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Research Article

A Multimethod Analysis of Pragmatic Skills in Children and Adolescents With Fragile X Syndrome, Autism Spectrum Disorder, and Down Syndrome

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Purpose: Pragmatic language skills are often impaired above and beyond general language delays in individuals with neurodevelopmental disabilities. This study used a multimethod approach to language sample analysis to characterize syndrome- and sex-specific profiles across different neurodevelopmental disabilities and to examine the congruency of 2 analysis techniques.
Method: Pragmatic skills of young males and females with fragile X syndrome with autism spectrum disorder (FXS-ASD, n = 61) and without autism spectrum disorder (FXS-O, n = 40), Down syndrome (DS, n = 42), and typical development (TD, n = 37) and males with idiopathic autism spectrum disorder only (ASD-O, n = 29) were compared using variables obtained from a detailed hand-coding system contrasted with similar variables obtained automatically from the language analysis program Systematic Analysis of Language Transcripts (SALT).
Results: Noncontingent language and perseveration were characteristic of the pragmatic profiles of boys and girls with FXS-ASD and boys with ASD-O. Boys with ASD-O also initiated turns less often and were more nonresponsive than other groups, and girls with FXS-ASD were more nonresponsive than their male counterparts. Hand-coding and SALT methods were largely convergent with some exceptions.
Conclusion: Results suggest both similarities and differences in the pragmatic profiles observed across different neurodevelopmental disabilities, including idiopathic and FXS-associated cases of ASD, as well as an important sex difference in FXS-ASD. These findings and congruency between the 2 language sample analysis techniques together have important implications for assessment and intervention efforts.

Pragmatic aspects of communication encompass a broad range of linguistic, nonlinguistic, and paralinguistic skills, including topic maintenance, turn taking, and speech acts, as well as nonverbal communication (e.g., gestures) and paralinguistic variations in rate, rhythm, and intonation (P. Brown & Levinson, 1987; Grice, 1975; Sperber & Wilson, 2002). Deficits in any component of pragmatics can significantly impact problem behavior and emotional regulation (Helland, Lundervold, Heimann, & Posserud, 2014; Ketelaars, Cuperus, Jansonius, & Verhoeven, 2010), academic functioning and language learning across domains (Bashir & Scavuzzo, 1992), and interpersonal relationships (Gallagher, 1993). Difficulties in pragmatic language have been reported for individuals with fragile X syndrome (FXS), autism spectrum disorder (ASD), and Down syndrome (DS), with considerable evidence of pragmatic impairment in FXS and ASD and more mixed findings for individuals with DS (Abbeduto & Hesketh, 1997; Hatton, 1998; G. E. Martin, Lee, & Losh, 2017; McDuffie, Thurman, Channell, & Abbeduto, 2016; Rice, Warren, & Betz, 2005). FXS, ASD, and DS are among the most common neurodevelopmental disabilities associated with language impairment, and thus, they are likely groups to be on the caseload of a speech-language pathologist. However, very few cross-population studies have been conducted comparing these groups directly to define syndrome-specific pragmatic profiles, and even fewer studies have examined pragmatic skills

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in girls, despite known sex differences in typical development (TD) for pragmatic language (Berghout, Salehi, & Leffler, 1987; Cook, Fritz, McCornack, & Visperas, 1985; Leaper, 1991). Clarifying pragmatic profiles and determining whether they differ across clinical populations or by sex are critical for informing targeted assessment and intervention efforts to promote communicative competence (Fidler, Philofsky, & Hepburn, 2007; Messinger et al., 2015; Rinehart, Cornish, & Tonge, 2011; Thompson, Caruso, & Ellerbeck, 2003).

In addition to a lack of knowledge on clinically meaningful syndrome- and sex-specific differences in pragmatic profiles, clinicians working with individuals with neurodevelopmental disabilities also face the challenge of valid assessment of these skills. Although key advantages of standardized tests of pragmatic language include relatively quick administration time and efficient scoring with standard references, information gained from these highly structured assessments may have limited generalizability to everyday contexts (e.g., conversational interaction; Adams, 2002; Prutting & Kittchner, 1987). Thus, more naturalistic assessment of pragmatic language has been recommended (Adams, 2002; Hyter, 2007; McTear & Conti-Ramsden, 1992; Prutting & Kittchner, 1987; Roth & Spekman, 1984) and may be more sensitive than standardized pragmatic assessments for children with neurodevelopmental disabilities (Klusek, Martin, & Losh, 2014a). Pragmatic coding systems used in the literature, however, are time intensive and typically not easy to apply in clinical practice.

To address these needs, the current study utilized a multimethod approach to language sample analysis, including a detailed hand-coding (manual, turn-by-turn coding) system assessing key pragmatic skills that has been used previously in research (Roberts, Martin, et al., 2007), along with automated analyses obtained from a language-sampling software that requires minimal data processing beyond transcription and that is therefore more accessible to clinicians: the Systematic Analysis of Language Transcripts (SALT; Miller & Iglesias, 2008). Both systems rely on language transcripts, but the use of these transcripts to obtain pragmatic variables differs. In the hand-coding system, coders are trained to systematically evaluate elements of pragmatic language; every conversational turn in the transcript is marked according to a specific set of guidelines. In contrast, SALT uses the transcripts directly to automatically compute a series of outcome variables and, thus, is more common in clinical practice. However, relative to hand coding, SALT may not detect some of the more nuanced, but clinically significant, pragmatic strengths and weaknesses common to individuals with neurodevelopmental disabilities. It is therefore important to evaluate SALT against hand-coding methods, to understand its potential utility in application with clinical syndromes impacting pragmatics.

Employing these two techniques, we compared the pragmatic skills of children and adolescents with FXS with and without ASD (FXS-ASD, FXS-Only or FXS-O), idiopathic ASD (ASD-O), and DS, as well as a control group of younger children with TD, during seminaturalistic (semistructured) social-communicative interactions. Girls were included in the FXS, DS, and TD groups (data on girls were not available for the ASD-O group). The literature describing pragmatic skills during naturalistic or seminaturalistic interactions, including sex differences when available, in each of the groups is summarized below. Based on this literature, the current study focused on the following key pragmatic features impacted in these groups: noncontingent language (loosely related or tangential, as well as off-topic or irrelevant language), perseveration (excessive verbal self-repetition of a word, phrase, sentence, or topic), initiations (nonobligatory, self-initiated contributions), and nonresponsiveness (failure to respond when a response is obligatory). With the exception of noncontingent language, each of these key pragmatic features had the potential to be captured by both hand coding and SALT schema in this study.

FXS

FXS, the most common inherited cause of intellectual disability, occurs in roughly one in 2,500 to one in 5,000 individuals (Coffee et al., 2009; P. J. Hagerman, 2008; Pesso et al., 2000). FXS results from > 200 trinucleotide cytosine–guanine–guanine repeats on the *FMR1* gene. This disruption completely or partially shuts down the production of the fragile X mental retardation protein (FRMP), which is known to play a critical role in synaptic maturation in the developing brain (Weiler et al., 1997). In females, the second unaffected X chromosome provides an additional healthy FMRP-producing copy of *FMR1*. As a result, females with FXS are typically not as severely impaired as males (R. J. Hagerman & Hagerman, 2002; Loesch et al., 2002), and thus, most research to date has focused on males.

