Research Article

A Duck Wearing Boots?! Pragmatic Language Strategies for Repairing Communication Breakdowns Across Genetically Based Neurodevelopmental Disabilities

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Purpose: The ability to repair breakdowns in communication is an important pragmatic language skill that helps to maintain clear and meaningful interactions. Examining this ability in genetically based neurodevelopmental disabilities in which pragmatics are affected can provide important information about the precise pragmatic skills impacted across different populations and also help to identify core mechanisms underlying pragmatic impairment that may inform tailored interventions.

Method: Individuals with idiopathic autism spectrum disorder (ASD-O; $n = 40$), fragile X syndrome with comorbid autism spectrum disorder (FXS-ASD; $n = 62$), FXS without ASD (FXS-O; $n = 38$), Down syndrome (DS; $n = 43$), and typical development (TD; $n = 42$) completed a picture description task in which an examiner prompted for clarification repeatedly to elicit communication repair attempts. Participants' response strategies were compared across diagnostic groups and by sex and examined in relationship to different cognitive abilities.

Results: Relatively few group differences were observed in responses to requests for clarification overall. Males with ASD-O responded less to clarification requests than males with FXS-ASD and FXS-O, and males with FXS-ASD responded more inappropriately than males with ASD-O and DS. All male groups became less responsive to prompts for communication repair across the series of requests. Males with TD and FXS-ASD used less effective strategies than females.

Conclusion: All groups showed some proficiency in repairing communication breakdowns, although individuals with ASD-O and FXS-ASD demonstrated some key areas of difficulty, highlighting the importance of considering ASD symptomatology in assessment and treatment of males with FXS. Findings also suggest that, across groups, multiple requests for clarification may lead to disengagement from the interaction. Finally, correlated skills observed across groups could implicate different underlying skills related to communication repair abilities across groups.

In communicative exchanges, misunderstood messages are typically met with a request for clarification (e.g., “what?” and “what do you mean?”). Such requests prompt the speaker to adjust his or her message to accommodate the listener's needs and establish shared understanding. This ability, known as *communication repair*, is an important pragmatic (i.e., social) language skill necessary

for successful exchange of information and effective and meaningful social interactions. A speaker may use a variety of strategies in responding to a request for clarification, varying in levels of specification and sophistication (e.g., repeating the utterance, revising grammatical structure, adding semantic information). In typical development (TD), repair skills emerge as young as 15 to 30 months of age, when the most common strategy is repetition of a message (Anselmi, Tomasello, & Acunzo, 1986; Gallagher, 1977, 1981). As children continue to gain expressive language skills, they begin to use more sophisticated strategies. For example, at age 5–6 years, children begin revising statements (e.g., “The boy is being chased by the dog” to “The dog is chasing the boy”) and, by age 8–9 years, begin adding new information (e.g., “A boy” to “A boy is riding a horse”; Brinton, Fujiki, Loeb, & Winkler, 1986; Gallagher, 1977).

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Communication repair skills can pose a particular challenge for individuals with neurodevelopmental disabilities impacting language, where misunderstandings may be frequent due to impairments in speech and language skills, as well as the underlying cognitive abilities that support communication (e.g., theory of mind [ToM]; Brady, Steeples, & Fleming, 2005; Brinton & Fujiki, 1991; Meadan, Halle, Watkins, & Chadsey, 2006). Given the potential for misunderstanding during conversational interactions, it is critical to understand whether individuals with different neurodevelopmental disabilities are able to employ repair strategies and, if so, the types of strategies utilized. This study examined communication repair abilities in verbal children and adolescents across three genetically based neurodevelopmental disabilities: idiopathic autism spectrum disorder (ASD-Only, or ASD-O), fragile X syndrome with and without comorbid autism spectrum disorder (FXS-ASD and FXS-O, respectively), Down syndrome (DS), and controls with TD.

Pragmatics and Communication Repair in Different Neurodevelopmental Disabilities

Some prior work has examined communication repair abilities in individuals with neurodevelopmental disabilities. Perhaps, surprisingly (given that social communication impairment is a defining feature of autism spectrum disorder [ASD]), some evidence suggests that individuals with ASD do deploy strategies for repairing communication breakdowns when explicitly prompted, including adding more information as breakdowns persist. However, those repair attempts were also accompanied by ineffective strategies, such as making off-topic statements or attempting to discontinue the conversation, and responses tended to be less specific or elaborative (Geller, 1998; Paul & Cohen, 1984; Volden, 2004). Prior studies in ASD, however, have included relatively small sample sizes (e.g., ranging from five to nine participants per group) and have not considered potentially related abilities that might inform the origins of different groups' difficulties with repair abilities. For instance, ToM, or the ability to infer mental states, which is often impaired in ASD, may impact the ability to perceive and respond to a communicative partner's need for message clarification.

Investigations of communication repair ability have also focused on individuals with intellectual disability of various etiologies. In general, this work suggests that repair skills do not appear specifically impaired beyond the effect of general language delays (Brady et al., 2005; Levy, Tennebaum, & Ornoy, 2003). Studies of this pragmatic skill in specific genetic syndromes associated with intellectual disability, such as FXS and DS (the most common genetic causes of intellectual disability), have been more limited but are important to determine the different strengths and weaknesses that may be differentially impacted and targeted clinically. FXS, caused by a mutation on the *FMR1* gene on the X chromosome, is the leading single-gene cause of ASD, with up to 75% of males with FXS meeting

diagnostic criteria for ASD and showing overlap in the types of pragmatic language impairments observed in ASD-O (Klusek, Martin, & Losh, 2014a, 2014b; Lee, Martin, Berry-Kravis, & Losh, 2016; Losh, Martin, Klusek, & Hogan-Brown, 2012a). Differences in symptom expression have also been observed between FXS and nonsyndromic ASD, making it particularly important to define carefully those areas of overlap and divergence, in order to understand how *FMR1* may contribute to ASD-relevant symptomatology. In this vein, and in line with prior work (see Abbeduto, McDuffie, & Thurman, 2014; Martin et al., 2017), this study employed a component-feature approach to further clarify specific pragmatic skills (i.e., communication repair) that may be differentially impacted in FXS and ASD, as well as DS. The limited studies on communication breakdown in FXS and DS have focused primarily on the ability to signal noncomprehension (confusion) when presented with unclear messages. For example, studies found that individuals with DS (Abbeduto et al., 2008; Martin et al., 2017) and FXS (Abbeduto et al., 2008; Thurman, Kover, Brown, Harvey, & Abbeduto, 2017) were less likely than TD controls to signal noncomprehension of a confusing message, with additional evidence that comorbid ASD negatively impacts this skill in FXS (Martin et al., 2017). No studies other than the study by Levy et al. (2003), which included one male with FXS, have examined the ability of individuals with FXS to repair their own statements, and only one case study of four children has investigated repair skills in DS, with findings showing appropriate responses to requests for clarification (Coggins & Stoel-Gammon, 1982).

