10-1-2014

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Publication Info

Preprint version *Journal of Speech, Language and Hearing Research*, Volume 57, Issue 5, 2014, pages 1692-1707.

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Abstract

Purpose: Impaired pragmatic language (i.e., language use for social interaction) is a hallmark feature of both autism spectrum disorder (ASD) and fragile X syndrome (FXS), the most common known monogenic disorder associated with ASD. However, few cross-population comparisons of ASD and FXS have been conducted, and it is unclear whether pragmatic language profiles in these conditions overlap. *Method*: This study used semi-naturalistic and standardized assessment methods to characterize pragmatic language abilities of 29 school-aged boys with idiopathic ASD, 38 with FXS and comorbid ASD, 16 with FXS without ASD, 20 with Down syndrome and 20 with typical development. *Results*: Similar severity of pragmatic language deficits was observed in both of the groups with ASD (idiopathic and fragile X-associated). ASD comorbidity had a detrimental effect on the pragmatic language skills of boys with FXS. Some different patterns emerged across the two pragmatic assessment tools, with more robust group differences observed in pragmatics assessed in a semi-naturalistic conversational context. *Conclusions*: These findings have implications for pragmatic language assessment and intervention, as well as for understanding the potential role of the fragile X gene, *Fragile X Mental Retardation-1*, in the pragmatic language phenotype of ASD.

Keywords: pragmatic language, social communication, autism, fragile X syndrome, discourse, endophenotype

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Pragmatic language competence, or the ability to use language in social contexts, is critical for supporting fluent social interactions (Bates, 1976; McTear & Conti-Ramsden, 1992; Prutting, 1982). Examples of pragmatic language skills include the selection of conversational topics fitting to the situation, appropriate word choice, and the ability to modify language in order to match the expectations and knowledge base of the communication partner. Problems using language in these ways limit participation in social interaction, potentially impacting other developmental domains (Chapman, 2000; Dickinson & McCabe, 1991; Fogel, 1993; Hewitt, 1998; McTear & Conti-Ramsden, 1992; Yoder & Warren, 1993). Thus, pragmatic language ability is a critical skill for social functioning and learning.

Pragmatic language impairment is a central characteristic of both autism spectrum disorder (ASD) and fragile X syndrome (FXS), two genetically-based neurodevelopmental disabilities that show substantial phenotypic overlap. Whereas autism is behaviorally defined, FXS is caused by an identifiable genetic mutation on the *Fragile X Mental Retardation-1 (FMR1)* gene on the X chromosome (Cohen, Pichard, & Tordjman, 2005). Understanding the precise phenotypic overlap between these disorders may therefore offer insights into the role of *FMR1* as a candidate gene in particular features of autism. It is unclear, however, whether pragmatic language profiles are similar in individuals with idiopathic ASD and FXS, as few studies cross-population comparison studies have been conducted. This study aimed to define overlapping and syndrome-specific pragmatic profiles in ASD and FXS through a comparison of boys with idiopathic ASD, boys with ASD and FXS, and boys with FXS only, across two different language contexts: a standardized assessment of pragmatic language and a semi-naturalistic conversational task. Below, ASD and FXS are briefly described as disorders of a neurogenic basis, followed by a review of pragmatic language impairment in these disorders and importance of context in assessing pragmatic language.

Autism Spectrum Disorder

ASD is a serious, lifelong disability that affects approximately 1 in 88 children (CDC, 2012). The

diagnosis of ASD is determined behaviorally by the presence of social and communication impairments, as well as repetitive or restricted behaviors (American Psychiatric Association, 2013). Converging evidence from twin, family, and molecular-genetic studies supports a large genetic component in the etiology of ASD (Devlin & Scherer, 2012; Miles, 2011). In spite of clear evidence for genetic underpinnings, ASD is both clinically and etiologically heterogeneous, which has slowed progress in mapping risk loci (Geschwind, 2011; Ronald & Hoekstra, 2011). The study of ASD within the context of associated monogenic conditions, such as FXS, has been proposed as a method for reducing etiologic complexity in the search for ASD genes (e.g., Abrahams & Geschwind, 2010; Belmonte & Bourgeron, 2006; Hagerman, Narcisa, & Hagerman, 2011). Such an approach provides a simplified genetic context from which to identify core features that are shared across etiologic subtypes of ASD, and which may be linked to a traceable genetic cause.

Fragile X Syndrome

FXS occurs in as many as 1 in 2,500 individuals (Fernandez-Carvajal et al., 2009; Hagerman, 2008) and is the most common known genetic disorder associated with ASD (Cohen et al., 2005). Unlike ASD, the genetic basis of FXS is relatively well-understood; it is caused by an expanded number of Cytosine-Guanine-Guanine (CGG) nucleic acid repeats on the *FMR1* gene (Hagerman & Hagerman, 2002). When the CGG expansion exceeds 200 copies, the gene methylates, or shuts down, and stops producing Fragile X Mental Retardation Protein (FMRP), which is a protein needed for brain development and functioning (Bassell & Warren, 2008; Irwin, Galvez, Weiler, Beckel-Mitchener, & Greenough, 2002; Weiler & Greenough, 1999). Because FMRP normally acts as a translator for other proteins, its absence has widespread consequences for the normal functions of other genetic pathways. Many protein systems that become dysregulated in the absence of FMRP have also been implicated in ASD, and it is through this interaction with other genes that the *FMR1* mutation increases risk for ASD (Hagerman, Au, & Hagerman, 2011; Hagerman, Hoem, & Hagerman, 2010).

Some ASD-like features are seen in almost all individuals with FXS (Hagerman et al., 2010). Assessment using gold-standard ASD diagnostic tools shows that 60-75% of individuals with FXS meet

the diagnostic criteria for ASD (Clifford et al., 2007; Hall, Lightbody, & Reiss, 2008). While it is clear that ASD and FXS share significant behavioral overlap, the nature of ASD in FXS is controversial. It has been hypothesized that idiopathic and fragile X-associated ASD stem from divergent underlying mechanisms, with fragile X-specific anxiety or intellectual disability underlying the ASD phenotype of FXS (Cohen, 1995; Cohen, Vietze, Sudhalter, Jenkins, & Brown, 1989; Hall, Lightbody, Hirt, Rezvani, & Reiss, 2010). While this theory has not been supported, with cross-population comparison studies failing to detect unique ASD symptom profiles in idiopathic and fragile X-associated ASD (Bailey et al., 1998; Dissanayake, Bui, Bulhak-Paterson, Huggins, & Loesch, 2009; Rogers, Wehner, & Hagerman, 2001), few studies have conducted a more fine-grained analysis of select ASD-associated features in these two neurodevelopmental conditions, which might reveal syndrome-specific profiles.

Pragmatic Language in Autism Spectrum Disorder and Fragile X Syndrome

The present study conducted a focused investigation of pragmatic language ability in ASD and FXS. As noted previously, pragmatic impairment is seen universally in ASD and is also a well-documented feature of FXS (Keysor & Mazzocco, 2002; Landa, 2000; Sudhalter & Belser, 2001; Tager-Flusberg, Paul, & Lord, 2005). During conversation, individuals with ASD struggle with turn-taking (Capps, Kehres, & Sigman, 1998; Paul et al., 1987), maintaining topics (Adams, Green, Gilchrist, & Cox, 2002; Tager-Flusberg & Anderson, 1991), and choosing appropriate diction (Ghaziuddin & Leonore, 1996). Perseveration (Ross, 2002), irrelevant details (Paul, Orlovski, Marcinko, & Volkmar, 2009), and unclear references (Fine, Bertolucci, Szatmari, & Ginsberg, 1994) are also exhibited in the conversation of individuals with ASD. Communicative repair is affected as well, with difficulties in adequately responding to the clarification requests of others (Geller, 1998; Volden, 2004). The narrative (i.e., storytelling) abilities of individuals with ASD also show atypical pragmatic features, such as the inclusion of inapropriate or irrelevant story components (Diehl, Bennetto, & Young, 2006; Loveland, McEvoy, & Tunali, 1990) and ambiguous references (Norbury & Bishop, 2003). When narrating, individuals with ASD fail to provide causal explanations to describe actions, experiences, and emotions (Capps, Losh, & Thurber, 2000; Diehl et al., 2006; Losh & Capps, 2003; Tager-Flusberg, 1995) and have difficulty

adopting the perspectives of others (García-Pérez, Hobson, & Lee, 2008). Because subclinical pragmatic difficulties are also seen in relatives of individuals with ASD as part of the broad autism phenotype, pragmatic impairment is hypothesized to represent a genetically meaningful trait marking vulnerability to ASD (Landa et al., 1992; Losh, Childress, Lam, & Piven, 2008; Piven, Palmer, Landa, Santangelo, & Childress, 1997).