Based on detailed turn-by-turn hand-coding approaches to language sample analysis, characteristic pragmatic features reported in males with FXS include noncontingent language and perseveration (Levy, Gottesman, Borochowitz, Frydman, & Sagi, 2006; Roberts, Martin, et al., 2007; Sudhalter & Belser, 2001; Sudhalter, Cohen, Silverman, & Wolf-Schein, 1990). More recently, Klusek et al. (2014a) examined social-communicative interactions using a comprehensive, 34-item observational rating system of pragmatic language skills based on ratings of videotaped language samples (another type of hand coding) and found that boys with FXS-O (without ASD) showed more pragmatic deficits overall relative to younger controls with TD and more inappropriate topic shifts (but not more perseveration) according to item-level analysis. Far fewer studies have focused on the pragmatic skills of females with FXS. In two studies, females with FXS waited longer to take their first turn (Lesniak-Karpiak, Mazzocco, & Ross, 2003) and made fewer requests for information on a topic (Mazzocco et al., 2006) than controls, suggesting that reduced initiations may be characteristic of the pragmatic profile of females. Research into sex differences in pragmatic language in FXS is also

lacking, although one study found greater repetition of rote phrases in males compared with females, yet no difference for the repetition of topics (Murphy & Abbeduto, 2007). No studies to date have examined sex differences in noncontingent language use in FXS.

FXS is also the most common known genetic cause of ASD, with approximately 40%–74% of males and 13%–45% of females with FXS meeting ASD criteria (D. B. Bailey, Raspa, Olmsted, & Holiday, 2008; Clifford et al., 2007; Hall, Lightbody, & Reiss, 2008; Kaufmann et al., 2004; Philofsky, Hepburn, Hayes, Hagerman, & Rogers, 2004; Rogers, Wehner, & Hagerman, 2001). Two studies have reported greater noncontingent language (Roberts, Martin, et al., 2007) and perseveration (G. E. Martin et al., 2012) in boys with FXS-ASD than those with FXS-O using a turn-by-turn hand-coding approach. In the study by Klusek et al. (2014a) described previously, boys with FXS-ASD and FXS-O did not differ significantly on items capturing the severity of inappropriate topic shifts or perseveration, suggesting that summary ratings of the severity of these features may not be as sensitive as turn-by-turn hand-coding characterizations. No studies have compared the pragmatic skills of girls with FXS-ASD and FXS-O during naturalistic or seminaturalistic interactions, leaving unknown whether pragmatic skills are impacted by ASD status similarly in boys and girls with FXS. Comparisons between FXS and ASD and between FXS and DS are provided in the following sections.

ASD

ASD is a neurodevelopmental disability defined behaviorally according to deficits in social interaction, communication, and restricted or repetitive behaviors and interests (American Psychiatric Association, 2013) and is estimated to occur in one in 59 children (Baio et al., 2018). Although pragmatic language is impaired in ASD by definition, particular deficits have been reported in studies using a hand-coding approach to language sample analysis. Like boys with FXS, children with ASD-O tend to produce noncontingent language (Capps, Kehres, & Sigman, 1998; Losh & Capps, 2003; Tager-Flusberg & Anderson, 1991) and perseveration (Ross, 2002). In addition, nonresponsiveness to communicative bids and infrequent initiations have been observed (Capps et al., 1998; Jackson et al., 2003; Loveland, Landry, Hughes, Hall, & McEvoy, 1988).

Given the high comorbidity of FXS and ASD, the comparison of these two groups is significant—beyond the clinical importance of guiding potentially tailored assessment and intervention efforts—for the identification of endophenotypes, or genetically linked traits, that may be shared across FXS and ASD and associated with the *FMR1* gene in particular. Sudhalter and colleagues have reported more tangential language (Sudhalter & Belser, 2001) and perseveration (Sudhalter et al., 1990) in FXS than ASD-O using turn-by-turn hand-coding methodology. However, they either excluded males with FXS and comorbid autism (Sudhalter et al., 1990) or did not report the autism status of their participants with FXS (Sudhalter & Belser, 2001). In the investigation by Klusek et al. (2014a) described in the previous section, boys with FXS-ASD and ASD-O showed a similar level of pragmatic impairment overall and did not differ significantly from each other in inappropriate topic shifts or perseveration. Thus, no study to date has compared pragmatic skills in FXS-ASD and ASD-O using a turn-by-turn hand-coding approach, which may be more sensitive to group differences than item-level analysis.

DS

DS occurs in approximately one in 700–800 live births and is the most common known genetic cause of intellectual disability (Parker et al., 2010; Pueschel, 1995). Social skills are a relative strength in this population (Dykens, Hodapp, & Evans, 2006; Freeman & Kasari, 2002; Moore, Oates, Hobson, & Goodwin, 2002; Wishart & Johnston, 1990). Indeed, based on turn-by-turn hand coding of social-communicative interactions, boys with DS are more contingent and less perseverative than boys with FXS or ASD (G. E. Martin et al., 2012; Roberts, Martin, et al., 2007; Tager-Flusberg & Anderson, 1991), although they tend to introduce new topics and elaborate on topics to a lesser extent than controls with TD and more often maintain topics by adding minimal information (Roberts, Martin, et al., 2007; Tannock, 1988). Klusek et al. (2014a) reported that boys with DS committed more pragmatic violations than younger mental age–matched male controls with TD overall but did not differ from them on topic shifts or perseveration. Using the same rating system as Klusek et al. (2014a); Lee et al. (2017) recently reported that boys with DS did not differ from male controls with TD in pragmatic language overall. However, girls with DS committed more pragmatic violations than female controls, and inappropriate topic shifts were more frequent in girls with DS than in boys. Thus, findings for males with DS are somewhat mixed, and no studies to date have applied turn-by-turn hand-coding methods to examining pragmatic skills in a group of females with DS.

Present Study

In summary, the existing literature, based mostly on males and hand-coding methods, suggests both similarities and differences across pragmatic language profiles in FXS, ASD, and DS. This study aimed to further delineate the pragmatic profiles of these groups by using a multimethod approach to language sample analysis and by examining sex differences. Groups included boys and girls with FXS-ASD, FXS-O, DS, and TD and boys with ASD-O (data on girls with ASD-O were not available). We utilized two language sample analysis techniques: a detailed turn-by-turn hand-coding system based on secondary processing of language transcripts (Roberts, Martin, et al., 2007) and a complementary and relatively less time-intensive language analysis software based on the language transcripts only

(SALT). Specifically, the present investigation addressed the following research questions:

1. Do boys with FXS-ASD, FXS-O, ASD-O, DS, and TD differ in pragmatic skills, and do boys from the clinical groups differ in the types of pragmatics skills that are impaired?
2. Do girls with FXS-ASD, FXS-O, DS, and TD differ in pragmatic skills, and do girls from the clinical groups differ in the types of pragmatic skills that are impaired?
3. Are there sex differences in pragmatic skills of children with FXS-ASD, FXS-O, DS, and TD?
4. What is the congruency between the two language sample analysis techniques? Specifically, (a) do patterns of differences vary according to method applied, and (b) do complementary variables across the two techniques align (correlate) with each other (both for all groups collapsed and all groups separately)?