Equally important to clarifying communication repair skill across neurodevelopmental disabilities is to examine sex differences within groups. No studies have examined sex differences in repair skills among individuals with neurodevelopmental disabilities, and only a few have examined sex differences in broader pragmatic language skills, generally producing inconsistent results. In FXS, females are typically less affected phenotypically with lower rates of ASD (owing to the presence of an unaffected copy of *FMR1* on females' second X chromosome; e.g., Clifford et al., 2007; Hagerman et al., 1992; Hall, Lightbody, Hirt, Rezvani, & Reiss, 2010; Klusek et al., 2014b; Lee et al., 2016). Therefore, it may be likely that females with FXS show a distinct pattern of pragmatic strengths and weaknesses from males, although evidence on the impact of sex on noncomprehension signaling (another pragmatic skill related to communication breakdowns, described above) is mixed (Abbeduto et al., 2008; Martin et al., 2017; Thurman et al., 2017). In DS, one study found that a small sample of parents reported higher rates of pragmatic impairment in males (Berglund, Eriksson, & Johansson, 2001), whereas a more recent study with an older age group found no differences in parent report of pragmatic skills (Lee et al., 2017). Lee et al. (2017) also reported no differences in overall pragmatic language as rated during a seminaturalistic interaction but some differences on ratings of more specific pragmatic skills, such as excessive detail, a higher frequency

of inappropriate topic shifts, and reduced eye contact in females when compared with males with DS. Finally, no differences were found between males and females with DS in their ability to signal noncomprehension in one study (Martin et al., 2017). Further research is needed to better understand potential sex differences in discrete pragmatic language skills across neurodevelopmental disabilities.

Abilities Associated With Communication Repair

The ability to repair communication breakdowns is supported by important developments in cognition and structural language (e.g., the ability to use different syntactic forms and vocabulary terms when revising messages). Examining such correlated abilities may help to reveal the origins of potential impairments in communication repair. For instance, ToM is critical for pragmatic competence in general (Happé, 1997) and accommodating a listener's needs and repairing communication breakdown in particular (Baron-Cohen, Wheelwright, Hill, Raste, & Plumb, 2001; Brady, McLean, McLean, & Johnston, 1995; Brady et al., 2005). First, in order to decipher why a message was misinterpreted, a speaker must evaluate the listener's state of knowledge and recognize differences with his or her own knowledge. Second, the speaker must adjust his or her message to match the listener's knowledge (Brady et al., 1995, 2005; Feldman & Kalmar, 1996). Being able to change repair strategies as a breakdown persists (e.g., across a series of prompts for clarification) requires the speaker to continually evaluate whether messages were successfully received, discard ineffective strategies, and adopt new strategies to assist the listener (Brinton & Fujiki, 1991; Volden, 2004).

ToM deficits are well documented in ASD-O (see Baron-Cohen, 2000, for a review) and have been repeatedly linked to pragmatic difficulties in ASD (Capps, Kehres, & Sigman, 1998; Losh & Capps, 2003, 2006; Losh et al., 2012a; Losh, Martin, Klusek, Hogan-Brown, & Sideris, 2012b; Tager-Flusberg & Sullivan, 1995). ToM impairments have also been reported in both FXS and DS (Abbeduto et al., 2001; Garner, Callias, & Turk, 1999; Grant, Apperley, & Oliver, 2007; Lewis et al., 2006; Losh et al., 2012b), but few studies have examined these abilities relative to pragmatic ability (Abbeduto et al., 2001; Losh, et al., 2012b). Further, ToM deficits have not previously been examined in relationship to communication repair in ASD, FXS, or DS, though, interestingly, associations between ToM and communication repair have been reported in patients with schizophrenia (Bosco, Bono, & Bara, 2012).

Present Study Objective

This study investigates how individuals with ASD-O, FXS-ASD, FXS-O, DS, and TD contend with communication breakdowns using a task designed specifically to probe for different types of communication repair strategies,

such as repeating a statement, adding novel information, or using nonverbal or paralinguistic strategies to emphasize speech (Brinton & Fujiki, 1991; Brinton et al., 1986). This task assesses an individual's ability to respond to persistent clarification requests, which has been argued to be more comparable to daily interactions than a response to a single request (Volden, 2004). Prior studies of individuals with TD, language disorders, and ASD demonstrate the task's validity and its value in teasing apart the constituent factors contributing to broader pragmatic difficulties (Brinton & Fujiki, 1991; Brinton et al., 1986; Volden, 2004). This study included neurodevelopmental disabilities where response to communication repair has not been studied previously (FXS, DS) and incorporated assessment of related abilities, such as ToM in all groups. Both males and females (in all groups except ASD-O) were also included to better understand the (potentially distinct) expression of different pragmatic skills across sexes.

Three primary questions were addressed: (a) Do strategies used to repair communication breakdown vary by group (ASD-O, FXS-ASD, FXS-O, DS, and TD) or sex? (b) Within and across groups, do strategies vary across a persistent breakdown (i.e., a series of requests for clarification—huh? What? I don't understand)? and (c) How do measures of cognition, language, and ToM relate to communication repair strategies utilized across groups? On the basis of reports of elevated rates of pragmatic language impairments in ASD-O and FXS-ASD (Klusek, Martin, & Losh, 2013; Klusek et al., 2014a; Losh et al., 2012a; Martin, Losh, Estigarribia, Sideris, & Roberts, 2013), we predicted that individuals with ASD (FXS-ASD and ASD-O) would produce more inappropriate responses and show less sophisticated repair strategies over successive prompts relative to all other groups. We also expected that females with FXS would show fewer differences than males, although predictions for sex differences in other groups were less clear. Finally, given the importance of inferring a communication partner's mental state in clarifying misunderstandings and on the basis of prior research demonstrating strong links between pragmatic language and ToM in typical and atypical development (Hughes & Leekam, 2004; Losh & Capps, 2003, 2006; Losh et al., 2012b; Milligan, Astington, & Dack, 2007), we predicted that better performance on a battery of ToM tasks would relate to use of more sophisticated strategies to repair the communication breakdown.