Individuals with FXS also struggle with pragmatic language, such as impaired use of communicative repair strategies (Abbeduto et al., 2008), difficulty with conversational topic maintenance (Roberts, Martin, et al., 2007; Sudhalter & Belser, 2001; Sudhalter, Cohen, Silverman, & Wolf-Schein, 1990; Wolf-Schein et al., 1987), and impaired narrative processing and formulation (Estigarribia et al., 2011; Simon, Pennington, Taylor, & Hagerman, 2001). Perseveration has been documented at length (Belser & Sudhalter, 2001; Martin et al., 2012; Roberts, Martin, et al., 2007; Sudhalter et al., 1990; Wolf-Schein et al., 1987). While it is clear that individuals with FXS struggle with pragmatics, the impact of ASD comorbidity on such deficits is less well understood, as most investigations have not accounted for co-occurring ASD. Some evidence suggests that ASD comorbidity has an added detrimental effect on pragmatic language abilities; boys with FXS and comorbid ASD exhibit more off-topic conversational turns (Roberts, Martin, et al., 2007), increased stereotyped and perseverative language (Martin et al., 2012; McDuffie et al., 2010), and perform more poorly on standardized pragmatic language assessments (Losh, Martin, Klusek, Hogan-Brown, & Sideris, 2012) than their counterparts without ASD.

Importantly, pragmatic language impairment in FXS is associated with *FMR1*-related molecular variation (CGG repeat length and percent methylation), providing a direct link between *FMR1* and pragmatic impairment (Losh, Martin, et al., 2012). Furthermore, premutation carriers of *FMR1* show subclinical pragmatic features that are similar in quality and severity with those seen in the broad autism phenotype, which supports a role of *FMR1* in a broad range of pragmatic language features associated with ASD (Losh, Klusek, et al., 2012). Thus, the delineation of pragmatic language features that may overlap or diverge in ASD and FXS is a promising method for identifying phenotypic commonalities that may stem from biological disruptions associated with *FMR1*.

Comparisons of Pragmatic Language in ASD and FXS

Only a handful of studies have directly compared pragmatic language features across ASD and FXS, with mixed results. A few early investigations compared select pragmatic features as they occurred in spontaneous or elicited language samples, although it is difficult to draw definitive conclusions from this work given the inconsistent handling of ASD comorbidity in FXS. For example, one research group analyzed the spontaneous conversation of males with idiopathic ASD and FXS (ASD status not reported) to find that males with FXS showed more repetitive speech (Belser & Sudhalter, 2001) and tangential language (Sudhalter & Belser, 2001) than males with idiopathic ASD. In another study, Sudhalter et al. (1990) compared the spontaneous language of individuals with ASD and individuals with FXS (who did not have ASD) and found that the individuals with ASD exhibited increased echolalia, whereas those with FXS showed more frequent perseveration. Although these reports offer some evidence that the pragmatic violations committed by individuals with ASD and FXS differ qualitatively, the inconsistent characterization of ASD across reports hinders our ability to understand the impact of ASD comorbidity on the pragmatic language skills of individuals with FXS.

A recent investigation by Losh et al. (2012) highlights the importance of multimodal pragmatic language assessment. This study examined pragmatic language abilities among boys with idiopathic ASD and boys with FXS who did or did not have comorbid ASD, using a standardized measure of pragmatic language (the Pragmatic Judgment subtest of the Comprehensive Assessment of Spoken Language; CASL-PJ; Carrow-Woolfolk, 1999) and a teacher-report measure of communication skills (the Children's Communication Checklist-2; CCC-2; Bishop, 2006). Performance of the groups with idiopathic ASD and FXS with comorbid ASD was similar on the CASL-PJ, and impaired relative to the group with FXS without ASD (whose performance was similar to that of boys with Down syndrome and boys with typical development). Conversely, the boys with idiopathic ASD, FXS with ASD, and FXS without ASD were all rated to have similar overall severity of pragmatic difficulties using the CCC-2. This research highlights the need to integrate multiple sources of pragmatic language information, as patterns differed across direct assessment and teacher-reported measures.

Rationale for Present Study

Two key gaps in the literature remain in understanding the potential overlap in the pragmatic profiles of ASD and FXS. First, ASD comorbidity has been inconsistently accounted for in prior studies of FXS, making it difficult to determine whether the pragmatic difficulties in this population might be better attributed to the presence of ASD than to FXS-related processes. The present study aimed to address this question by comparing pragmatic language profiles of boys with FXS who did and did not have ASD to boys with idiopathic ASD. Two control groups were included: boys with Down syndrome (DS) and boys with typical development (TD). The group with DS was included as an intellectual disability comparison group to determine whether pragmatic language features might be attributable to general cognitive delays rather than to processes specific to ASD or FXS. Younger boys with TD (who were similar in language age to the disability groups) were included to provide a benchmark for expectations of pragmatic competence at this developmental level, necessary for evaluating the performance of the disability groups.

Second, few studies have incorporated multiple sources of information in the assessment of pragmatics in ASD or FXS, despite evidence that a multi-method approach to language assessment informs convergent validity and improves diagnostic accuracy (Brewer & Hunter, 2006). Incorporating evidence from both standardized and naturalistic assessments of pragmatic language may provide a more comprehensive portrait of social-communication skills in these populations, as well as inform how the choice of assessment technique may account for inconsistencies across prior reports. While standardized pragmatic language assessment tools offer many advantages (e.g., quick administration, norm-referencing), the decontextualized form of standardized tools is thought to limit generalizability to real-life settings (e.g., Adams, 2002; Prutting & Kittchner, 1987). In fact, the highly structured format of standardized assessments may over-estimate pragmatic skills in individuals with ASD, who are known to perform better in contexts where there is less need to interpret contextual information (Clark & Rutter, 1981; Loukusa et al., 2007; Williams, Goldstein, & Minshew, 2006). Thus, naturalistic measures are considered the gold standard method for pragmatic language assessment (Adams, 2002; Hyter, 2007;

McTear & Conti-Ramsden, 1992; Prutting & Kittchner, 1987; Roth & Spekman, 1984). However, naturalistic approaches are not without drawbacks, as they tend to be time-intensive to implement. This study incorporated both standardized and semi-naturalistic pragmatic language assessments to capture comprehensive, ecologically valid pragmatic language phenotypes of ASD and FXS, which may inform syndrome specificity. A *semi*-naturalistic measure, rather than a naturalistic measure, was used to improve efficiency and to ensure that the language sampling context was relatively similar across groups.

Method

Participants

Study participants included 29 boys with idiopathic ASD (ASD only; ASD-O), 38 boys with FXS and comorbid ASD (FXS-ASD), 16 boys with FXS without ASD (FXS-only; FXS-O), 20 boys with Down syndrome (DS), and 20 boys with typical development (TD) who were of a similar language age. The mean chronological age of the disability groups was 11.31 years (SD 3.32, range 3.18-17.93), and the mean age of the boys with TD was 4.82 (*SD* 1.00, range 3.54-6.69); see Table 1. Only boys participated in the study because females with FXS are generally less affected than males and less likely to have ASD (Clifford et al., 2007; Hagerman & Hagerman, 2002).

Participants were regularly using phrases of at least three words and English was the primary language spoken at home. Pure-tone hearing thresholds were screened at 500, 1000, 2000 and 4000 Hz with a MAICO MA 40 audiometer; children were excluded for failing the screener at 30 dB in the better ear. All boys with FXS had a diagnosis of the full mutation, and were recruited without reference to ASD-status. ASD was ruled out in the boys with DS and TD using the Autism Diagnostic Observation Schedule (ADOS; Lord, Rutter, DeLavore, & Risi, 2001). Four boys with DS were initially recruited but later dropped from the study after scoring above diagnostic cutoffs for ASD. Boys with TD with a history of developmental or language delays were excluded. In addition, boys with TD were dropped from the study if they scored below 1.5 standard deviations of the mean on the standardized vocabulary and cognitive assessments described below. This cut-off was chosen because it is a threshold commonly used in clinical settings to identify developmental delays (Spaulding, Plante, & Farinella, 2006). Recruitment

was based in the Eastern and Midwestern regions of the United States. Participants were ascertained through advertisement at genetic clinics, parent support groups, physicians' offices, and through the Research Participant Registry Core of the Carolina Institute for Developmental Disabilities at the University of North Carolina at Chapel Hill.