We hypothesized that FXS-ASD and ASD-O groups would demonstrate the greatest pragmatic difficulties and that boys with FXS-ASD and ASD-O would show overlapping profiles in the types of pragmatic skills impacted. We further hypothesized that boys with FXS would exhibit greater deficits than girls but that we would find an opposite sex difference in the DS group, based on the existing literature. In comparing gold standard hand-coding methods to SALT, we expected that hand coding may be more sensitive for detecting group differences.

Method

Participants

Participants comprised three clinical groups and a control group with TD (see Table 1 for participant characteristics), including 46 boys and 15 girls with FXS-ASD, 13 boys and 27 girls with FXS-O, 29 boys with ASD-O, 20 boys and 22 girls with DS, and 19 boys with TD and 18 girls with TD. Participants were taking part in a large-scale longitudinal study of pragmatic language ability (Losh, Martin, Klusek, Hogan-Brown, & Sideris, 2012; G. E. Martin, Barstein, et al., 2017) and were recruited from parent support groups, childcare centers, schools, research registries, and genetic clinics in the eastern, southeastern, and midwestern United States. Of note, given the lower incidence of ASD in females (both overall and in FXS) compared with males (A. Bailey et al., 1993; D. B. Bailey et al., 2008; Clifford et al., 2007; I. L. Cohen, Brown, et al., 1989; Klusek, Martin, & Losh, 2014b; Mazzocco, Kates, Baumgardner, Freund, & Reiss, 1997), girls with idiopathic ASD were not included in this study. Participants were recruited with the goal of matching groups on nonverbal mental age; however, this was not achieved given the recruitment challenges inherent to research with rare conditions. Therefore, analyses controlled for nonverbal mental age, receptive and expressive vocabulary age equivalents, and mean length of utterance (MLU) in morphemes (measures described next). These covariates were selected because pragmatic deficits may be linked to either cognitive or structural language deficits (see G. E. Martin, Lee, et al., 2017, for discussion) and because of the significant differences across groups on the covariates (see Table 1). For further characterization of groups, please also see Table 2 for nonverbal cognition, receptive vocabulary, and expressive vocabulary standard scores.

Inclusion criteria included speaking English as the primary language at home and using three or more words in an utterance. The FXS group had the *FMR1* full mutation (cytosine–guanine–guanine expansion of > 200 on *FMR1*). Participants were excluded if they failed a hearing screening with a threshold greater than 30 dB HL in the better ear across 500, 1000, 2000, and 4000 Hz. Participants in the TD and DS groups were also excluded if they met criteria for ASD on the Autism Diagnostic Observation Schedule (ADOS; Lord, Rutter, DeLavore, & Risi, 2001). Children with TD had no history of developmental or language delays.

Procedure

Assessments were administered in the child's home or school or at a testing space in a research laboratory, based on family preference and availability. Sessions were audiotaped using a digital audio recorder (Marantz PMD670) and videotaped using a SONY Digital 8 camcorder (Model DCR-TVR27). Procedures were approved by the University of North Carolina at Chapel Hill and Northwestern University Institutional Review Boards. Informed consent was provided by the parent or guardian.

ASD Classification

ASD status was determined using the ADOS (Lord et al., 2001), a semistructured, play-based assessment. The ADOS yields scores of autism, autism spectrum, and nonspectrum. Participants who met criteria for either autism or autism spectrum were included in the ASD groups. Because data came from a larger longitudinal study in which multiple ADOSs were usually available for a given child (i.e., 29% had one ADOS, 54% had two, 15% had three, and 2% had four), calibrated autism severity scores based on the ADOS (Gotham, Pickles, & Lord, 2009) were averaged across multiple ADOSs when available to determine ASD status (see Table 1 for average severity scores). The utilization of longitudinal ADOS assessments is consistent with our prior work (i.e., Barstein, Martin, Lee, & Losh, 2018; Klusek et al., 2014a; G. E. Martin, Barstein, et al., 2017) and deemed to be the best estimate of, and most valid approach to, determining ASD status using these longitudinal data. Staff members administering and scoring the ADOS achieved reliability either through direct training with the test developers or through intralab reliability per standards outlined by the test developers. Of note, one participant with ASD-O did not meet ASD criteria using the ADOS. However, because he had a prior clinical diagnosis

Table 1. Participant characteristics.

Variable	FXS-ASD *M* (*SD*) Range	FXS-O *M* (*SD*) Range	ASD-O *M* (*SD*) Range	DS *M* (*SD*) Range	TD *M* (*SD*) Range
Males	*n* = 46	*n* = 13	*n* = 29	*n* = 20	*n* = 19
Chronological age	10.4 (2.4)[a]	9.7 (3.3)[ab]	9.0 (2.4)[b]	**10.9 (2.1)[a]**	4.7 (1.0)[c]
	6.6–15.1	6.1–15.0	4.2–12.8	**6.8–14.9**	3.2–6.7
Nonverbal mental age[a]	5.0 (0.49)[a]	**5.5 (0.99)[ab]**	6.1 (1.6)[b]	5.3 (0.85)[a]	5.2 (1.2)[a]
	3.5–6.0	**4.4–8.3**	3.9–10.5	4.3–8.3	3.6–7.5
Receptive vocabulary age[b]	5.6 (1.4)[ab]	6.5 (2.6)[a]	6.0 (2.0)[ab]	5.2 (1.4)[b]	5.9 (1.6)[ab]
	2.4–8.8	3.4–13.8	3.1–10.5	2.4–7.5	2.2–8.7
Expressive vocabulary age[c]	5.0 (1.1)[a]	**5.4 (1.5)[ab]**	5.8 (1.6)[b]	5.5 (1.3)[ab]	5.5 (1.6)[ab]
	2.7–7.3	**3.8–9.3**	3.4–8.9	3.6–8.6	2.9–8.3
MLU[d]	3.5 (0.69)[ab]	**4.0 (0.73)[ac]**	4.2 (1.2)[c]	3.2 (0.77)[b]	4.9 (0.65)[d]
	2.3–4.9	**2.3–4.7**	1.9–6.4	1.8–4.8	3.7–6.1
Autism severity[e]	6.6 (1.6)[a]	2.4 (1.0)[b]	7.5 (2.0)[c]	1.5 (0.56)[b]	1.5 (0.61)[b]
	4–10	1–3.5	2–10	1–3	1–3
Females	*n* = 15	*n* = 27		*n* = 22	*n* = 18
Chronological age	9.3 (3.8)[a]	9.5 (3.7)[a]		**9.2 (2.2)[a]**	5.4 (2.5)[b]
	4.9–15.9	4.2–14.9		**6.0–14.2**	3.2–11.8
Nonverbal mental age[a]	5.4 (0.89)[a]	**7.3 (2.7)[b]**		5.0 (0.74)[a]	6.2 (2.8)[ab]
	4.0–7.3	**3.9–14.9**		3.8–6.8	3.9–14.9
Receptive vocabulary age[b]	6.8 (3.2)[ab]	8.6 (3.6)[b]		4.8 (1.9)[c]	6.3 (3.1)[ac]
	2.4–15.5	3.2–16.3		2.1–9.8	2.7–16.1
Expressive vocabulary age[c]	5.9 (2.2)[a]	**8.7 (4.0)[b]**		4.7 (1.4)[a]	5.9 (2.3)[a]
	2.9–11.3	**4.1–19.8**		1.9–7.8	3.2–12.2
MLU[d]	4.1 (1.1)[a]	**4.9 (1.1)[b]**		3.3 (0.96)[a]	5.1 (1.5)[b]
	2.3–5.6	**3.2–7.1**		2.3–6.6	3.1–7.9
Autism severity[e]	6.3 (1.7)[a]	2.1 (0.81)[b]		1.7 (0.59)[bc]	1.4 (0.58)[c]
	4–9.5	1–3.5		1–3	1–3