Method

Participants

A group totaling 225 children and adolescents participated, including individuals with ASD-O (40 males), FXS-ASD (50 males, 12 females), FXS-O (13 males, 25 females), DS (22 males, 21 females), and TD (22 males, 20 females). Participants were part of a broader longitudinal study of pragmatic development in ASD, FXS, DS, and TD (Hogan-Brown, Losh, Martin, & Mueffelman,

2013; Klusek et al., 2013, 2014a, 2014b; Lee et al., 2017; 2016; Losh et al., 2012b; Martin et al., 2017). For the larger study, individuals were tested annually up to three times. The communication repair task was administered at the first and third time point, although only the first time point is reported in the current paper, with concurrent measures of cognition, social cognition (i.e., ToM), and language. Data from females with ASD-O were not available as the study was not funded to include this group. Eligibility criteria for the study included combining three or more words, speaking English as the first and primary language in the home, and passing a hearing screening (excluded if threshold greater than 30 dB HL across 500, 1000, 2000, and 4000 Hz in the better ear). Participants with DS and TD were initially screened for ASD using the Social Communication Questionnaire (Rutter, Bailey, & Lord, 2003) and excluded from analyses if they met ASD criteria on the Autism Diagnostic Observation Schedule (ADOS, described below; Lord, Rutter, DiLavore, & Risi, 2001) at any time point.

Table 1 provides a demographic breakdown of the groups. Groups differed across measures of cognition and language; thus, all analyses controlled for mental age equivalent (Leiter International Performance Scale–Revised; Roid & Miller, 1997), receptive vocabulary age equivalent (Peabody Picture Vocabulary Test–Third Edition; Dunn & Dunn, 1997, or Peabody Picture Vocabulary Test–Fourth Edition; Dunn & Dunn, 2007), expressive vocabulary age equivalent (Expressive Vocabulary Test; Williams, 1997), and mean length of utterance (MLU) obtained from a conversational sample from the ADOS. Males with ASD-O had greater nonverbal mental ages than all other males ($ps < .05$). Females with FXS-O

had greater nonverbal mental ages than females with DS and FXS-ASD ($ps < .01$). Males with ASD-O had greater expressive vocabulary age equivalents than males with DS and FXS-ASD ($ps < .05$), and males with TD had greater expressive vocabulary age equivalents than males with FXS-ASD ($p < .05$). Females with FXS-O had greater expressive vocabulary age equivalents than all other female diagnostic groups ($ps < .01$). Males with FXS-O had greater receptive vocabulary age equivalents than males with DS and TD ($ps < .05$). Females with FXS-O had greater mean receptive vocabulary age equivalences compared with females with DS and FXS-ASD ($ps < .05$). Males with TD had a greater MLU than all other male diagnostic groups ($ps < .05$). Males with DS had a lower MLU than males with ASD-O ($p < .001$) and FXS-O ($p < .01$). Males with FXS-ASD had a lower MLU than males with FXS-O ($p < .05$) and DS ($p < .001$). Males with DS and FXS-ASD were significantly older than their female counterparts ($ps < .05$). Females with FXS-O had significantly greater nonverbal and expressive language age equivalents and MLU than males with FXS-O ($ps < .05$). Participants were recruited from schools, genetic clinics, developmental clinics, physicians' offices, and research registries. The study protocol was reviewed and approved by institutional review boards at the University of North Carolina and Northwestern University.

Procedure

Testing took place in a research laboratory or in a quiet room in participants' homes or schools. All assessments

Table 1. Participant characteristics by group.

Category	M (SD) Range								
	ASD-O	FXS-ASD		FXS-O		DS		TD	
	Males (n = 40)	Males (n = 50)	Females (n = 12)	Males (n = 13)	Females (n = 25)	Males (n = 22)	Females (n = 21)	Males (n = 22)	Females (n = 20)
Chronological age	9.1 (2.6) 4.2–13.3	10.7 (2.4) 6.6–15.1	8.7 (4.1) 4.5–15.9	9.7 (3.3) 6.1–15.0	9.2 (3.7) 4.2–14.9	10.9 (2.1) 6.8–14.9	9.0 (2.3) 6.0–14.2	5.0 (1.4) 3.3–8.8	5.4 (2.4) 3.2–11.8
Leiter	7.1 (3.2) 3.9–19.7	5.0 (0.5) 3.5–6.0	5.1 (0.8) 4.0–6.1	5.5 (1.0) 4.4–8.3	7.0 (2.3) 3.9–12.8	5.3 (0.8) 4.3–8.3	5.0 (0.8) 3.8–6.8	5.5 (1.5) 3.6–9.2	6.1 (2.7) 3.9–14.9
EVT	6.3 (2.2) 3.4–13.8	5.0 (1.0) 2.7–7.3	5.6 (1.5) 3.3–7.6	5.4 (1.5) 3.8–9.3	8.4 (3.9) 4.1–19.8	5.3 (1.3) 3.6–8.6	4.6 (1.5) 1.9–7.8	6.0 (2.1) 2.9–12.3	6.1 (2.4) 3.8–12.2
PPVT	6.8 (3.0) 3.1–17.0	5.7 (1.3) 2.4–8.8	6.4 (2.0) 2.4–8.6	6.5 (2.6) 3.4–13.8	8.3 (3.4) 3.2–15.5	5.2 (1.4) 2.4–7.5	4.8 (1.9) 2.1–9.8	6.3 (2.0) 2.2–11.6	6.4 (3.0) 2.7–16.1
MLU	4.3 (1.1) 2.2–6.4	3.5 (0.7) 1.8–4.9	3.9 (1.2) 2.3–5.6	4.0 (0.73) 2.3–4.7	4.7 (1.1) 3.2–7.1	4.8 (0.7) 1.8–4.8	3.3 (1.0) 2.3–6.6	3.5 (0.7) 3.1–6.1	5.3 (1.5) 3.2–7.9

Note. ASD-O = ASD only; FXS-ASD = fragile X syndrome with autism spectrum disorder; FXS-O = FXS only; DS = Down syndrome; TD = typical development. Leiter = Leiter–Revised (Nonverbal Mental Age); EVT = Expressive Vocabulary Test (Age Equivalent); PPVT = Peabody Picture Vocabulary Test (Age Equivalent); MLU = mean length of utterance.

were audiotaped with a Marantz portable solid-state recorder (PMD670) and videotaped.