Participants were drawn from a larger pool of children participating in an ongoing longitudinal study of pragmatic language in FXS, which has been described previously (Losh, Martin et al., 2012). Participants were selected for inclusion in the present study if they had available data for the diagnostic, cognitive, and language assessments of interest (described below). Given the longitudinal design of the larger study, in some instances data from several different time points were available for a given participant. In these cases, data were chosen to optimize group-level matching on vocabulary, according to a raw score composite of the Peabody Picture Vocabulary Test-III (PPVT; Dunn & Dunn, 1997) and the Expressive Vocabulary Test (EVT; Williams, 1997). Matching was based on vocabulary (as opposed to cognitive ability) in order to examine pragmatic deficits above and beyond what could be attributed to general language skills. To achieve group matching, two boys with ASD-O were excluded because they had receptive or expressive vocabulary scores that fell above the range of the other groups. The groups did not differ on receptive or expressive vocabulary (*ps* > .400), although there were some differences in mean length of utterance and nonverbal cognitive ability across groups (see Table 1). Vocabulary, mean length of utterance, and nonverbal mental age were co-varied in analyses. The groups were similar in race, household income, and maternal education level (*ps* > .112); see Table 2. The standardized pragmatic language data for forty-two children (5 with ASD-O, 9 with FXS-ASD, 3 with FXS-O, 7 with DS, 18 with TD) have been previously analyzed by Losh et al. (2012).

Procedures

Assessments were administered as part of a broader research protocol, which lasted approximately 4-6 hours, including time for breaks. Testing took place in a university-affiliated research laboratory, the child's school, or in a quiet room in the child's home. Procedures were approved by the

Institutional Review Boards of the University of North Carolina at Chapel Hill and Northwestern University.

Characterization of ASD. The Autism Diagnostic Observation Schedule (ADOS; Lord et al., 2001) was administered to confirm clinical diagnoses of ASD in the boys with ASD-O, and to determine ASD comorbidity among the boys with FXS. The ADOS involves direct observation of social-communicative and restricted/repetitive behaviors during a play-based, semi-structured interaction between the participant and an examiner. The ADOS was coded by examiners who had achieved reliability either through direct training with the developers of the ADOS or through intra-lab reliability in accordance with the standards of the instrument developers. All boys in the groups with ASD (ASD-O and FXS-ASD) met diagnostic criteria for "autism" or "autism spectrum" on the revised diagnostic algorithms(Gotham et al., 2008; Gotham, Risi, Pickles, & Lord, 2007). Eleven of the children with FXS had been administered the ADOS at three independent time points through participation in related longitudinal studies of language and speech characteristics that followed the same cohort of boys at younger or older ages (Roberts, Hennon, et al., 2007; Zajac, Harris, Roberts, & Martin, 2009). All available diagnostic information was considered in determining ASD status in attempt to determine the best-estimate diagnosis. Four boys met criteria for ASD at 3/3 time points and three met criteria at 2/3 time points; these boys were characterized as FXS-ASD. Two boys scored as non-spectrum at 3/3 time points and two scored as non-spectrum at 2/3 time points; these boys were assigned to the FXS-O group. The best-estimate diagnosis agreed with concurrent ADOS classification for all but one of the boys, who had met diagnostic criteria for ASD at the concurrent assessment but had scored below diagnostic thresholds at two earlier time points. Concurrent ADOS diagnostic information was used for the remaining participants. The ADOS was also used as a continuous measure of ASD symptoms; severity scores were computed in accordance with Gotham, Pickles, and Lord (2009). For those participants who had been administered the ADOS several times, severity scores from all available time points were averaged to compute a best-estimate autism severity score. This included the 11 boys with FXS discussed above, as well as six boys with DS and one with boy with TD.

Cognitive ability. Nonverbal cognitive ability was assessed using the Brief IQ Composite of the Leiter International Performance Scale-Revised (Leiter-R; Roid & Miller, 1997). Age equivalent scores were used in analysis.

Receptive and expressive language. Receptive and expressive vocabulary were measured with age equivalent scores from the Peabody Picture Vocabulary Test-III (PPVT; Dunn & Dunn, 1997) and the Expressive Vocabulary Test (EVT; Williams, 1997), respectively. In addition to standardized vocabulary measures, mean length of utterance in morphemes (MLU) was computed as an index of morphosyntactic development in young children (Brown, 1973). Systematic Analysis of Language Transcripts (SALT; Miller & Chapman, 2008) conventions were used to transcribe 55 intelligible child conversational turns from "play" and 55 from "non-play" contexts (e.g., conversation) occurring during the ADOS. This strategy ensured that the context of the language sample was comparable across groups, who varied in "talkativeness". The language samples were transcribed by trained research assistants who had achieved morpheme agreement of 80% or higher as compared to a gold standard transcript for two samples from each diagnostic group. Consistent with SALT conventions, MLU was calculated on novel, intelligible, non-routine utterances.

Standardized assessment of pragmatic language. The Pragmatic Judgment subtest of the Comprehensive Assessment of Spoken Language (CASL-PJ; Carrow-Woolfolk, 1999) was used as a standardized measure of the knowledge and use of pragmatic language. In the CASL-PJ, participants are told short stories about children in various social situations and are scored on their ability to provide a pragmatically appropriate response explaining what the children should do or say in each scenario. Specific pragmatic behaviors assessed in the CASL-PJ include communicative intent, turn taking, emotional expression, and pragmatic appropriateness. The CASL-PJ is normed on individuals aged 3-21 years, and is a reliable index of pragmatic language ability with test-retest reliability coefficients ranging from .66-.85 across age groups. Age equivalent scores were used in analysis, which are preferable to raw scores as they are on an equal-interval scale. A higher score age equivalent score indicates more advanced skills.

Semi-naturalistic assessment of pragmatic language. The Pragmatic Rating Scale-School Age (PRS-SA; Landa, 2011) was used to rate pragmatic language behaviors occurring during semi-structured social interaction. The ADOS, which generally lasts 40-60 minutes, was used as a semi-naturalistic conversational context from which to rate pragmatic language skills. The ADOS is an ideal context for sampling conversation because the semi-structured format provides continuity across administrations but is flexible in following the child's lead (Tager-Flusberg et al., 2009). Eighty-eight children were administered the ADOS module 3 (for verbally fluent individuals), and 35 were administered module 2 (for use with individuals who have phrase speech). There was a roughly equal distribution of both modules across diagnostic groups.

The PRS-SA assesses 34 features related to pragmatic language, such as verbosity, social appropriateness, scripting, redundancy, failure to initiate topics, reduced communicative intent, inadequate turn taking, and the use of nonverbal behaviors such as eye contact and communicative gestures. Items are rated for severity on a scale of "0" to "2" according to operational definitions of each trait. Items are summed to produce a total score, with a higher score indicating greater severity of pragmatic language difficulties. The PRS-SA also provides five theoretically-derived subdomain scores, which were explored in this study with factor analysis (described in *Data Reduction*). All PRS-SA ratings were conducted by the first author (JK), who had achieved reliability with the developer of the PRS-SA. The coder was blind to the diagnosis of 86% of participants (it was not possible to maintain blinding to all participants, as the coder had assisted with recruitment and testing for the study). 15% of the sample was randomly selected and second-scored by an independent, blind rater who had also achieved coding reliability with the developer of the PRS-SA. Inter-rater reliability was as follows: Intraclass Correlation Coefficient (ICC: 3, 2): .96 for the overall sample, .74 for ASD-O, .83 for FXS-ASD, .73 for FXS-O, .89 for DS, and .84 for TD. ICC values of 0.40- 0.75 represent "fair" to "good" agreement, and values greater than .75 signify "excellent" agreement (Fleiss, Levin, & Paik, 2004; Landis & Koch, 1977).