Note. Different superscripts within a row indicate significant differences at $p < .05$. If groups share the same letter, differences were not significant. Bold texts indicate significant sex differences at $p < .05$. All ages given in years. FXS-ASD = fragile X syndrome with autism spectrum disorder; FXS-O = fragile X syndrome only; ASD-O = autism spectrum disorder only; DS = Down syndrome; TD = typical development.

[a]Leiter International Performance Scale–Revised. [b]Peabody Picture Vocabulary Test–III. [c]Expressive Vocabulary Test. [d]Mean length of utterance in morphemes. [e]Autism Diagnostic Observation Schedule.

of ASD (as did all those with ASD-O) and scored in the autism range on the Autism Diagnostic Interview–Revised (ADI-R; Lord, Rutter, & Le Couteur, 1994), he was included in the ASD-O group. Please note that we attempted to collect ADI-R information on all participants with FXS and ASD. However, due to missing data, only the ADOS was used to determine group membership.

Cognitive and Structural Language Abilities

Nonverbal mental age was determined using the Leiter International Performance Scale–Revised (Roid & Miller, 1997). Structural language abilities were assessed using the Peabody Picture Vocabulary Test–III (Dunn & Dunn, 1997), a measure of receptive vocabulary; the Expressive Vocabulary Test (Williams, 1997), a measure of expressive vocabulary; and MLU (R. Brown, 1973), a measure of expressive morphosyntactic complexity. To compute MLU, ADOS language samples were transcribed using SALT (Miller & Iglesias, 2008) software, as described below.

Language Sampling and Transcription

Language samples from the ADOS were transcribed using audio and video by trained research assistants who had first achieved morpheme-to-morpheme agreement of 80% or higher as compared with a gold standard transcript for two samples from each diagnostic group. In addition, 10% of language samples per group were randomly selected and transcribed for reliability. Reliability for utterance segmentation was 88.36%, and the average intraclass correlation coefficient (ICC; Shrout & Fleiss, 1979) for MLU was .97 across groups. All participants received either Module 2 or Module 3 of the ADOS. See Table 3 for a breakdown of modules by group; chi-square tests by diagnosis and sex indicated no significant differences for module breakdown across groups (all *p*s > .438). Consistent with prior work (Klusek et al., 2014a), the first 55 intelligible participant turns from make-believe play and the first 55 intelligible participant turns from selected nonplay tasks were transcribed. The nonplay context included natural conversation after the construction task and during any cleanup time, the demonstration task, picture description, creating a story, birthday party, and snack. Transcripts included equal numbers of play and nonplay turns in an effort to account for potential differences in intelligibility and talkativeness across groups. That is, a goal of this procedure was to avoid having controls with TD achieve the required number of intelligible turns sooner in the

Table 2. Standard scores for nonverbal IQ and receptive/expressive vocabulary.

Variable	FXS-ASD *M* (*SD*) Range	FXS-O *M* (*SD*) Range	ASD-O *M* (*SD*) Range	DS *M* (*SD*) Range	TD *M* (*SD*) Range
Males	*n* = 46	*n* = 13	*n* = 29	*n* = 20	*n* = 19
Noverbal IQ[a]	**52.4 (12.2)[a]**	**65.1 (14.7)[b]**	75.3 (22.4)[c]	**53.2 (10.3)[a]**	113.3 (10.9)[d]
	36–79	**42–89**	40–137	**38–73**	98–139
Receptive vocabulary[b]	**62.4 (16.0)[a]**	**75.4 (15.4)[b]**	74.5 (18.2)[b]	56.2 (12.7)[a]	113.7 (10.7)[c]
	23–103	**39–97**	35–114	35–73	89–127
Expressive vocabulary[c]	**51.4 (15.4)[a]**	**64.4 (16.4)[b]**	70.6 (19.8)[b]	52.2 (15.3)[a]	110.9 (9.4)[c]
	40–100	**40–79**	40–115	40–89	96–126
Females	*n* = 15	*n* = 27		*n* = 22	*n* = 18
Noverbal IQ[a]	**71.5 (19.3)[a]**	**88.9 (20.9)[b]**		**59.4 (10.2)[c]**	116.8 11.7[d]
	38–113	**48–135**		**42–77**	97–139
Receptive vocabulary[b]	**81.9 (21.0)[a]**	**95.1 (14.9)[b]**		60.1 (11.8)[c]	109.6 (11.7)[d]
	47–132	**66–123**		44–93	88–127
Expressive vocabulary[c]	**72.1 (21.0)[a]**	**94.5 (16.9)[b]**		52.4 (11.2)[c]	108.2 (9.3)[d]
	40–111	**59–126**		40–75	93–128

Note. Different superscripts within a row indicate significant differences at $p < .05$. If groups share the same letter, differences were not significant. Bold texts indicate significant sex differences at $p < .05$. FXS-ASD = fragile X syndrome with autism spectrum disorder; FXS-O = fragile X syndrome only; ASD-O = autism spectrum disorder only; DS = Down syndrome; TD = typical development.

[a]Leiter International Performance Scale–Revised. [b]Peabody Picture Vocabulary Test–III. [c]Expressive Vocabulary Test.

assessment than the clinical populations, resulting in differences in the sampling context across the groups. Consistent with the definition used by Roberts, Martin, et al. (2007), a turn was defined as an utterance or string of utterances that continued until the participant either voluntarily stopped speaking, was interrupted, there was a topic change, or there was a pause of greater than 5 s between utterances. Reliability for turn identification was 97.04%.

Pragmatic Language

ADOS language transcripts were analyzed for pragmatic abilities using two language sample analysis techniques: (a) a detailed hand-coding system for assessing key pragmatic skills in social-communicative interaction (Roberts, Martin, et al., 2007) and (b) SALT (Miller & Iglesias, 2008). The ADOS provides an ideal context for seminaturalistic language sampling given that the administration is consistent across participants, yet simultaneously allows for flexibility in following the child's lead (Tager-Flusberg et al., 2009). Further, as noted above, only certain activities from the ADOS deemed to be most representative of natural social-communicative interaction were coded. Both hand-coding and SALT were completed based on the same language samples, but transcripts differed slightly in length in order to conservatively provide leeway for hand-coding analyses (i.e., transcribers were directed to transcribe 110 turns in case they miscounted the number of turns necessary for hand coding). In line with prior work (Roberts, Martin, et al., 2007), hand coding analyzed 100 intelligible turns (first 50 play based and 50 non–play based). Rather than manually edit all transcripts, all 110 intelligible turns were considered for SALT. All hand-coded and SALT variables were analyzed as proportions to account for the very minimal differences in transcript length (i.e., approximately 10 more turns used for SALT versus hand-coding analyses).

