d = 1.67). There was a moderate effect size for the difference between the TD and FXS-ASD groups on the Questions and Negations subscale. All other effect sizes for comparisons with TD were greater than 1.0, indicating very large effects.

Across all four subscales, there were no significant differences between the two FXS groups. The boys with DS scored lower on the Questions and Negations subscale than the boys with FXS-O (d = .72) and the boys with FXS-ASD (d = .52), showing moderate effect sizes. The DS group did not differ from either FXS group on the other subscales.

Discussion

In this investigation, we compared the syntactic skills of boys with FXS without ASD, boys with FXS with ASD, boys with DS, and TD boys. In all of our analyses, we controlled for nonverbal mental age and maternal education levels. We found that the boys with FXS with

and without ASD and the boys with DS had lower overall syntactic skills (as measured by MLU and IPSyn Total scores) than the TD boys. The DS group also scored lower than both FXS groups on these overall syntactic measures, and the two FXS groups did not differ. When we compared the groups' performances on the more narrow measures of Noun Phrases, Verb Phrases, Questions and Negations, and Sentence Structure, we found that the FXS-ASD and DS groups scored lower than the TD group across all four of these subscales. The FXS-O group scored lower than the TD group on three of the subscales: Noun Phrases, Verb Phrases, and Sentence Structure. Interestingly, the FXS-O and TD groups did not differ on the Questions and Negations subscale. The two FXS groups did not differ on any of the subscales, and both scored higher on the Questions and Negations subscales than the DS group.

These findings indicate that on nearly all measures of expressive syntax, boys with FXS, regardless of autism status, score lower than would be expected based on their nonverbal mental ages. The one exception was on the Questions and Negations subscale, where boys with FXS-O did not differ from TD boys. These findings are consistent with those reported by Roberts, Hennon, and colleagues (2007), whose participants overlapped considerably with those in the current study, and Paul and colleagues (1984). However, our findings conflict with those of other researchers, such as Sudhalter and colleagues (1991, 1992), who found that the expressive syntactic skills of individuals with FXS were similar to those of younger TD children. As noted in the literature review, methodological differences in the studies may account for these differences in findings. Our participants were somewhat younger than those in the studies conducted by Sudhalter et al. (1991, 1992). We also strictly controlled for nonverbal developmental levels and maternal education levels. Our finding that boys with FXS-O and TD boys scored similarly on the Questions and Negations subscale repeats the finding reported by Roberts, Hennon, et al. (2007), whose FXS-O and TD samples included many of the children studied in the current investigation. Anecdotal reports suggest that individuals with FXS engage in frequent question-asking. Perhaps this behavior is related to relatively spared syntactic construction of questions and negations. Alternatively, the Questions and Negations subscale of the IPSyn contains fewer items than other subscales and may be less sensitive to group differences.

We were somewhat surprised that the FXS groups did not differ according to autism status, given previous findings that some children with autism (but not FXS) demonstrate deficits on standardized measures of syntax when compared with controls matched on both chronological and mental age (Kjelgaard & Tager-Flusberg, 2001; Landa & Goldberg, 2005) and that overall expressive language skills have been found to be lower in children (Philofsky et al., 2004) and adolescents (Lewis et al., 2006) with comorbid FXS and autism than in individuals with FXS only. However, the participants in the studies of children with autism but not FXS were high-functioning, whereas our participants with FXS and ASD also had intellectual disabilities. For the boys with FXS in the current investigation, the presence of ASD does not seem to additionally impact syntactic production. Our findings complement those of Lewis et al. (2006), who found that the overall expressive language skills of adolescent boys with FXS (who also had intellectual disabilities) did not differ according to autism status.

Our findings also indicated that the expressive syntactic skills of boys with DS were lower than would be expected based on nonverbal mental age, adding to the well-established body of research that suggests that individuals with DS demonstrate particular difficulty with expressive syntax (Abbeduto & Chapman, 2005; Chapman, 2006; Chapman & Hesketh, 2000; Laws & Bishop, 2003; Rice et al., 2005). Notable deficits in expressive syntax have been reported for adolescents with DS using both standardized assessments and conversational and narrative language samples (see Chapman & Hesketh, 2000, for a review). Eadie and colleagues (2002) have found similar syntactic deficits in the conversational language samples of younger children with DS (M = 7 years of age).