Data Reduction and Analysis

To support the use of the theoretically-derived pragmatic subdomains of the PRS-SA, exploratory factor analysis was conducted with PASW Statistics 18 (2009). The model was fit under weighted least squares estimation with a geomin rotation. Items were treated as categorical variables, and polychoric correlations were used to produce the asymptotic covariance matrix for analysis (Joreskog, 1994). Examination of the scree plot showed a distinct leveling after the first factor, and eigenvalues for the first eight factors were greater than one, with a significant drop from the first to second factor (6.10 to 3.01). A one-factor model provided the most theoretically meaningful constructs. Thus, results suggested that a one-factor model was the best fit for the data. A confirmatory factor analytic model was then conducted in Mplus (Muthen & Muthen, 2006) to determine whether the data might converge on the pre-identified subscales. Like the exploratory model, the confirmatory factor analysis was fit under weighted least squares estimation with a geomin rotation, with polychoric correlations to handle the categorical items. The model using the pre-identified subscales as factors failed to converge, and even the preliminary solution indicated high between-factor correlations, implying that the one-factor solution was sufficient. Therefore, group comparisons on the PRS-SA subscales are not presented.

Prior to analyses, data were first examined for skewedness, kurtosis, and heteroscedasticity; no corrections were necessary. For the group comparisons on pragmatic language, two separate sets of analyses were run in order to account for ASD symptoms in FXS either categorically or continuously. First, group differences on the pragmatic language variables were examined with ASD status in the boys with FXS considered as a categorical trait (i.e., the group was divided into subgroups of boys with FXS-ASD and FXS-O, as described previously). For these models, multivariate analysis of covariance (MANCOVA) was used to test whether the PRS-SA and CASL-PJ scores differed by group, controlling for nonverbal mental age (Leiter-R), receptive vocabulary (PPVT), expressive vocabulary (EVT), and morphosyntactic skills (MLU). Independent samples *t*-tests indicated that there were no significant differences between the PRS-SA scores of participants who had been administered the ADOS module 2 versus module 3 ($p = .192$) and thus module type was not controlled for in group comparisons. Planned pair-wise comparisons were conducted to test for specific group differences. False discovery was

controlled for by adjusting at the level of the omnibus *F*-test, using the Benjamini-Hochberg correction procedure (Benjamini & Hochberg, 1995).

The second set of analyses took a continuous approach to account for ASD, using a series of linear regression analyses to explore the ADOS severity score as a unique predictor of pragmatic language ability in the groups with ASD-O and FXS-all (i.e., the full FXS sample). Nonverbal mental age, receptive and expressive vocabulary, MLU, and autism severity were entered in a hierarchical fashion into two different regression models predicting PRS-SA and CASL-PJ performance. These analyses were conducted only in the groups with ASD-O and FXS because of the limited range of autism severity scores in the groups with DS and TD.

Although factor analysis did not support the use of the PRS-SA subscales to examine pragmatic subdomains, additional analyses were conducted to explore possible group differences at the item level. MANCOVA was used to test the effect of group on each of the PRS-SA items, controlling for nonverbal mental age (Leiter-R), and language abilities (PPVT, EVT, and MLU). The Benjamini-Hochberg correction procedure was applied at the level of the omnibus *F*-test to correct for multiple comparisons (Benjamini & Hochberg, 1995). Finally, simple Pearson correlations were conducted between the PRS-SA and CASL-PJ to examine the relationship between the measures.

Results

Group Comparisons on Pragmatic Language Total Scores

Descriptive statistics presenting uncorrected group means, standard deviations, and ranges on the PRS-SA and CASL-PJ are presented in Table 3. MANCOVA revealed a significant group effect on the PRS-SA and CASL-PJ scores [Pillai's Trace = 0.37, F (8, 214) = 16.98, $p < .001$]. Follow-up univariate analysis testing the specific effect of group on PRS-SA showed a significant overall effect [F (4, 117) = 42.78, $p < .001$]. Post-hoc comparisons indicated that the boys with ASD-O and FXS-ASD showed similar severity of pragmatic impairment on the PRS-SA ($p = .248$, $d = .04$), which was significantly elevated in comparison to all other groups. The PRS-SA scores of the boys with ASD-O were higher (indicating greater impairment) than those of boys with FXS-O ($p < .001$, $d = .31$), DS ($p < .001$, $d =$

1.18), and TD ($p < .001$, $d = 1.92$). The boys with FXS-ASD also had significantly elevated scores compared to boys with FXS-O ($p < .001$, $d = .31$), DS ($p < .001$, $d = .47$), and TD ($p < .001$, $d = .98$). The boys with FXS-O and DS did not differ on PRS-SA total score ($p = .277$, $d = .02$), and both groups showed significantly higher (i.e., more impaired) scores than the boys with TD ($p < .001$, $d = .85$ and $p = .002$, $d = .38$, respectively). Group comparisons are presented in Figure 1.

Follow-up univariate analyses of CASL-PJ performance also showed a significant overall effect for group [F (4, 117) = 3.35, $p = .013$]. Pair-wise group comparisons indicated that the boys with ASD-O had significantly lower (i.e., more impaired) CASL-PJ scores than did boys with TD ($p = .002$, $d = .27$) and FXS-O ($p = .015$, $d = .16$) but did not differ from the boys with FXS-ASD ($p > .395$, $d = .01$) or DS ($p = .350$, $d = .02$). Boys with FXS-ASD had significantly lower scores than those with TD ($p = .025$, $d = .08$), and lower mean scores than boys with FXS-O with the difference approaching significance ($p = .057$, $d = .16$). The difference between the scores of the boys with DS and TD approached significance ($p = .053$, $d = .18$), with better performance in the boys with TD. Group comparisons are presented in Figure 2.

Item-level Group Comparisons on the PRS-SA

MANCOVA indicated a significant effect for group [Pillai's Trace = 2.20, F (128, 328) = 3.12, $p < .001$]. Benjamini-Hochberg corrected follow-up comparisons showed significant group effects for a number of PRS-SA items; Table 4 presents pair-wise comparisons for individual items. Overall, item-level patterns mirrored the results of the group comparisons on the PRS-SA total score (i.e., the groups with ASD-O and FXS-ASD showed similar pragmatic language violations, which were more severe than those exhibited by the boys with FXS-O, DS, and TD). There were a few notable exceptions to this pattern. First, odd signaling of humor and inappropriate volume was observed more frequently in the boys with ASD-O than in those with FXS-ASD. Second, while the boys with FXS-O generally showed less pragmatic violations than the boys with ASD-O or FXS-ASD, scripting, inappropriate topic shifts, and atypical intonation occurred at a similar severity across the groups with ASD-O, FXS-ASD, and FXS-O (and were was impaired relative to the groups with DS and TD). Third, the boys with DS had the highest

ratings of stuttering/cluttering of all of the groups, and this item was coded more frequently in the groups with FXS than in those with ASD-O or TD. The groups with FXS-ASD and DS were less intelligible than all other groups. Finally, all of the disability groups performed similarly on the perseveration and rate of speech items, with impaired performance relative to the boys with TD. No significant group effects were detected for the following items: atypical vocal noises (p = .054), character speech (p = .074), vegetative sounds (p = .175), mismanagement of interpersonal space (p = .241), interrupting (p = .306), dysfluent language formulation (p = .381), grammar/vocabulary errors (p = .493), ability to clarify (p = .826), and formality (p = .875).

Autism Severity as a Predictor of Pragmatic Language Ability

PRS-SA. As depicted in Table 5, the combined effects of nonverbal mental age, receptive and expressive vocabulary, and MLU did not account for significant variance in the PRS-SA scores of the boys with ASD-O, but did account for 34% of the variance in PRS-SA performance among the boys with FXS. The addition of autism severity to the model accounted for significant unique variance in both groups; after accounting for cognitive and language factors, autism severity accounted for 35% of the variance in the PRS-SA score among the boys with ASD-O, and 33% of the variance among the boys with FXS.

CASL-PJ. The combined effects of nonverbal mental age, receptive and expressive vocabulary, and MLU ability accounted for significant unique variance in CASL-PJ scores in both ASD-O and FXS (see Table 6). The cognitive and language variables accounted for 70% of the variance in CASL-PJ scores of the boys with ASD-O and 78% of the variance in the scores of the boys with FXS. The addition of autism severity to the model did not account for additional unique variance in CASL-PJ in the group with ASD-O. In FXS, autism severity uniquely accounted for 6% of the variance in CASL-PJ beyond the effects of the cognition and language.