Hand coding. The hand-coding system used in this study was based on the system previously described in Roberts, Martin, et al. (2007). Transcripts for two boys with ASD-O and one girl with FXS-O included less than 100 but greater than 80 intelligible turns (81, 97, and 99 turns, respectively); these participants were retained in analyses given that the outcome variables were proportions. Two coders were trained extensively on this system and were responsible for all coding. Coders utilized the transcript and the video data. Twelve percent of the files were randomly selected and coded for reliability, with the mean intercoder agreement based on ICC greater than or equal to .94 for each of the variables, with the ICC breakdown by specific category as follows: noncontingent language (.94), perseveration (.95), initiations (.96), and nonresponsiveness (.96).

Noncontingent language was coded in one of two situations. First, noncontingency was coded if a child's turn did not clearly change the topic but failed to meet the informational expectation of the previous turn (e.g., the examiner says to the child, "I love to build sand castles," and the child says "Oh, thank you"). Second, noncontingent language was coded when a child's turn changed the topic abruptly, occurring without adequate pause time (at least 5 s) and/or before the previous topic had reached an obvious conclusion (e.g., the examiner asks the child, "Can I play with you for a little bit?" and the child responds, "Spoon and fork," in a context unrelated to play). The noncontingent language variable was derived by dividing all noncontingent turns by all turns.

Perseveration was coded when the participant repetitively used words, phrases, sentences, or topics either across

Table 3. Autism Diagnostic Observation Schedule (ADOS) module number by group.

Variable	FXS-ASD	FXS-O	ASD-O	DS	TD
Males	*n* = 46	*n* = 13	*n* = 29	*n* = 20	*n* = 19
ADOS module	23 M2, 23 M3	5 M2, 8 M3	11 M2, 18 M3	11 M2, 9 M3	9 M2, 10 M3
Females	*n* = 15	*n* = 27		*n* = 22	*n* = 18
ADOS module	8 M2, 7 M3	9 M2, 18 M3		12 M2, 10 M3	8 M2, 10 M3

Note. No significant differences between groups (all *p*s > .438). M2 and M3 refer to ADOS modules 2 and 3, respectively. FXS-ASD = fragile X syndrome with autism spectrum disorder; FXS-O = fragile X syndrome only; ASD-O = autism spectrum disorder only; DS = Down syndrome; TD = typical development.

utterances within a turn or across turns. Three occurrences were necessary before a code of perseveration was given, and the variable used in analyses was the proportion of turns containing perseveration. The example below illustrates a child (C) perseverating on dolls during an interaction with the examiner (E).

C: Go dolls. Go dolls. Go dolls.
E: Okay.
C: Go dolls.
E: You sound like my daughter. She doesn't play with dolls.
C: Go dolls.
E: She's kind of an athletic girl. I bet you're an athletic girl too. Is that right?
C: Go dolls.
…
E: So what do you like to do at home?
C: Play dolls.
E: Do you play outside?
C: Play dolls.

Initiations were nonobligatory turns; each turn was coded as being obligatory or nonobligatory. Obligatory turns followed a direct question or directive when a response was required (e.g., the examiner asks the child, "What kind of makeup do you wear?" and the child responds, "Oh, just like pink makeup."). In contrast, nonobligatory turns were self-initiated (e.g., the examiner says, "My dog knows she's not supposed to eat the cat food," and the child says, "Tell her not to be naughty."). The initiations variable was calculated by dividing all nonobligatory turns by all turns.

Nonresponsiveness was coded when the examiner made a direct request that required a turn from the child and the child did not respond within 3 s (e.g., the child does not respond to the question "What else do you like to play?"). This variable was calculated by dividing all nonresponses by the total of nonresponses and obligatory (required) responses.

SALT. SALT variables included self-repetition (a user-defined, manually entered exclusionary postcode for syntax analysis) along with two computational linguistic (automatic) variables, which are standard in SALT: (a) spontaneous utterance and (b) response to questions. These variables were selected based on their conceptual similarity to the hand-coding variables described above (no standard SALT variable corresponds to noncontingent language). Note that SALT variables are based on complete and intelligible utterances. Because hand coding was based on turns (which could contain one or multiple utterances), all variables were computed as proportions. With the exception of response to questions, proportions were calculated by dividing the variable of interest by the total number of complete and intelligible utterances. Response to questions was derived by dividing the total number of child responses to questions by the total number of examiner questions that were asked. Ten percent of files from each diagnostic group were transcribed for reliability. The average ICCs for SALT variables ranged from .83 to .95 across groups, with the ICC breakdown by specific category as follows: self-repetition (.83), spontaneous utterances (.87), and response to questions (.95).

Self-repetition (similar to the perseveration variable in the hand-coding scheme) was coded when an utterance was an exact, or nearly exact (i.e., differing only in pronunciation or addition of a filler), repetition of a previous utterance. This was a user-defined SALT code for excluding these utterances from syntax analysis, based on the recommendation of Scarborough (1990) and consistent with other research on children with neurodevelopmental disabilities (e.g., Estigarribia, Martin, & Roberts, 2012; Price et al., 2008; Roberts, Hennon, et al., 2007). To identify *spontaneous* (self-initiated) *utterances*, a complement to the hand-coding initiations variable, the SALT program identifies child utterances that do not follow examiner questions or intonation prompts (i.e., do not follow examiner utterances ending with a question mark or tilde). SALT identified *response to questions* by searching for child utterances that immediately followed a question from the examiner (i.e., examiner utterance ending with a question mark). This variable was calculated by dividing the number of child utterances that followed an examiner question by the total number of examiner questions asked. We chose this variable given its inverse relationship to the nonresponsiveness hand-coding variable.

Analysis Plan

To characterize pragmatic language profiles across the groups (boys and girls with FXS-ASD, FXS-O, DS, and TD and boys with ASD-O), we conducted a series of analysis of covariance (ANCOVA) models, controlling for

nonverbal mental age and structural language (receptive vocabulary age, expressive vocabulary age, and MLU). ANCOVAs addressed group differences for boys and girls separately and sex differences within groups (except for the ASD-O group, which included males only) on the hand-coding (noncontingency, perseveration, initiations, nonresponsiveness) and SALT (self-repetition, spontaneous utterance, response to questions) variables. Planned comparisons were conducted following each ANCOVA even when the overall model was nonsignificant given our hypotheses and to guard against Type 2 error. Effect sizes for differences were computed as Cohen's *d*. Consistent with J. Cohen (1988), an effect size of 0.2 was considered small, 0.5 was considered medium, and 0.8 was considered large.

Finally, for each of the nine groups and for all groups combined, we examined bivariate correlations between hand-coding and conceptually related SALT variables. Correlations were run with all groups combined (in addition to the groups separately) because the correlations addressed a predominately methodological question about the relationship between SALT and hand coding where all data points were considered valuable and to maximize sample size.