We also found some differences in expressive syntax between the FXS and DS groups, indicating that each group has a unique language profile. On MLU and IPSyn Total scores, the DS group scored lower than both FXS groups. On the IPSyn subscales, the three groups performed differently only on the Questions and Negations subscale, with the DS group scoring lower than both of the FXS groups. Boys with DS appear to have lower overall syntactic abilities and produce less complex questions and negations than boys with FXS. Recent studies of the pragmatic (Roberts, Martin, et al., 2007), phonological (Roberts et al., 2005), and receptive language (Abbeduto et al., 2003; Price et al., 2007) skills of individuals with DS and individuals with FXS support distinct communication profiles for the two groups, with individuals with DS showing greater deficits in phonology and receptive language but individuals with FXS showing greater deficits in pragmatics.

We also examined the correlations between overall syntactic measures and nonverbal cognitive levels. Syntax was correlated with nonverbal cognitive levels for all groups; however, the strongest correlations were for the TD and DS groups, who had the most advanced and least advanced syntactic skills, respectively. These results suggest that language development is tied to nonverbal cognitive development; however, the relatively weaker correlations between syntax and nonverbal cognitive levels for the FXS groups indicate that syntactic development

may be less related to nonverbal cognitive development for these groups than for other groups of children. When we examined the relationship between overall receptive language and nonverbal cognition, we found similar results—that is, that nonverbal cognition and language were related for children with FXS, children with DS, and TD children but to a lesser degree for children with FXS compared with the other groups (Price et al., 2007). Perhaps this pattern indicates that the underlying mechanisms of language and cognitive development for children with FXS differ from those of children with DS and TD children.

Clinical Implications

Several implications for assessment and intervention can be drawn from our study. The IPSyn appears to be a useful tool for assessing syntactic production for individuals with FXS or DS and may also be appropriate for children with other types of disabilities. The IPSyn profiles syntax skills in four areas (noun phrases, verb phrases, questions/negations, and sentence structure), allowing for identification of specific syntactic intervention targets. Because overall syntactic skills are below developmental expectations for children with FXS and children with DS, expressive syntax is an important area to assess and treat for children with both of these disorders. Children with DS and children with FXS-ASD scored lower than the TD children on all overall measures of syntax and all of the specific subscales of the IPSyn, indicating that all areas of syntax may warrant intervention. Scores on the IPSyn subscales indicated that basic syntactic skills had been mastered, suggesting that relatively more advanced syntactic structures (e.g., noun phrases, verb phrases, questions and negations, sentence structure) be targeted in intervention. Examples of targets for noun phrases might include producing multiword noun phrases and using adjectives in noun phrases. Verb phrase targets might incorporate production of modals, third person singular *–s,* and past tense *–ed*. Targets for questions and negations might include production of *wh–* questions with inverted auxiliaries and use of negative auxiliaries. In addition, more advanced sentence structures such as use of infinitive phrases and conjoined phrases might be targeted. For children with FXS-O, the syntactic structure of questions and negations appears to be relatively spared; therefore, intervention efforts may best target other areas of syntactic development (i.e., noun phrases, verb phrases, and sentence structure).

Strengths and Limitations

A primary strength of the current study is the large sample sizes of children with FXS, children with DS, and TD children. We also compared the syntactic skills of children with FXS with and without ASD, a comparison not previously made in the literature. We controlled for the effects of nonverbal cognition and maternal education levels. We also used overall measures of syntax as well as more specific indices of noun phrase, verb phrase, question/negation, and sentence structure development. It would be interesting, in future studies, to explore even more specific strengths and weaknesses in syntax, such as those of particular grammatical morphemes and structures. Our study was limited in that only two syndromes were compared. In future studies, other comparison groups, particularly children with autism (without FXS), should be included to further determine which characteristics of FXS are shared by autism or other disabilities. The inclusion of only boys with FXS was also a limitation of our study. Future studies should also investigate the syntactic production of girls with FXS. Whereas the current study investigated syntactic skills at only one time point, future research should investigate growth in syntactic skills over time in children with FXS with and without autism. Finally, our study examined syntactic skills elicited during conversational language samples. Although this procedure may most closely simulate the everyday interactions in which children participate, there is evidence that narrative language samples elicit more advanced syntax in adolescents and young adults with DS and other forms of intellectual disability (Abbeduto, Benson, Short, & Dolish, 1995; Miles, Chapman, & Sindberg, 2006). Future studies should consider whether this difference is also found for younger children with DS, FXS, and other types of disabilities. Exploration of the IPSyn as a tool for analysis of syntax produced during narrative samples is also warranted.

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