Relationship between Pragmatic Language Assessments

A significant, though moderately weak, association was detected between the PRS-SA total score and the CASL-PJ age equivalent score in the overall sample (r = -.31, p = .001), indicating that

performance on the CASL-PJ decreased as the severity of pragmatic language violations on the PRS-SA increased. Within-group correlations revealed similar patterns, with significant associations between PRS-SA and CASL-PJ in boys with FXS-O (r = -.50, p = .049), and the correlation approaching significance in boys with FXS-ASD (r = -.33, p = .065). Associations in the groups with ASD-O, DS, and TD followed a similar trend although correlations were not significant (ps >.130). Correlations are presented in Table 7.

Discussion

Boys with idiopathic ASD and FXS with ASD showed similar severity of pragmatic language impairments on both standardized and semi-naturalistic assessments, which builds on growing evidence of shared behavioral profiles in idiopathic ASD and ASD associated with the *FMR1* mutation (Bailey et al., 1998; Dissanayake et al., 2009; Rogers et al., 2001). While boys with FXS with and without ASD did not differ on the standardized pragmatic language measure, evaluation of pragmatic abilities in a semi-naturalistic setting using the PRS-SA revealed greater impairment in boys with FXS with comorbid ASD, suggesting that the presence of co-occurring ASD plays a significant role in these boys' abilities to communicate in real-life social contexts. Boys with DS, on the other hand, showed pragmatic language skills that were generally similar to those of the boys with FXS-O but more impaired than the boys with TD. Finally, whereas ASD severity accounted for a substantial proportion of variance on the PRS-SA for the ASD-O and FXS groups, language and cognitive variables were more predictive of performance on the CASL-PJ. The results of this study highlight the need for a multimodal approach to pragmatic language assessment in clinical settings, and underscore the importance of considering ASD comorbidity in the evaluation and treatment of individuals with FXS.

A number of prior reports have addressed symptom overlap in ASD and FXS using global measures of autism symptoms (such as the ADOS), with some reporting virtually indistinguishable symptom profiles in individuals with idiopathic versus FXS-associated ASD (Bailey et al., 1998; Dissanayake et al., 2009; Rogers et al., 2001), and others finding differences in the symptom profiles of these groups (Wolff et al., 2012). However, omnibus measures of ASD symptoms may not have the sensitivity to precisely map phenotypic overlap or divergence of ASD-O and FXS-ASD. By using a more

fine-grained approach to characterize pragmatic language, a central feature of ASD, this study adds strong support for FXS as a genetic model that may lend insight into pathophysiological mechanisms that are common across etiological subgroups of ASD. Evidence for shared behavior in ASD and FXS has implications for the eventual identification of causal pathways that may be common across disorders, and which may be traced back to biological pathways that are disrupted by the *FMR1* mutation (eventually leading to the development of interventions directly targeting these disrupted pathways).

Furthermore, evidence was found for substantial phenotypic overlap in a complex social-communicative skill, pragmatic language, which contradicts the view that behavioral overlap in ASD-O and FXS-ASD may be limited to lower-level behaviors, such as motor stereotypies (Wolff et al., 2012). While some prior work has suggested that ASD-related symptoms in FXS (such as pragmatic difficulties) present as a consequence of cognitive deficits (e.g., Hall et al., 2010; Loesch et al., 2007), the results of this study strongly suggest that pragmatic language difficulties in FXS are more closely tied to ASD comorbidity than to the presence of intellectual disability. On direct assessment ratings of pragmatics in a semi-naturalistic context, the boys with FXS-ASD showed greater pragmatic deficits than did the boys with FXS-O and DS who were of a similar mental age (and these differences held even after mental age was controlled for statistically). ASD symptoms also accounted for a substantial amount of variability in performance on the PRS-SA (although this was not necessarily the case with the CASL-PJ). Thus, it appears that the presence of ASD in FXS is associated with pragmatic impairments that are elevated beyond mental-age based expectations, and are more severe than both boys with FXS-O as well as boys with other genetically based intellectual disabilities (i.e., DS). This is significant because pragmatic language is an inherently social skill that is deeply rooted in the ability to process and engage with the social world. Thus, the comparison of pragmatic language phenotypes in neurodevelopmental disabilities such as ASD and FXS may lend insight into the neural basis underlying complex social behaviors, such as pragmatic language, and how underlying genetic variation may give rise to such phenotypes.

Analyses of specific types of pragmatic language behaviors were similarly revealing. In particular, the boys with ASD-O and FXS-ASD showed overall similar profiles on both pragmatic

measures, with the groups differing only in the ability to signal humor, and in the speech-related features of volume, intelligibility, and stuttering/cluttering that may impact pragmatics. It is not unexpected that stuttering/cluttering and poor intelligibility were exhibited more frequently by the boys with FXS (with and without ASD) than the boys with ASD-O; males with FXS are often described as having cluttered speech characterized by a rapid, fluctuating rate with dysfluent components and repetitions (Hanson, Jackson, & Hagerman, 1986) whereas stuttering/cluttering are not characteristic features of idiopathic ASD. On the other hand, it is somewhat surprising that the boys with ASD-O exhibited greater difficulties in signaling appropriate humor than the boys with FXS-ASD. Individuals with ASD are known to struggle with abstract social concepts such as humor (Ozonoff & Miller, 1996) and the inability to appreciate jokes is thought to stem from impaired social understanding, which is core to ASD (Baron-Cohen, 1988; Siegal, Carrington, & Radel, 1996; Yirmiya, Erel, Shaked, & Solomonica-Levi, 1998). Social-cognitive impairments have also been documented among individuals with FXS (Grant, Apperly, & Oliver, 2007; Lewis et al., 2006; Losh, Martin, et al., 2012), and therefore similar performance in skills that rely on social understanding, such as humor signaling, might be expected in individuals with ASD-O as well as FXS-ASD. Given that shyness and anxiety are characteristic of FXS (Hagerman, 2002), perhaps the boys with FXS-ASD were less likely than those with ASD-O to become comfortable enough to joke with the examiner, resulting in less opportunities to exhibit inappropriate humor (participants who did not attempt to make any humorous remarks were not coded on this item by default).

While overall the group with FXS-O showed less pragmatic difficulties on the PRS-SA than the groups with ASD, some items diverged from this pattern: atypical intonation, scripting, and inappropriate topic shifting were detected at similar rates in the groups with FXS-O, FXS-ASD, and ASD-O, which were elevated in comparison to the groups with DS and TD. Scripting (i.e., stereotyped language) and atypical intonation are trademark features of the ASD language phenotype, so it is somewhat unexpected that these features were also observed among the boys with FXS who did not have ASD. However, ASD-like behaviors have been reported to occur in as many as 90% of boys with FXS, even when diagnostic thresholds are not met (Hagerman et al., 1986), perhaps accounting for this finding. Unlike the findings of

Sudhalter et al. (1990), increased perseveration was not detected in the boys with FXS-O compared to ASD-O. This discrepant finding may be related to differences in the definition of perseveration. The PRS-SA focuses on topic-level perseveration whereas the methods of Sudhalter et al. also captured phrase and sentence-level repetitions in their characterization of perseveration.

While the presence of ASD had a clear negative impact on pragmatic abilities overall, the pragmatic language skills of the disability groups without ASD were not wholly unaffected. Both the boys with FXS-O and DS demonstrated greater pragmatic violations than younger, language-age matched boys with TD on the PRS-SA, suggesting that pragmatic performance in these neurodevelopmental disabilities lags behind developmental expectations, even in the absence of co-occurring ASD. However, closer examination of item-level patterns seems to support pragmatics as a relative strength for individuals with DS, as the group differences between those with DS and TD were restricted to the speech-related features of rate, stuttering/cluttering, and intelligibility. In fact, the boys with DS exhibited the greatest difficulties with stutter/cluttering and intelligibility of all the groups, which is consistent with a number of studies suggesting that stuttering and cluttering are exceedingly common among individuals with DS, and may impact intelligibility (Devenny & Silverman, 1990; Kumin, 1994; Van Borsel & Vandermeulen, 2008). While some other studies have documented pragmatic language challenges in individuals with DS compared with mental-age matched children with TD, such as difficulties with topic elaboration (Roberts, Martin, et al., 2007) and repairing communicative breakdowns (Abbeduto et al., 2008), evidence is mixed and pragmatics is generally considered a relative strength of the language profile of individuals with DS (Martin, Klusek, Estigarribia, & Roberts, 2009).