Results

Group Differences in Boys

Hand Coding

The overall model for noncontingent language in boys was significant, $F(4, 118) = 5.89$, $p < .001$, driven by increased noncontingent language in boys with FXS-ASD ($ps < .05$) and ASD-O ($ps < .05$) compared to boys with FXS-O, DS, and TD (ds 0.78-0.97). Similar findings emerged for perseveration, $F(4, 118) = 2.90$, $p = .025$. Specifically, boys with FXS-ASD and ASD-O were more perseverative than boys with DS ($ps < .05$, $ds = 0.63–0.67$) and FXS-O ($ps < .05$, $ds = 0.67–0.69$). No significant differences were detected between the boys with TD and the boys with FXS-ASD ($p = .177$, $d = 0.37$) or ASD-O ($p = .153$, $d = 0.43$). Significant differences were also found for initiations, $F(4, 118) = 3.14$, $p = .017$. Boys with ASD-O initiated less often than all the other groups ($ps < .05$, $ds = 0.60–1.10$), including boys with FXS-ASD ($p = .006$; $d = 0.67$). Significant differences for nonresponsiveness, $F(4, 118) = 3.21$, $p = .015$, were similarly driven by boys with ASD-O, as they were significantly more nonresponsive than boys with FXS-O and DS ($ps < .05$, $ds = 0.88–0.95$). Boys with ASD-O were also more nonresponsive than boys with FXS-ASD ($p = .055$, $d = 0.46$) and TD ($p = .136$, $d = 0.45$), although these differences were not statistically significant.

SALT

No group differences in self-repetition emerged, $F(4, 118) = 0.84$, $p = .501$; $ds = 0.01–0.46$. Findings for spontaneous utterances mirrored findings for the initiations hand-coding variable, $F(4, 118) = 3.80$, $p = .006$. Boys with ASD-O produced significantly fewer spontaneous utterances than all other groups ($ps < .05$, $ds = 0.57–1.10$). Findings for SALT variable of response to questions were similar to those found for the nonresponsiveness hand-coding variable, $F(4, 118) = 3.22$, $p = .015$, with the ASD-O group responding to questions significantly less often than boys with FXS-O ($p = .020$, $d = 0.81$), DS ($p = .004$, $d = 0.88$), and FXS-ASD ($p = .005$, $d = 0.69$), but not differing significantly from boys with TD ($p = .505$, $d = 0.20$). See Table 4 for comparisons of male groups.

Group Differences in Girls

Hand Coding

For girls, the overall model for noncontingency was marginally significant, $F(3, 74) = 2.43$, $p = .072$. Planned pairwise comparisons indicated that girls with FXS-ASD used significantly more noncontingent language compared with all other groups ($ps < .05$, $ds = 0.69–0.84$). The overall model for perseveration was also not significant, $F(3, 74) = 1.96$, $p = .128$, but planned comparisons showed that girls with FXS-ASD were significantly more perseverative than girls with DS ($p = .024$, $d = 0.79$). Girls with FXS-ASD also used more perseveration than girls with TD ($p = .099$, $d = 0.59$), although this difference was not significant. The overall model for initiations was marginally significant, $F(3, 74) = 2.65$, $p = .055$, in girls. Pairwise comparisons indicated that girls with TD initiated significantly less than girls with FXS-ASD and DS ($ps < .05$, $ds = 0.84–0.85$) and marginally less than girls with FXS-O ($p = .078$, $d = 0.55$). The overall model for nonresponsiveness was significant, $F(3, 74) = 5.32$, $p = .002$, driven by girls with FXS-ASD and TD being more nonresponsive than girls with FXS-O ($ps < .011$, ds 0.87–0.90) and DS ($ps < .008$, ds 0.93–0.96).

SALT

There were no group differences for self-repetition, $F(3, 74) = 0.50$, $p = .681$, $ds = 0.02–0.35$. Findings for spontaneous utterances were similar to those found for the initiations hand-coding variable, $F(3, 74) = 2.72$, $p = .051$; the TD group had fewer spontaneous utterances compared with all other groups ($ps < .05$, $ds = 0.72–0.86$). Findings for response to questions were similar to those for the nonresponsiveness hand-coding variable, $F(3, 74) = 3.30$, $p = .025$; girls with TD responded to questions significantly less often than girls with FXS-O ($p = .026$, $d = 0.70$) and DS ($p = .003$, $d = 1.01$), although the comparisons between girls with FXS-ASD and FXS-O ($p = .846$, $d = 0.06$) and girls with FXS-ASD and DS ($p = .247$, $d = 0.39$) were not significant. See Table 5 for comparisons of female groups.

Sex Differences

Boys with TD initiated more than their female counterparts, $F(1, 31) = 6.22$, $p = .018$, $d = 0.82$, with this pattern marginally significant in boys and girls with FXS-O, $F(1, 34) = 3.96$, $p = .055$, $d = 0.67$. Complementary SALT analyses similarly revealed that boys with TD produced more spontaneous utterances than girls with TD, $F(1, 31) = 12.7$, $p = .001$, $d = 1.18$. Girls with FXS-ASD and

Table 4. Adjusted means and standard errors in males.

Variable	FXS-ASD *M* (*SE*)	FXS-O *M* (*SE*)	ASD-O *M* (*SE*)	DS *M* (*SE*)	TD *M* (*SE*)
Hand coding					
Noncontingent language	.18 (.01)[a]	.11 (.02)[b]	.18 (.02)[a]	.11 (.02)[b]	.10 (.02)[b]
Perseveration	.12 (.01)[a]	.07 (.02)[b]	.12 (.02)[a]	.07 (.02)[b]	.09 (.02)[a,b]
Initiations	.59 (.02)[a]	.63 (.03)[a]	.52 (.02)[b]	.58 (.03)[a]	.58 (.03)[a]
Nonresponsiveness	.05 (.01)[a,b]	.02 (.02)[a]	.08 (.01)[b]	.02 (.02)[a]	.05 (.02)[a,b]
SALT					
Self-repetition	.26 (.01)[a]	.27 (.02)[a]	.27 (.02)[a]	.28 (.02)[a]	.30 (.02)[a]
Spontaneous utterances	.72 (.02)[a]	.77 (.03)[a]	.66 (.02)[b]	.74 (.02)[a]	.75 (.03)[a]
Response to questions	.73 (.02)[a]	.74 (.03)[a]	.65 (.02)[b]	.75 (.03)[a]	.67 (.03)[a,b]

Note. If groups share the same letter, differences were not significant. Different superscripts within a row indicate significant differences at $p < .05$. FXS-ASD = fragile X syndrome with autism spectrum disorder; FXS-O = fragile X syndrome only; ASD-O = autism spectrum disorder only; DS = Down syndrome; TD = typical development; SALT = Systematic Analysis of Language Transcripts.

TD were more nonresponsive than boys with FXS-ASD, $F(1,55) = 5.43$, $p = .023$, $d = 0.69$, and TD, $F(1,31) = 5.00$, $p = .033$, $d = 0.69$, respectively. This finding mirrored the SALT finding for response to questions in the FXS-ASD group, $F(1, 55) = 5.13$, $p = .027$, $d = 0.67$, with a marginally significant similar pattern found for the TD group, $F(1, 31) = 3.38$, $p = .076$, $d = 0.60$. No other significant sex differences emerged.