ASD Classification

The ADOS (Lord, Rutter, DiLavore, & Risi, 2001) was used to classify ASD. The ADOS is a gold-standard diagnostic measure of ASD that consists of a series of structured and semistructured interactions between an examiner and subject. Examiners were trained either with the instrument's developers or through intralab training. Coders had previously achieved at least 80% reliability with either the instrument developers or through intralab training in accordance with the standards of the instrument developers. ASD classification was determined using the ADOS-2 revised algorithm 10-point severity score, which derives a cutoff (score of 4) for ASD classification (Gotham, Pickles, & Lord, 2009). Consistent with published work (i.e., Klusek et al., 2014a; Martin et al., 2017), all available ADOS assessments from the larger longitudinal study were considered to determine ASD status. The average severity score across time points was considered the most valid and conservative representation of ASD status given the longitudinal nature of the larger study.

Communication Repair Task

To provide a context for communication breakdown, participants were asked to describe a series of nonsensical pictures to an examiner (e.g., a pig vacuuming) who ostensibly could not see the picture. The pictures were drawn from Webber's "What's Wacky?" language cards intended for grades prekindergarten through fifth grade (Webber, 1998) and converted to electronic images for presentation on a computer screen. Each participant was administered eight experimental and 10 control pictures. All 18 stimuli were randomly ordered into four different sets, and participants were randomly assigned an item set. During the control condition, the examiner immediately indicated understanding (e.g., "oh that's so silly!"). During the experimental trials, following the child's description, the examiner feigned a misunderstanding through a series of three prompts requesting clarification ("Huh?" "What?" "I don't understand"). Based on administration procedures developed by Brinton et al. (1986), the stacked sequence of prompts was intended to simulate a persistent breakdown in communication in order to elicit strategies in an attempt to reach mutual understanding between the speaker and listener. After the participant responded to the third prompt, the examiner acted as though he or she finally understood the content of the picture (e.g., "Oh, I get it!"). During the control condition, the examiner simply acknowledged the participant's description.

Videos were transcribed by blinded research assistants trained to 80% or higher morpheme-to-morpheme reliability. Trials were not coded for repair strategy type if any component was unintelligible. Coding was conducted blind to group status and based on both transcripts and video. A second coder independently scored samples from 10% of each diagnostic group for reliability assessment. The intraclass

correlation coefficient across groups for all prompts across all strategy types was .982. Coding was adapted from Brinton et al. (1986) and Volden (2004). In response to a request for clarification, the categories below were coded. Codes were categorized into nonresponse, verbal repair strategies, and supplementary repair strategies. For all codes but nonresponse (see description below), proportions of repair type were calculated by dividing the total instance of each specific repair strategy by codable (i.e., intelligible) responses.

Nonresponse: Does not respond to a request for clarification within 5 s of the request. This proportion was calculated by dividing the number of instances a participant did not respond by the total examiner prompts for clarification.

Verbal strategies (presented in order of sophistication, low to high):

1. **Inappropriate** (includes all three, below):
Look. Directs the examiner to look at the screen despite knowing that the examiner is not allowed to look.
Off-Topic. Statements that do not relate to the image or task at hand are made, such as "it's raining outside."
Resistance. Resistance to respond to the task instructions, such as "move to the next picture."
2. **Repetition:** Repeats one or several word(s) present in prior responses without adding new information, using a new grammatical structure, or making the original description more grammatically correct. For example, "Girl on swing" to "Girl" or "Girl on swing."
3. **Revision:** Holds semantic content/meaning constant but alters or corrects grammar of an utterance, such as using synonymous labels or different syntactic structure. For example, "He's on the helicopter swinging" to "He's swinging on the helicopter."
4. **Addition:** Adds specific, accurate semantic information. For example, "Horse and cowboy" to "Horse on the cowboy's back."
5. **Background:** Offers context that provides a framework in which prior responses could be interpreted. For example, "Birds are nesting in someone's hair" to "You know how barrettes that you put in your hair. She has birds in her hair."
6. **Metacomment:** Acknowledges or speaks about the process for repair, such as stating, "I don't know how to say it better."

Supplementary strategies:

1. **Suprasegmental:** Increases loudness, emphasizes words or word junctures, uses more precise articulation, or reduces rate of speech.
2. **Gesture:** Uses behaviors that represent objects or actions symbolically, such as pantomiming, or to direct attention, such as pointing.

To illustrate these various codes, Table 2 shows an example of an interaction between the examiner and child.

Cognition and Language

The Leiter International Performance Scale–Revised (Roid & Miller, 1997) was used to assess nonverbal mental age. The Expressive Vocabulary Test (Williams, 1997) and Peabody Picture Vocabulary Test–Third or Fourth Edition (Dunn & Dunn, 1997, 2007) were used to assess expressive and receptive vocabulary, respectively. Age equivalents, rather than standard scores, were used in analyses to account for cases when raw scores fell outside of the range of standard scores (e.g., floor effects). Grammatical development was indexed by MLU in morphemes (Brown, 1973) from language samples from the ADOS, transcribed using Systematic Analysis of Language Transcripts conventions (Miller & Chapman, 2008). Ten percent of files from the total sample for each diagnostic group were transcribed for reliability, and the average intraclass correlation coefficient was .972 for MLU across groups.