Clinical Implications

A number of clinical implications can be drawn from this study. First, the evaluation of pragmatic skills from a semi-naturalistic conversational context proved effective in differentiating groups, and revealed more striking variations in pragmatic language than did the CASL-PJ, a standardized assessment (which differentiated boys with ASD-O and FXS-ASD from those with TD, but did not differentiate the performance of the other groups). Only a weak association was detected between the results of the two

pragmatic assessments, which may suggest that the PRS-SA and CASL-PJ measures were capturing different aspects of pragmatic behavior or different underlying impairments, a possibility supported by findings that performance on the PRS-SA was strongly predicted by autism severity whereas general cognitive and structural language abilities proved more predictive of performance on the CASL-PJ. Overall, the CASL-PJ was able to detect frank pragmatic language violations, whereas the PRS-SA, which directly samples conversational behavior, was better suited for uncovering more subtle violations. The PRS-SA also assesses a broad range of behaviors, likely contributing to its ability to more sensitively index of pragmatic abilities than the CASL-PJ, which taps a more limited range of pragmatic features. Our findings are consistent with prior work highlighting the strengths of naturalistic measures in the assessment of pragmatics (e.g., Adams, 2002; Hyter, 2007; McTear & Conti-Ramsden, 1992; Prutting & Kittchner, 1987; Roth & Spekman, 1984). Naturalistic assessment preserves the intricacies of social-communication that are difficult to replicate in standardized testing, such complex information processing, an unstructured format, and the need to process contextual cues (Clark & Rutter, 1981; Loukusa et al., 2007; Williams, Goldstein, & Minshew, 2006). Thus, naturalistic measures are thought to increase generalizability (Adams, 2002; Prutting & Kittchner, 1987). Naturalistic assessment techniques are also ideal for measuring treatment effects, as they are sensitive to small changes in skills and they can be administered repeatedly without risk of learning effects. Of course, limitations of naturalistic methods are that they often require extensive training and may be more labor-intensive than standardized measures or informant-based measures such as the CCC-2. While standardized assessments may provide useful information regarding the child's performance relative to peers, observation of performance on an ecologically valid setting may provide more detailed information regarding strengths and weaknesses.

Findings indicate that ASD comorbidity has a significant detrimental effect on the pragmatic language skills of individuals with FXS, underscoring the importance of considering ASD comorbidity in the evaluation and treatment of this population. The finding that boys with FXS-ASD performed significantly worse than the boys with FXS-O in the semi-naturalistic context suggests that pragmatic language deficits may be most characteristic of those individuals with comorbid ASD (in other words, it

appears that pragmatic language difficulties are core to ASD within the context of FXS, rather than to FXS itself). While the standardized pragmatic language measure failed to differentiate the performance of the boys with FXS-ASD and FXS-O, a trend was detected for poorer performance in the group with FXS-ASD. Given the small sample size of the group with FXS-O (n = 16) this study may have been underpowered to detect group differences of small effect. Indeed, a previous study from our group, which included a larger sample of boys with FXS-O, did detect greater pragmatic deficits on the CASL-PJ in boys with FXS-ASD in comparison to boys with FXS-O (Losh, Martin, et al., 2012). Other studies have also detected greater pragmatic language violations in the spontaneous discourse of boys with FXS-ASD as compared to boys with FXS-O (Martin et al., 2012; Roberts, Martin, et al., 2007). Because ASD is highly likely to co-occur in FXS and has a detrimental effect on developmental outcomes, all individuals with FXS should be evaluated for ASD. Presently, it is unknown whether interventions designed for individuals with idiopathic ASD are effective for individuals with fragile X-associated ASD. However, it is likely that the treatment of individuals with FXS-ASD would warrant a different intensity and type of services than that for FXS-O, given the divergent behavioral phenotypes of these groups. Future research investigating the efficacy of ASD-tailored treatments for individuals with FXS and co-occurring ASD will be important for determining best clinical practices for this population.

Limitations and Directions

This study has several limitations. First, a single diagnostic tool (the ADOS) was used to characterize ASD, while best practice dictates the use of multiple sources of information in determining the presence of ASD. The incorporation of other ASD diagnostic instruments, such as the Autism Diagnostic Interview-Revised (Lord, Rutter, & Le Couteur, 1994), would have allowed for more precise characterization of ASD. Second, this study included only boys and it is unclear whether the findings of this study would generalize to females who, in the case of FXS, will be less severely affected due to the presence of an unaffected X chromosome. Finally, the results of this study are based on the presentation of these disorders at a single point in time, with limited ability to account for developmental patterns or prospective features that may predict pragmatic language impairment. Longitudinal studies are needed to

understand the emergence of pragmatic language deficits in individuals with ASD and FXS across development, which might help identify developmental periods that are most optimal for intervention. Future directions of this work might include the examination of other naturalistic pragmatic language assessment methods that may better characterize this multifaceted skill in these population comparisons, such as turn-by-turn pragmatic hand coding systems.

Table 1

Group Characteristics

	Group					
	ASD-O *n* = 29	FXS-all *n* = 54	FXS-ASD *n* = 38	FXS-O *n* = 16	DS *n* = 20	TD *n* = 20
	M (*SD*) Range	*M* (*SD*) Range	*M* (*SD*) Range	*M* (*SD*) Range	*M* (*SD*) Range	*M* (*SD*) Range
Chronological age	9.22 (3.15)[a] 3.18-14.56	11.84 (3.11)[b] 5.59-17.82	12.23 (2.91)[b] 6.58-12.23	10.93 (3.49)[a,b] 5.59-16.38	12.90 (2.75)[b] 8.38-17.90	4.82 (1.00)[c] 3.54-6.69
Nonverbal mental age[1]	6.71 (2.04)[a] 2.33-10.50	5.20 (0.73)[b] 3.50-8.25	5.13 (0.60)[b] 3.50-6.67	5.34 (0.98)[b] 4.00-8.25	5.66 (1.23)[b] 4.33-9.58	5.27 (1.16)[b] 2.58-7.50
Receptive vocabulary age[2]	6.68 (2.17)[a] 1.75-10.50	6.53 (1.49)[a] 3.50-9.33	6.15 (1.51)[a] 3.50-9.33	6.93 (2.52)[a] 2.75-13.83	5.93 (2.06)[a] 2.42-10.92	6.07 (1.29)[a] 3.75-8.67
Expressive vocabulary age[3]	6.12 (1.179)[a] 2.58-9.42	6.39 (1.88)[a] 2.75-13.83	5.47 (1.41)[a] 3.85-9.92	5.91 (1.71)[a] 3.42-9.25	5.89 (1.36)[a] 3.42-8.33	5.67 (1.37)[a] 3.33-8.83
Mean length of utterance	4.70 (1.52)[a] 1.81-9.33	3.69 (1.00)[b] 1.80-7.30	3.46 (0.85)[b] 1.80-6.05	4.23 (1.16)[a,b] 2.27-7.30	2.22 (0.85)[b] 1.91-5.08	4.88 (0.59)[a] 4.12-6.06

Note. [1]Age equivalent on the Leiter International Performance Scale-Revised or the Wechsler Abbreviated Scale of Intelligence; [2]Peabody Picture Vocabulary Test age equivalent [3]Expressive Vocabulary Test age equivalent. Means in the same row with different superscripts differ significantly at $p < .05$.