Correlations Between Conceptually Related Hand-Coding and SALT Variables

With all groups combined, significant correlations were detected for all three pairs of conceptually related variables: perseveration and SALT self-repetition ($r = .31$, $p < .001$), initiations and SALT spontaneous utterances ($r = .82$, $p < .001$), and nonresponsiveness and SALT response to questions ($r = -.63$, $p < .001$). Note that the last correlation is negative, as expected, given the inverse relationship of the two variables. Examining the groups separately, perseveration and SALT self-repetition were significantly associated in boys with ASD-O ($r = .38$, $p = .043$), girls with DS ($r = .43$, $p = .049$), and girls with FXS-ASD ($r = .85$, $p < .001$) and marginally associated in girls with FXS-O ($r = .33$, $p = .095$). Initiations and SALT spontaneous utterances were significantly associated in all groups (rs > .62, ps < .002). Finally, nonresponsiveness and SALT response to questions were significantly associated among boys with ASD-O and DS ($r = -.64$, $p < .001$; $r = -.63$, $p = .003$), boys and girls with FXS-ASD ($r = -.69$, $p < .001$; $r = -.83$, $p < .001$), and girls with TD and FXS-O ($r = -.84$, $p < .001$; $r = -.42$, $p = .028$) and marginally associated in girls with DS ($r = -.36$, $p = .099$).

Discussion

This study applied both a detailed pragmatic language hand-coding system and semiautomated language analysis program to characterize pragmatic profiles across boys and girls with fragile X syndrome with and without autism spectrum disorder (FXS-ASD, FXS-O), Down syndrome (DS), and typical development (TD) and boys with idiopathic ASD (ASD-O). Sex differences were also examined to further inform the nature of pragmatic impairments across neurodevelopmental conditions. Results suggest important areas of overlap and divergence across groups and indicate relatively strong convergence across the two language sample analysis techniques with some exceptions.

Group and Sex Differences (Hand Coding)

Consistent with previous research findings (Capps et al., 1998; Losh & Capps, 2003; Roberts, Martin et al., 2007; Tager-Flusberg & Anderson, 1991), boys with FXS-ASD and boys with ASD-O used more noncontingent language compared to boys with FXS-O, DS, and TD, with medium to large effect sizes. This was the first study to examine noncontingent language in girls with FXS, and similarly, girls with FXS-ASD were more noncontingent than girls in all other groups, again with medium to large effect sizes. Findings for perseveration largely mirrored those for noncontingent language, with boys with FXS-ASD and ASD-O using more perseveration than boys with FXS-O and DS (consistent with previous research on males; G. E. Martin et al., 2012; Ross, 2002) and girls with FXS-ASD using more perseveration than girls with DS, all with medium effect sizes. Although girls with FXS-ASD did not differ significantly from girls with TD on perseveration, a medium effect size suggests that we may have also found a significant difference between these two groups with larger samples. Together, these results suggest that increased noncontingency and perseveration may be defining characteristics of the pragmatic language phenotype of ASD regardless of FXS status or, potentially, sex (inclusion of girls with ASD-O in future studies will help to clarify the latter point).

Despite the meaningful areas of pragmatic language overlap between the two male ASD groups, some important differences also emerged. Boys with ASD-O initiated turns less often than boys in all other groups, including boys with FXS-ASD, with medium to large effect sizes. They were also less responsive than boys with FXS-O and

Table 5. Adjusted means and standard errors in females.

Variable	FXS-ASD *M* (*SE*)	FXS-O *M* (*SE*)	DS *M* (*SE*)	TD *M* (*SE*)
Hand coding				
Noncontingent language	.15 (.02)[a]	.09 (.02)[b]	.09 (.02)[b]	.08 (.02)[b]
Perseveration	.08 (.02)[a]	.06 (.01)[a,b]	.03 (.02)[b]	.04 (.02)[a,b]
Initiations	.60 (.03)[a]	.56 (.03)[a,b]	.60 (.03)[a]	.49 (.03)[b]
Nonresponsiveness	.09 (.02)[a]	.03 (.01)[b]	.03 (.02)[b]	.09 (.02)[a]
SALT				
Self-repetition	.23 (.02)[a]	.23 (.02)[a]	.24 (.02)[a]	.26 (.02)[a]
Spontaneous utterances	.74 (.03)[a]	.73 (.03)[a]	.74 (.03)[a]	.64 (.03)[b]
Response to questions	.68 (.04)[a]	.69 (.03)[a]	.73 (.03)[a]	.59 (.03)[b]

Note. Different superscripts within a row indicate significant differences at $p < .05$. If groups share the same letter, differences were not significant. FXS-ASD = fragile X syndrome with autism spectrum disorder; FXS-O = fragile X syndrome only; DS = Down syndrome; TD = typical development; SALT = Systematic Analysis of Language Transcripts.

DS, with large effect sizes, whereas boys with FXS-ASD did not differ from either of these groups in nonresponsiveness. For boys with ASD-O, these results are consistent with prior reports of reduced initiations and increased nonresponsiveness (Capps et al., 1998; Jackson et al., 2003; Loveland et al., 1988). These differences support a symptom-specific, or endophenotype-based, approach to understanding the overlap of FXS-ASD and ASD-O, in order to identify specific pragmatic features (e.g., noncontingent language and perseveration) that may be linked to the *FMR1* gene. Although boys with FXS-ASD did not show reduced responsiveness in the current study, girls with FXS-ASD were less responsive than girls with FXS-O and DS, with large effect sizes, and were also less responsive than boys with FXS-ASD (medium effect size), suggesting that clinical needs may be different for boys and girls with FXS-ASD. Girls with FXS-ASD and ASD-O should be compared in future studies to more fully understand the overlap of FXS-ASD and ASD-O.

Some findings were less consistent with previous research. Except for a marginal difference in initiations between males and females with FXS-O (although with a medium effect size suggesting that we may have been underpowered for this comparison), neither group of girls with FXS showed reduced initiations in this study, which is somewhat inconsistent with two studies in which females with FXS showed reduced initiations compared with female controls without FXS (Lesniak-Karpiak et al., 2003; Mazzocco et al., 2006). Of note, these previous studies used role-play conversations with an unfamiliar adult (acting as a "stranger" who was minimally responsive and affectively neutral), and so our findings are not directly comparable to this more contrived situation. Instead, girls in our study interacted with a trained examiner who was responsive and supportive and followed the child's lead. Together, discrepant findings across studies suggest that, not surprisingly, the interaction style of the communication partner may have an important impact on a child's performance.

Boys and girls with DS did not show any deficits on any of the variables measured in this study. This is consistent with previous studies of noncontingent language and perseveration in boys (G. E. Martin et al., 2012; Roberts, Martin, et al., 2007; Tager-Flusberg & Anderson, 1991). Results are somewhat inconsistent with those of Lee et al. (2017) who reported more inappropriate topic shifts in girls with DS than in boys based on a single item from an observational rating scale, with a sample partially overlapping with the one studied here. That study employed a rating scale based on videotaped interactions only and required a relatively low threshold for capturing noncontingent language, where only two striking instances of inappropriate topic shifting over the course of the entire language sample would result in a participant reaching the maximum score possible for inappropriate topic shifts. The current study, on the other hand, considered the proportion of all turns that included noncontingent language and was based on detailed annotated and verbatim transcripts of these interactions.