ToM

Participants completed one of two comparable batteries of tasks to measure ToM, depending on when they entered the study, that included assessment of perspective taking (i.e., recognizing that others may have a different perspective than oneself), diverse desires (i.e., recognizing that others may want something different than oneself), diverse beliefs (i.e., recognizing that others may have beliefs different than one's own), false belief (i.e., recognizing that others have beliefs that may contrast with reality), explicit false belief (i.e., recognizing how a person's actions may follow on the basis of their knowledge contrasting with reality), and knowledge access (i.e., recognizing the knowledge an individual possesses on the basis of the information that has been given to them in contrast to others; Slaughter, Peterson, & Mackintosh, 2007; Wellman & Liu, 2004). The first version contained primarily verbal presentation of the tasks. Based on performance of early participants, the second battery was modified to consist of nonverbal presentation of similar tasks in addition to more basic tests of desire and intention (i.e., recognizing individuals may have desires different than their own, such as enjoying

a food that is disliked by the participant) and appearance versus reality (i.e., recognizing that something may appear different than it is, such as a sponge that looks like a rock; Flavell, Flavell, & Green, 1983; Lewis & Mitchell, 1994; Matthews, Dissanayake, & Pratt, 2003; Repacholi & Gopnik, 1997; Slaughter et al., 2007). To produce a single composite ToM score for all participants that was comparable across batteries, factor analysis scores across batteries were derived, which were standardized to have a mean of 10 and an *SD* of 1 (see Losh et al., 2012b, for further detail).

Analysis Plan

Individuals were excluded from all analyses if they had fewer than five total intelligible responses across all requests ($n = 4$; including one male with FXS-ASD, one male with ASD-O, and two females with DS).

Group Comparisons of Communication Repair Skills

In order to characterize overall group differences on repair strategy type, a series of one-way analyses of covariance (ANCOVAs), followed by planned comparisons using Bonferroni correction to account for multiple comparisons, were conducted for males and females separately for each response type: nonresponse, verbal strategies (inappropriate, repetition, revision, addition, background, metacomment), and supplemental strategies (gestures, suprasegmentals), controlling for nonverbal mental age, expressive vocabulary age, receptive vocabulary age, and MLU, with diagnostic group as the independent variable. ANCOVAs, with sex as the independent variable, controlling for identical variables, were also used to examine sex differences within the FXS, DS, and TD groups.

Group Comparisons Across the Clarification Request Series

Individuals were only included in repeated-measures analyses if they had at least five intelligible responses at each request for clarification, which led to the further exclusion of seven males with ASD-O, 10 males and three females with FXS-ASD, one male and five females with FXS-O, six males and seven females with DS, and four males and two females with TD from repeated analyses only. To determine changes in response type across the series of prompts for clarification, three (prompt order)-by-five (diagnostic group) repeated-measures ANCOVAs were utilized with each of the repair categories (nonresponsive, six verbal, two supplemental) as the dependent variables for males and females separately. Planned comparisons using Bonferroni corrections were conducted to determine between-groups differences. Within groups, sex differences in changes in response type were examined through a three (prompt order)-by-two (sex) repeated-measures ANCOVA, controlling for nonverbal mental age, expressive vocabulary age, receptive vocabulary age, and MLU.

Correlates of Communication Repair Skills

Bivariate Pearson correlations were conducted within each group to determine the relationship between each

Table 2. Example of an interaction between the examiner (E) and child (C).

Sample	Utterance	Code
Original description	C Boy's getting a haircut.	
Prompt 1	E Huh?	
Repair 1	C Octopus is cutting his hair.	Addition
Prompt 2	E What?	
Repair 2	C Octopus is cutting his hair.	Repetition
Prompt 3	E I don't understand.	
Repair 3	C There's an octopus cutting his hair.	Revision
Understanding	E Oh, an octopus is cutting a boy's hair?	

of the repair strategy types and mental age, expressive and receptive vocabulary age equivalence, MLU, and ToM.

Results

Group Comparisons of Communication Repair Skills

Separate univariate ANCOVAs revealed main effects for diagnostic group for the males in nonresponsiveness, $F(4, 131) = 3.86, p = .01$, resistance to the task, $F(4, 131) = 4.1, p = .004$, and suprasegmentals, $F(4, 131) = 4.4, p = .002$ (Figure 1). Follow-up pairwise comparisons revealed that differences in nonresponsiveness were driven by males with ASD-O failing to respond to requests for clarification at a higher rate than males with FXS-ASD and FXS-O, $ps < .05$, although they did not differ from the TD or DS groups, $ps > .11$. Males with FXS-ASD showed more resistance to the task than males with DS, $p < .01$, and ASD-O, $p < .01$, although they did not differ from the TD or FXS-O groups, $ps > .12$. Males with DS used significantly more suprasegmentals than males with ASD-O, $p < .01$, and FXS-ASD, $p < .01$, although they did not differ from males with TD when correcting for multiple comparisons, $ps > .09$. No group differences were observed for females, $ps > .28$ (Figure 2).

For sex comparisons, separate univariate ANCOVAs for each diagnostic group revealed a main effect of sex in the TD group in use of inappropriate strategies, $F(1, 35) = 7.7, p = .01$, a main effect of sex in the FXS-ASD group in the

use of gestures, $F(1, 51) = 4.8, p = .03$, and a marginal effect of sex in resistance to the task, $F(1, 53) = 3.8, p = .06$. In particular, males with TD were more likely to use inappropriate responses (resisting the task, asking the examiner to look, making off-topic statements) than females with TD. Additionally, males with FXS-ASD gestured less and showed resistance at a higher rate than females. No additional sex differences emerged, $ps > .10$.

Group Comparisons Across the Clarification Request Series

Mean proportions of response type across each prompt for clarification are displayed in Figures 3–5. Results of the three (prompt order)-by-five (diagnostic group) repeated-measures ANCOVAs examining patterns of response across the stacked series of prompts for clarification revealed a significant main effect of prompt for nonresponsiveness, $F(2, 131) = 3.46, p = .03$, and use of repetition, $F(2, 140) = 3.2, p = .047$, in males (Figure 3). Across groups, males increased in nonresponsiveness from Prompts 1 to 3 and Prompts 2 to 3, $ps < .05$, and decreased in use of repetition from Prompts 1 to 3 and 2 to 3, $ps < .05$. There was no significant main effect for group or significant interaction of prompt by diagnosis for males for any other response type, $ps > .33$. For females, there were no significant main effects for group or prompt, $ps > .21$. There was a significant prompt by diagnosis interaction in the use of repetition for females, $F(6, 104) = 2.36, p < .05$ (Figure 4). Whereas females with

Figure 1. Overall mean proportion of strategy use across groups: males. Adjusted means and standard errors are depicted. Analyses controlled for Leiter–Revised mental age, Expressive Vocabulary Test age equivalence, Peabody Picture Vocabulary Test age equivalence, and mean length of utterance. Proportion of nonresponse trials (by total number of examiner prompts for clarification); all other proportion variables exclude nonresponse trials. ^Inappropriate composed of Look, Off-Topic, and Resistance. * $p < .05$, nonresponse: ASD-O > FXS-ASD, FXS-O; resistance: FXS-ASD > DS, ASD-O; suprasegmentals: DS > ASD-O, FXS-ASD. Autism spectrum disorder (ASD-O), fragile X syndrome with comorbid ASD (FXS-ASD), FXS without ASD (FXS-O), Down syndrome (DS), and typical development (TD).