Table 2

Demographic Characteristics

	% with Group					
	ASD-O	FXS-all	FXS-ASD	FXS-O	DS	TD
Race						
Caucasian	89.7	83.3	81.6	87.5	80.0	75.0
African American	10.3	1.9	--	6.3	15.0	5.0
Asian	--	5.6	7.0	--	--	--
Multi-racial	--	1.9	2.6	--	--	2.1
Not Reported	--	7.4	7.9	6.3	5.0	10.0
Income						
<20*k*	3.4	--	--	--	--	--
20*k*-39*k*	13.8	3.7	5.3	--	--	10.0
40*k*-59*k*	6.9	7.4	10.5	--	--	10.0
60*k*-79*k*	17.2	7.4	7.9	6.3	5.0	20.0
>80*k*	27.7	44.4	50.0	31.3	35.0	30.0
Not Reported	31.0	37.0	26.3	62.5	60.0	30.0
Maternal Education Level						
High School	17.2	16.7	18.4	12.5	10.0	20.0
Associate	6.9	14.8	15.8	18.8	20.0	10.0
Bachelor	37.9	40.7	31.6	62.5	20.0	35.0
Master	20.7	13.0	15.8	6.3	25.0	20.0
Doctorate	--	9.3	10.5	6.3	15.0	10.0
Not Reported	17.2	5.5	7.8	--	10.0	5.0

Table 3

Descriptive Statistics of Group Performance on the Pragmatic Language Measures

	Group					
	ASD-O	FXS-all	FXS-ASD	FXS-O	DS	TD
	M (*SD*) Range	*M* (*SD*) Range	*M* (*SD*) Range	*M* (*SD*) Range	*M* (*SD*) Range	*M* (*SD*) Range
PRS-SA Total Score	36 (6.70) 17-49	29.91 (9.00) 7-47	33.26 (7.38) 18-47	21.94 (7.44) 7-33	22.35 (6.34) 12-36	12.90 (6.99) 1-26
CASL-PJ Age Equivalent	5.00 (1.72) 2.42-8.33	4.92 (1.54) 2.42-9.17	4.58 (1.30) 2.42-6.83	5.68 (1.77) 3.75-9.17	4.71 (1.47) 2.58-8.33	5.45 (1.36) 3.42-7.75

Note. PRS-SA = Pragmatic Rating Scale-School Age; CASL-PJ = Pragmatic Judgment subtest of the Comprehensive Assessment of Spoken Language.

Table 4

Model Adjusted Means and Group Comparisons on Individual Pragmatic Rating Scale-School Age Items

	Group				
	ASD-O	FXS-ASD	FXS-O	DS	TD
Item	*M* (*SE*)	*M* (*SE*)	*M* (*SE*)	*M* (*SE*)	*M* (*SE*)
Limited topic initiation	**1.12 (1.98)^{a,b}**	**1.19 (1.69)^{a}**	**0.58 (0.24)^{b,c}**	**0.74 (0.23)^{a,b,c}**	**0.18 (0.22)^{c}**
Inappropriate topic shifts	**1.42 (0.20)^{a}**	**1.40 (0.17)^{a}**	**1.45 (0.24)^{a}**	**0.76 (0.23)^{b}**	**0.47 (0.22)^{b}**
Interrupting	1.37 (0.19)^{a}	0.96 (0.16)^{a}	0.90 (0.23)^{a}	0.77 (0.22)^{a}	0.88 (0.21)^{a}
Failure to acknowledge	**1.67 (0.16)^{a}**	**0.86 (0.14)^{a}**	**0.34 (0.20)^{b}**	**0.22 (0.19)^{b}**	**0.37 (0.18)^{b}**
Reduced topic elaboration	**1.24 (0.13)^{a}**	**1.40 (0.11)^{a}**	**0.79 (0.61)^{b}**	**0.51 (0.15)^{c}**	**0.41 (0.15)^{b,c}**
Perseveration	**1.36 (0.21)^{a}**	**1.24 (0.18)^{a}**	**0.75 (0.25)^{a,b}**	**0.81 (0.24)^{a,b}**	**0.41 (0.15)^{b}**
Vocal noises	1.08 (0.19)^{a}	0.96 (0.16)^{a}	0.70 (0.23)^{a}	0.49 (0.22)^{a}	0.35 (0.21)^{a}
Poor reciprocal conversation	**1.42 (0.14)^{a}**	**1.40 (0.12)^{a}**	**0.79 (0.17)^{b}**	**0.50 (0.16)^{b}**	**0.40 (0.15)^{b}**
Formality	0.33 (0.12)^{a}	0.26 (0.11)^{a}	0.18 (0.15)^{a}	0.13 (.14)^{a}	0.30 (0.14)^{a}
Scripting	**1.35 (0.17)^{a}**	**1.35 (0.14)^{a}**	**1.20 (0.21)^{a}**	**0.26 (0.20)^{b}**	**0.11 (0.19)^{b}**
Grammar/vocabulary errors	1.89 (0.10)^{a}	1.87 (0.08)^{a}	1.88 (0.12)^{a}	2.05 (0.11)^{a}	1.75 (0.10)^{a}
Poor intelligibility	**0.71 (0.12)^{a}**	**1.37 (0.10)^{b}**	**0.77 (0.14)^{a}**	**1.58 (0.17)^{b}**	**0.23 (0.13)^{c}**
Atypical rate	**1.58 (0.17)^{a}**	**1.68 (0.15)^{a}**	**1.22 (0.21)^{a}**	**1.70 (0.20)^{a}**	**0.35 (0.19)^{b}**
Atypical intonation	**1.71 (0.13)^{a}**	**1.58 (0.11)^{a}**	**1.17 (0.16)^{a}**	**0.19 (0.16)^{b}**	**0.12 (0.15)^{b}**
Inappropriate volume	**1.65 (0.17)^{a}**	**1.15 (0.14)^{b}**	**0.74 (0.20)^{b,c}**	**0.60 (0.19)^{c}**	**0.80 (0.18)^{b,c}**
Character speech	0.69 (0.18)^{a}	0.77 (0.16)^{a}	0.44 (0.22)^{a}	0.13 (0.21)^{a}	0.23 (0.20)^{a}
Dysfluent language formulation	1.05 (0.19)^{a}	0.62 (0.16)^{a}	0.40 (.023)^{a}	0.76 (0.22)^{a}	0.65 (0.21)^{a}
Stuttering/cluttering	**0.13 (0.16)^{a}**	**0.97 (0.14)^{b}**	**0.80 (0.20)^{b}**	**1.64 (0.19)^{c}**	**0.23 (0.18)^{a}**
Mismanagement of interpersonal space	1.50 (0.18)^{a}	1.58 (0.16)^{a}	1.34 (0.22)^{a}	1.08 (0.21)^{a}	1.12 (0.20)^{a}
Odd/limited gesture use	**1.53 (0.15)^{a}**	**1.45 (0.13)^{a}**	**0.87 (0.18)^{b}**	**0.44 (0.18)^{b}**	**0.45 (0.17)^{b}**
Hand/finger mannerisms	**1.23 (0.18)^{a}**	**0.81 (0.15)^{a,b}**	**0.76 (0.22)^{a,c}**	**0.30 (0.21)^{c}**	**0.40 (0.20)^{b,c}**
Atypical facial expressions	**1.61 (0.19)^{a}**	**1.22 (0.16)^{a}**	**0.54 (0.23)^{b}**	**0.52 (0.22)^{b}**	**0.15 (0.21)^{b}**
Atypical gaze	**1.54 (0.13)^{a}**	**1.73 (0.11)^{a}**	**0.85 (0.16)^{b}**	**0.39 (0.15)^{c}**	**0.64 (0.15)^{b,c}**
Inappropriate vegetative sounds	0.66 (0.20)^{a}	0.69 (0.17)^{a}	0.41 (0.24)^{a}	0.79 (0.23)^{a}	0.06 (0.22)^{a}
Odd signaling of humor	**1.18 (0.16)^{a}**	**0.70 (0.13)^{b}**	**0.34 (0.19)^{b,c}**	**0.08 (0.18)^{c}**	**0.12 (0.17)^{c}**
Inability to clarify	0.62 (0.14)^{a}	0.45 (0.12)^{a}	0.36 (0.17)^{a}	0.36 (0.16)^{a}	0.47 (0.15)^{a}

Note. Covariate-adjusted means, controlling for nonverbal mental age, receptive and expressive vocabulary, and

mean length of utterance. Higher scores indicate greater impairment. Groups in the same row sharing the same letter did not differ significantly at $p < .05$.
*$p < .05$, **$p < .01$, ***$p < .001$

Table 5

Regression Coefficients Depicting Predictors of Performance on the Pragmatic Rating Scale-School Age

			B (SE)	*β*	R^2	$R^2\Delta$	$F\Delta$
	Step 1	Constant	47.76 (4.51)		.04		0.26
		Leiter-R	0.80 (1.08)	0.25			
		PPVT	-0.24 (1.32)	-0.08			
		EVT	-0.63 (1.55)	-0.41			
		MLU	-0.35 (1.00)	-0.08			
ASD-O	Step 2	Constant	22.94 (6.74)		.31	.35	10.72**
		Leiter-R	0.91 (0.91)	0.28			
		PPVT	-1.12 (1.15)	-0.36			
		EVT	0.30 (1.33)	0.08			
		MLU	-0.84 (0.86)	-0.19			
		ADOS Severity	2.14 (0.65)	0.59**			
	Step 1	Constant	37.01 (8.99)		.34		6.18***
		Leiter-R	0.55 (2.17)	0.05			
		PPVT	-4.48 (1.08)	-0.94***			
		EVT	4.67 (1.29)	0.78**			
		MLU	-2.00 (1.28)	-0.22			
FXS-all	Step 2	Constant	12.20 (7.38)		.67	.33	46.83***
		Leiter-R	2.52 (1.58)	0.20			
		PPVT	-4.33 (0.77)	0.15***			
		EVT	3.62 (0.93)	-0.44***			
		MLU	0.09 (0.96)	0.01			
		ADOS Severity	2.18 (0.32)	0.44***			

Note. Leiter-R = Leiter International Performance Scale- Revised; PPVT = Peabody Picture Vocabulary Test; EVT = Expressive Vocabulary Test; MLU = mean length of utterance; ADOS = Autism Diagnostic Observation Schedule.
*$p < .05$, **$p < .01$, ***$p < .001$.