Finally, girls with TD initiated less (large effect size) and were more nonresponsive (medium effect size) than boys with TD. Previous research with preschool and school-age children with TD has also shown that boys are more likely to initiate than girls (Berghout et al., 1987; Cook et al., 1985). More surprisingly, in this study, girls with TD also initiated less frequently than girls with FXS-ASD and DS with large effect sizes (and marginally less than girls with FXS-O with a medium effect size) and were more nonresponsive than girls with FXS-O and DS (large effect sizes). As pragmatic sex differences in TD may be more sociocultural rather than biological in nature (Sigelman & Holtz, 2013), perhaps girls with FXS and DS are not picking up on social cues or being provided the same social cues as girls with TD. It is also possible that some girls with FXS or DS are more likely to initiate to control the conversation, thereby making it more predictable for them in light of cognitive or language deficits (i.e., a compensatory strategy).

Comparison of Two Language Sample Analysis Techniques

This study utilized a multimethod approach to language sample analysis for characterizing pragmatic language and

to evaluate the utility of a more efficient semiautomated technique (SALT) relative to gold standard hand coding. The hand-coding methodology was the primary focus of analyses and highlighted group and sex differences across all variables. Semiautomated SALT analyses, which relied on the language transcripts only, converged with some but not all of the hand-coding findings. Analyses with SALT were generally consistent in detecting differences for the automated analysis of initiations and responses, with similar effect sizes as those found for hand coding in most cases. Aside from noncontingent language, for which there was no complementary SALT variable, one notable exception to this trend of relatively strong alignment was perseveration (or "self-repetition" in SALT, a user-defined code). In fact, effect sizes for group comparisons on SALT self-repetition were all negligible to small with no significant differences detected, contrasting with medium effect sizes for several significant group comparisons for perseveration as described above. However, even though group differences on SALT self-repetition variable were not detected, perseveration and self-repetition were significantly correlated for all groups combined, as well as in boys with ASD-O and girls with FXS-ASD (and in girls with DS for whom perseveration was not found to be pronounced), suggesting that both variables are picking up on similar language features in these groups. Notably, the same relationship was not found for boys with FXS-ASD, suggesting that the two variables are not tapping the same behavior for this group in particular (our largest group, indicating that the absence of a significant correlation is not due to a lack of statistical power). Perseveration in the hand-coding system allowed for more variable expression of repetitive language, including repetition of topics through different word combinations (versus the more strict definition of SALT self-repetition, an exclusionary user-defined code for syntax analysis, which required an identical or nearly identical repetition). In addition, two repetitions were necessary before a code of perseveration was given in the hand-coding system because perseveration should be "excessive," whereas just one repetition was necessary for the SALT code. Together, findings suggest that standard transcription procedures may yield valuable information for some aspects of pragmatic language that can be measured through semiautomated techniques such as SALT but that the more time-intensive approach of hand coding is necessary for accurately capturing perseveration and noncontingent language.

Study Strengths, Limitations, and Directions

This study has several important strengths. We examined pragmatic language in three genetically based neurodevelopmental disabilities, including a relatively large group of children with FXS, the rarest of the conditions included in this study. We applied two complementary language sample analysis techniques to compare an array of pragmatic language skills across these different syndromes and controlled for nonverbal mental age and structural language to isolate pragmatic ability in particular. Importantly, we also investigated the overlap of FXS and ASD by comparing boys with FXS-ASD and boys with ASD-O and considered sex differences and overlap in the FXS and DS groups, contributing to a very small body of existing work on sex-specific patterns in these populations.

Some limitations and directions for research include our focus on examiner–child interactions only, which may not be representative of a child's performance in other situations. Future studies should examine pragmatic skills with a range of communication partners, including caregivers and peers, to more fully understand the strengths and needs of children with FXS, ASD, and DS. It will also be important for future work to include girls with ASD-O to increase our understanding of sex differences in idiopathic ASD and the overlap of FXS and ASD. Additionally, although this study attempted to take a comprehensive approach to describing group differences in pragmatic profiles, we did not examine potential underlying mechanisms that could contribute to pragmatic deficits (and, perhaps, differentially) across groups, beyond controlling for mental age and structural language. For example, deficits in theory of mind and executive function have often been linked to pragmatic difficulties in idiopathic ASD (e.g., Baron-Cohen, Leslie, & Frith, 1985; Joseph, 1999; I. Martin & McDonald, 2003), and anxiety and excessive arousal have been argued to account for pragmatic deficits in FXS (e.g., Belser & Sudhalter, 1995; I. L. Cohen, 1995; I. L. Cohen, Vietze, et al., 1989; Cornish, Sudhalter, & Turk, 2004). Identifying underlying mechanisms of pragmatic difficulties and whether they differ across groups, even for similar atypical pragmatic features (e.g., noncontingent language and perseveration in FXS-ASD and ASD-O), would inform intervention planning.

Clinical Implications

These findings have several important clinical implications. Although pragmatic impairment by definition is present in all those who meet criteria for ASD, our findings highlight the complexity of the expression of pragmatic impairment in ASD by showing areas of both similarity and difference in idiopathic and syndromic ASD (ASD-O and FXS-ASD, respectively). Although both male ASD groups showed increased noncontingent language and perseveration, only those with ASD-O showed difficulties initiating conversational turns and responding to conversational bids. These differences suggest potentially different targets for intervention depending on the etiology of ASD and, perhaps, indicate more social motivation in boys with FXS-ASD, which may prove to be a positive prognostic indicator for treatment success in this group. Girls with FXS-ASD on the other hand were more nonresponsive than their male counterparts, indicating that clinicians should also consider the role of sex in assessment and intervention for children with FXS.

In this study, we used the ADOS as our language sample and understand that not all clinicians will be trained

in the ADOS. However, the ADOS is similar to a traditional language sample in many ways (where the adult will comment, avoid too many yes/no questions, follow the child's lead, etc.) and, as such, has been recommended as a language-sampling context (Tager-Flusberg et al., 2009). These findings are therefore likely to extend to similar conversational language elicitation contexts. Clinicians should also see Timler (2018) for a detailed description of a language-sampling protocol for school-age children and adolescents.

Finally, whereas both hand-coding and SALT approaches were generally consistent in detecting differences for initiations and responses, the more detailed, time-intensive hand-coding approach was necessary for detecting differences in perseveration (and, of course, noncontingent language, for which there was no corresponding SALT code). Although hand-coding efforts such as those employed here may not be feasible for a clinician, custom coding (i.e., user-defined codes) is possible in SALT, as evidenced by our self-repetition variable. Clinicians may use definitions of perseveration and noncontingent language provided in the current article to create custom codes as part of a comprehensive language assessment. If SALT is unavailable to a clinician, alternative strategies for a language sample analysis for assessing pragmatics have been described in the literature (e.g., Timler, 2018), and our findings, of course, also show that one can examine these features using hand coding given the time to do so.

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