Figure 2. Overall mean proportion of strategy use across groups: females. Adjusted means and standard errors are depicted. Analyses controlled for Leiter–Revised mental age, Expressive Vocabulary Test age equivalence, Peabody Picture Vocabulary Test age equivalence, and mean length of utterance. Proportion of nonresponse trials (by total number of examiner prompts for clarification); all other proportion variables exclude nonresponse trials. ^Inappropriate comprised of Look, Off-Topic, and Resistance. Autism spectrum disorder (ASD-O), fragile X syndrome with comorbid ASD (FXS-ASD), FXS without ASD (FXS-O), Down syndrome (DS), and typical development (TD).

Figure 3. Mean proportion of strategy use by prompt, across groups: males. Adjusted means and standard errors are depicted. Analyses controlled for Leiter–Revised mental age, Expressive Vocabulary Test age equivalence, Peabody Picture Vocabulary Test age equivalence, and mean length of utterance. Proportion of nonresponse trials (by total number of examiner prompts for clarification); all other proportion variables exclude nonresponse trials. ^Inappropriate comprised of Look, Off-Topic, and Resistance. * $p < .05$, indicating an overall main effect of prompt for nonresponse and repetition, but no significant main effect of group or significant group by prompt interaction. Autism spectrum disorder (ASD-O), fragile X syndrome with comorbid ASD (FXS-ASD), FXS without ASD (FXS-O), Down syndrome (DS), and typical development (TD).

Figure 4. Mean proportion of strategy use by prompt, across groups: females. Adjusted means and standard errors are depicted. Analyses controlled for Leiter–Revised mental age, Expressive Vocabulary Test age equivalence, Peabody Picture Vocabulary Test age equivalence, and mean length of utterance. Proportion of nonresponse trials (by total number of examiner prompts for clarification); all other proportion variables exclude nonresponse trials. ^Inappropriate comprised of Look, Off-Topic, and Resistance. * $p < .05$, indicating a significant group by prompt interaction for repetition but no overall main effects of prompt or group. Autism spectrum disorder (ASD-O), fragile X syndrome with comorbid ASD (FXS-ASD), FXS without ASD (FXS-O), Down syndrome (DS), and typical development (TD).

FXS-O and DS decreased their use of repetition strategies, females with TD and FXS-ASD remained relatively stable.

Within groups, results of the three (prompt order)-by-two (sex) repeated-measures ANCOVAs revealed a significant prompt by sex interaction in the TD group for

repetition, $F(2, 58) = 4.86$, $p < .05$, with males showing decreased repetition across the series of prompts, $p < .05$, and females remaining relatively stable. No other groups showed a main effect of prompt or sex or an interaction of prompt by sex, $ps > .21$.

Figure 5. Mean proportion of supplemental strategy use by prompt, across groups. Adjusted means and standard errors are depicted. Analyses controlled for Leiter–Revised mental age, Expressive Vocabulary Test age equivalence, Peabody Picture Vocabulary Test age equivalence, and mean length of utterance. Autism spectrum disorder (ASD-O), fragile X syndrome with comorbid ASD (FXS-ASD), FXS without ASD (FXS-O), Down syndrome (DS), and typical development (TD).

Table 3. Correlations between proportion of repair strategy use and cognition/language.

Strategy		ASD-O	FXS-ASD		FXS-O		DS		TD	
		Males	Males	Females	Males	Females	Males	Females	Males	Females
Nonresponse	Leiter	-.16	-.16	.19	.40	.47	-.26	-.15	-.42	.07
	PPVT	-.17	-.16	.32	.45	.37	-.27	-.18	-.42	.11
	EVT	-.11	-.03	.36	.46	.13	-.12	-.14	-.36	.16
	MLU	-.06	-.27	-.06	.25	.20	-.32	-.23	-.06	.20
Inappropriate	Leiter	-.15	.002	-.70	-.25	-.44	-.28	-.15	-.13	-.22
	PPVT	-.05	-.03	-.47	-.31	-.42	-.47	-.21	.02	-.17
	EVT	-.05	-.06	-.66	-.24	-.42	-.43	-.15	.02	-.15
	MLU	.17	.10	-.42	.34	-.54	-.08	-.12	.19	-.10
Repetition	Leiter	-.24	-.31	-.51	-.55	-.51	-.30	-.52	-.50	-.59
	PPVT	-.21	-.34	-.51	-.51	-.58	-.39	-.58	-.45	-.57
	EVT	-.39	-.33	-.70	-.61	-.49	-.51	-.58	-.47	-.67
	MLU	-.30	-.30	-.18	-.60	-.47	-.50	-.70	-.39	-.57
Revision	Leiter	.35	.17	.37	-.14	.39	.11	.53	-.07	.02
	PPVT	.25	.48	.47	-.18	.43	.25	.63	-.24	.15
	EVT	.37	.37	.61	-.17	.28	.30	.65	-.25	.23
	MLU	.002	.25	.21	-.04	.19	.21	.70	-.001	.35
Addition	Leiter	.09	.29	.71	.76	.40	.35	.32	.47	.65
	PPVT	-.02	.14	.36	.72	.48	.44	.39	.55	.62
	EVT	.07	.18	.63	.76	.39	.49	.21	.56	.65
	MLU	.19	.001	.18	.19	.51	.26	.24	.14	.48
Metacomment	Leiter	.04	-.26	.22	.11	-.15	.09	-.07	.13	-.21
	PPVT	.01	-.25	.36	.15	-.04	.11	.02	.17	-.22
	EVT	.10	-.22	.40	.04	-.01	.14	.19	.02	-.28
	MLU	-.01	.12	.47	-.74	-.19	.07	.38	.30	-.22
Suprasegmental	Leiter	-.04	.36	.68	-.53	-.42	.05	.16	-.36	-.47
	PPVT	-.05	.13	.20	-.38	-.51	.29	.04	-.34	-.34
	EVT	-.10	.12	.43	-.51	-.38	-.04	.10	-.28	-.35
	MLU	.16	.09	.40	-.73	-.23	.05	-.21	-.13	-.38
Gesture	Leiter	.11	.13	-.02	.78	-.13	.28	.11	-.17	-.28
	PPVT	-.02	.15	-.46	.59	-.08	.27	.13	-.25	-.21
	EVT	.01	-.06	-.26	.62	.01	.14	-.10	-.19	-.28
	MLU	.10	-.06	-.39	.07	.02	-.13	.01	.31	-.18