Table 6

Regression Coefficients Depicting Predictors of Performance on the Pragmatic Judgment Subtest of the Comprehensive Assessment of Spoken Language

			B (SE)	*β*	R^2	$R^2\Delta$	$F\Delta$
	Step 1	Constant	0.11 (0.87)		.70		13.27***
		Leiter-R	-0.09 (0.16)	0.07			
		PPVT	0.50 (0.19)	0.63*			
		EVT	0.23 (0.23)	0.24			
		MLU	0.16 (0.15)	0.15			
ASD-O	Step 2	Constant	0.78 (1.18)		.71	.01	0.70
		Leiter-R	-0.09 (0.16)	-0.11			
		PPVT	0.54 (0.20)	0.68*			
		EVT	0.18 (0.23)	0.19			
		MLU	0.19 (0.15)	0.17			
		ADOS Severity	-0.10 (0.11)	-0.10			
	Step 1	Constant	0.27 (0.91)		.78		40.76***
		Leiter-R	-0.16 (0.22)	-0.08			
		PPVT	0.49 (0.11)	0.60***			
		EVT	0.33 (0.13)	0.33*			
		MLU	0.14 (0.13)	0.09			
FXS-all	Step 2	Constant	2.00 (0.91)		.82	.06	15.06***
		Leiter-R	-0.13 (0.19)	-0.14			
		PPVT	0.48 (0.10)	0.59***			
		EVT	0.41 (0.12)	0.40**			
		MLU	-0.01 (0.12)	-0.02			
		ADOS Severity	-0.15 (0.04)	-0.25***			

Note. Leiter-R= Leiter International Performance Scale- Revised; PPVT = Peabody Picture Vocabulary Test; EVT = Expressive Vocabulary Test; MLU = mean length of utterance; ADOS = Autism Diagnostic Observation Schedule.
*$p < .05$, **$p < .01$, ***$p < .001$.

Table 7

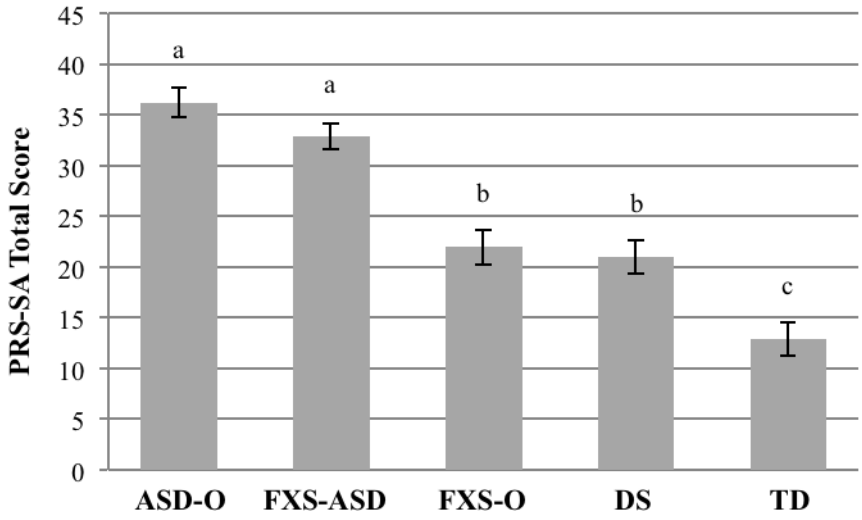
Correlations between the Pragmatic Rating Scale-School Age and the Pragmatic Judgment Subtest of the Comprehensive Assessment of Spoken Language

	PRS-SA					
	Full Sample	ASD-O	FXS-ASD	FXS-O	DS	TD
CASL-PJ	-.31*	-.25	-.32	-.50*	-.36	-.20
n	118	29	35	16	19	20

Note. PRS-SA = Pragmatic Rating Scale-School Age; CASL-PJ = Pragmatic Judgment subtest of the Comprehensive Assessment of Spoken Language.
*$p < .05$.

Figure 1

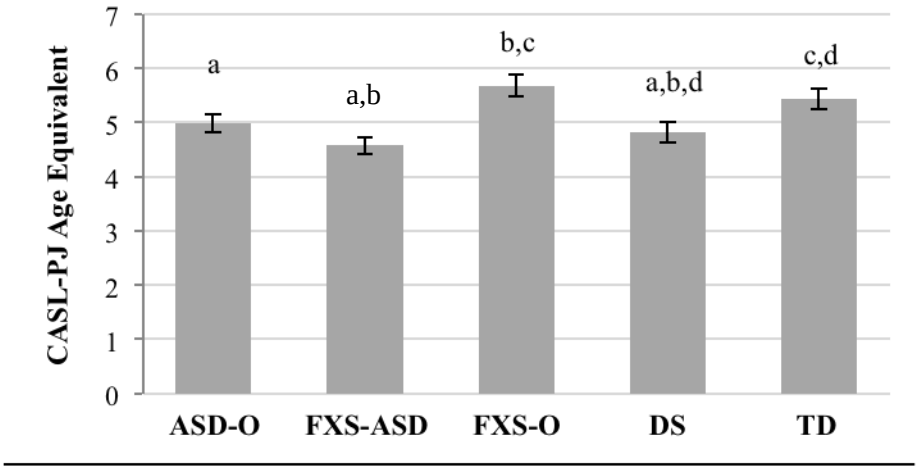
Group Comparisons on the Pragmatic Rating Scale-School Age



Note. Covariate-adjusted means, controlling for nonverbal mental age, receptive and expressive vocabulary, and mean length of utterance. Higher scores indicate greater impairment.
Groups sharing the same letter did not differ significantly at $p < .05$.

Figure 2

Group Comparisons on the Pragmatic Judgment subtest of the Comprehensive Assessment of Spoken Language



Note. Covariate-adjusted means, controlling for nonverbal mental age, receptive and expressive vocabulary, and mean length of utterance. Lower scores indicate greater impairment.
Groups sharing the same letter did not differ significantly at $p < .05$.

Acknowledgements

This paper was completed as part of the first author's doctoral dissertation, which was supported in part by the James J. Gallagher Dissertation Award of Frank Porter Graham Child Development Institute. This work was funded by the National Institute of Child Health and Human Development (R01HD0388190-62A, R01HD038819-09S1, R01HD044935-06A), the National Institute on Deafness and Other Communication Disorders (1R01DC010191-01A1, R03DC010880), the National Institute of Maternal Health (R01MH091131-01A1), the National Fragile X Foundation, the Ireland Family Foundation, and the Research Participant Registry Core of the Carolina Institute for Developmental Disabilities (P30HD03110). The authors would like to thank Linda Watson, Heather Cody Hazlett, and Jane Roberts for their comments on an earlier version of this manuscript, Abigail Hogan-Brown for her assistance with the pragmatic language coding, Christine Rothermel for her help with the tables, and John Sideris for his guidance on the statistical analyses. We also thank Rebecca Landa for permitting our use of the Pragmatic Rating Scale-School Age in this project, and for providing training and guidance on its implementation. We gratefully acknowledge the late Dr. Joanne Roberts, who was awarded the original NICHD grants that supported the initial phases of this research, and the children and families who participated.

NOTE: This article will be published in its final form by the Journal of Speech, Language, and Hearing Research; http://jslhr.pubs.asha.org.

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