Note. r values reported. Bold font indicates $p < .05$. All correlations include age equivalences as covariates. ASD-O = ASD only; FXS-ASD = fragile X syndrome with autism spectrum disorder; FXS-O = FXS only; DS = Down syndrome; TD = typical development; Leiter = Leiter-Revised; EVT = Expressive Vocabulary Test; PPVT = Peabody Picture Vocabulary Test; MLU = mean length of utterance.

Correlates of Communication Repair Skills

Measures of cognition and language were significantly correlated with the majority of communication repair strategies across diagnostic groups, though correlations were more limited in males with ASD-O (see Table 3). ToM also showed a number of correlations with repair skills in males with DS, FXS-O, and TD but not in the ASD-O or FXS-ASD groups. In males with DS, better ToM was associated with fewer off-topic responses, $r(1, 22) = -.45$, $p = .04$, and greater tendency to add information in responses, $r(1, 22) = .45$, $p = .04$. In males with FXS-O, better ToM ability was associated with decreased requests to look at the picture, $r(1, 14) = -.63$, $p = .02$. Finally, in males with TD, better ToM was related to a greater tendency to provide background information, $r(1, 20) = .59$, $p = .01$.

ToM was also related to repair strategies among females in the DS, FXS-O, and TD groups. Better ToM in females with DS was associated with fewer off-topic responses and more revisions, $r(1, 19) = -.53$, $p = .02$; $r(1, 19) = .51$, $p = .03$, respectively. In both females with

FXS-O and TD, better ToM was related to fewer inappropriate responses, $r(1, 24) = -.47$, $p = .02$; $r(1, 14) = -.60$, $p = .02$, respectively.

Discussion

This study examined the ability to repair communication breakdowns in children and adolescents with different genetically based neurodevelopmental disabilities impacting pragmatics—ASD-O, FXS-ASD, FXS-O, and DS—compared with children with TD, in an effort to further characterize communication repair skills, identify related cognitive and language mechanisms, and identify possible targets for intervention in each group. Communication repair abilities are an important aspect of pragmatic language necessary for successful communicative exchanges and a potential source of communication difficulties in neurodevelopmental disabilities affecting communication and cognition. Overall, all clinical groups showed relative proficiency with the task, showing several similarities to TD controls in repair strategies employed. However, some

important group differences were also observed, along with a few sex-specific patterns.

Overall, clinical groups showed minimal differences from TD controls, suggesting relative competence in this specific pragmatic skill, at least in the context of this structured task, and when accounting for cognitive and structural language (vocabulary and syntax) delays. Patterns across prompts also revealed similarities across groups, lending important insight as to how best to structure requests for clarification for these individuals. For example, males became increasingly nonresponsive as the series of prompts persisted, and a trend of increased inappropriate responses was observed across both males and females, with the exception of females with FXS-ASD. These findings suggest that, despite some basic competence, persistent breakdowns in communication may lead to greater rates of frustration for both clinical groups and younger TD children. Further, with the exception of females with TD and FXS-ASD, all groups also decreased their repetition of prior statements as the prompting continued. As suggested by Brinton et al. (1986), it is possible that high rates of repetition early on in the sequence are due to the speaker inferring that the examiner's question is due to their unintelligibility, a common experience for many individuals with neurodevelopmental disabilities. As requests for clarification proceed, however, it may become clearer to the speaker that the request indicated a lack of understanding as opposed to an inability to perceive the message. These studies provide insight into possible ways to tailor requests for clarification across groups, in that they suggest that repeated prompting is not necessarily the most fruitful strategy for clarifying misunderstandings, and more direct and specific questions may be required. Future studies are needed, however, to investigate effective strategies to improve communication repair skills in individuals with neurodevelopmental disabilities who use complex language, as the majority of studies have focused on prelinguistic children (e.g., Keen, Sigafos, & Woodyatt, 2001).

It is, perhaps, surprising that we did not find greater differences between ASD-O and TD groups, given that pragmatic language impairment is a core feature of ASD (Landa, 2000; Tager-Flusberg, Paul, & Lord, 2005; Young, Diehl, Morris, Hyman, & Bennetto, 2005). However, males with ASD-O performed comparably to controls in some prior studies (Baltaxe, 1977; Geller, 1998; Paul & Cohen, 1984; Volden, 2004). Similarly, prior studies of ASD-O document better pragmatic language performance in more structured tasks with less demand on interpersonal interaction (Losh & Capps, 2003; Losh & Gordon, 2014; Martin et al., 2017; Nadig, Lee, Singh, Bosshart, & Ozonoff, 2010). Indeed, prior work examining repair strategies within the context of less structured conversational interaction have revealed some difficulties in communication repair even in very high-functioning individuals with ASD (Volden, 2004). It is possible that the structured nature of the present task served to scaffold performance and mask impairments that may be evident in more naturalistic settings.

Although they did not differ significantly from TD controls, both males with FXS-ASD and those with ASD-O demonstrated further difficulties in communication repair relative to other clinical groups. Males with FXS-ASD demonstrated significantly higher rates of inappropriate responses than males with ASD-O and DS, consistent with prior work demonstrating increased pragmatic impairments, including noncontingent (off-topic) language and fewer signals of noncomprehension of a message, in individuals with FXS-ASD compared with FXS-O, DS, and TD groups (Losh et al., 2012a; Martin et al., 2017, 2013, 2012; Roberts, Weisenfeld, Hatton, Heath, & Kaufmann, 2007). The current findings extend this literature to suggest that communication repair represents a specific area of difficulty for males with FXS-ASD. Males with ASD-O demonstrated a different area of impairment, in that they showed greater rates of nonresponsiveness relative to males with FXS-ASD and FXS-O. These findings suggest that, when working with males with FXS-ASD and ASD-O in a clinical context, it may be important to devote additional attention to assessing communication repair across persistent breakdowns in a more structured and deliberate way, such as using the present task, to evaluate this area of difficulty observed in both groups.

Findings that ASD symptomatology did not influence repair strategies similarly in males with ASD-O and FXS-ASD stand in contrast to prior studies documenting areas of strong similarity in pragmatic impairments in ASD-O and FXS-ASD, such as the frequency and types of pragmatic language violations committed in conversation and displayed in a standardized measure of pragmatic judgment (Klusek et al., 2014a; Losh et al., 2012a). It should be noted that males with FXS-ASD in the present sample had a significantly lower nonverbal mental age when compared with males with ASD-O. Although it is possible that impaired repair skills in FXS-ASD resulted from more severe intellectual disability (for review, see Abbeduto, McDuffie, & Thurman, 2014), both nonverbal mental age and vocabulary and syntax (MLU) were covaried in analyses, suggesting that the impaired performance in the FXS-ASD group was not simply explained by lower cognitive or language functioning. Rather, that the nature of communication repair challenges varies across these groups illustrates the importance of examining discrete pragmatic abilities to understand the potentially variable manifestations of ASD symptoms in ASD-O and FXS. Future studies should examine further the repair skills in these populations across different contexts and with different types of conversational partners to further characterize similarities and differences in this specific pragmatic skill.

In DS, it appears that communication repair skills are a relative strength, as both males and females responded appropriately to requests for clarification and varied their strategies in response to different prompts. Moreover, males with DS evidenced a significantly higher rate of strategies involving suprasegmentals than males with ASD-O and FXS-ASD, which may provide an effective, complementary strategy for clarifying communication when misunderstandings

occur. Poor speech intelligibility is a commonly reported characteristic of individuals with DS (see Kent & Vorperian, 2013, for a review), and the use of suprasegmental cues may serve as a particularly adaptive technique to address more frequent misunderstandings among this group. This possibility is supported by the increase in suprasegmental strategies in males with DS from the first to the second prompt, consistent with the possibility that individuals with DS may have initially assumed that a breakdown was due to unintelligibility.

There were few sex differences across groups in the use of different types of repair strategies, with the notable exception that females with FXS-ASD and TD were less likely to exhibit resistance to the task than their male counterparts. Additionally, females with FXS-ASD were more likely to use gestures than males. These findings are not necessarily surprising, given that females with FXS (with and without ASD) are typically less severely affected than males, due to the presence of a second X chromosome (thus one normally functioning copy of *FMRI*; Hagerman et al., 1992). Of note, previous studies of sex differences in communication repair skills in FXS have yielded mixed results; whereas two studies indicated better signaling of noncomprehension of a confusing message in females with FXS compared with males (Abbeduto et al., 2008; Thurman et al., 2017), research from our group did not detect sex differences in individuals with FXS-O or FXS-ASD (Martin et al., 2017). Additionally, TD females tend to develop social communicative skills earlier than males (Blakemore, Berenbaum, & Liben, 2008). One important consideration for future work is that this study was limited by the lack of a female ASD-O group. Because sex differences in social communication skills have been observed in ASD-O (Mandy et al., 2012) and given the differences noted among females with FXS-ASD, it will be important to include direct comparisons of females with ASD-O and FXS-ASD to investigate possible sex-specific profiles of communication repair skills in ASD and FXS (with and without ASD).

Finally, in addition to examining group and sex differences in communication repair ability, we also examined potential underpinning skills of this ability across groups. We hypothesized that impairments in ToM would relate to less sophisticated repair strategies across groups, as successful repair of communication breakdowns requires the speaker to recognize what information the listener may not possess. Whereas ToM abilities correlated with a range of repair strategies in males and females with FXS-O, DS, and TD, they were not associated with repair skills in either of the ASD groups. This finding stands in contrast to repeated reports of associations between ToM and pragmatic language in ASD-O (Capps et al., 1998; Capps, Losh, & Thurber, 2000; Losh & Capps, 2003, 2006; Loveland & Tunali, 1993; Surian, 1996; Tager-Flusberg, 2000; Tager-Flusberg & Sullivan, 1995) and FXS-ASD (Losh et al., 2012b) and may suggest that, for the ASD groups, deficiencies in the specific pragmatic skill of communication repair are tied more strongly to other factors, such as working memory, flexibility and set-shifting, or other

executive function skills (e.g., storing in working memory and recalling prior responses, showing flexibility by modifying prior responses). Considering additional skills that may relate to repair abilities in individuals with ASD (with and without FXS-ASD) will be an important focus for future work to help inform the complex causes of different communication difficulties in idiopathic and syndromic ASD.

In conclusion, children and adolescents with ASD-O, FXS-ASD, FXS-O, DS, and TD showed many strengths in communication repair ability but also some important differences, particularly among males with ASD-O and FXS-ASD. Group differences in repair skills may help to inform clinical practice by highlighting relative strengths and weaknesses displayed by different populations. In order to further refine and extend clinical applications of these findings, it will be important for future work to examine repair skills together with additional pragmatic skills to establish comprehensive pragmatic profiles that may be associated with particular diagnoses. Additionally, understanding of correlated abilities can help to inform knowledge of the origins of communication repair difficulties and guide intervention by focusing efforts on (potentially different) underlying contributing skills for each clinical group. Finally, future work would also profit from examining potential developmental changes in communication repair skills as related language and cognitive abilities progress and social and communicative challenges change with age. It will also be important to study communication repair skills across a variety of more practical and real-world settings (e.g., conversational, vocational) and with a variety of conversational partners (e.g., parents, peers) that may more sensitively capture challenges in this specific pragmatic skill across groups.

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