HHS Public Access

Author manuscript

Clin Linguist Phon. Author manuscript; available in PMC 2020 January 01.

Published in final edited form as:

Clin Linguist Phon. 2019 ; 33(5): 420–436. doi:10.1080/02699206.2018.1536727.

The Use of Demonstratives and Personal Pronouns in Fragile X Syndrome and Autism Spectrum Disorder

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Abstract

Demonstratives (e.g. here, that, these) and personal pronouns are early developing components of language, which are often impaired in young children with autism spectrum disorder (ASD). Additionally, demonstrative and personal pronoun use are linked to joint attention and language ability early in life for individuals with ASD. Fragile X syndrome (FXS) is a genetic disorder with a significant amount of overlap in its behavioural phenotype with ASD. The present study examined demonstrative and personal pronoun production during a conversation sample in adolescent boys with ASD and adolescent boys with FXS with a co-diagnosis of ASD (FXS +ASD). Findings indicated that grammatical complexity was related to both qualitative and quantitative aspects of demonstrative and personal pronoun production in boys with ASD, while grammatical complexity was related to the total number of demonstratives and personal pronouns produced in the boys with FXS+ASD. ASD severity was not related to demonstrative or personal pronoun production in ASD, although it was negatively correlated with the total number of personal pronouns produced by the boys with FXS. Additionally, groups did not differ significantly in production of personal pronouns, but they did differ significantly in multiple aspects of demonstrative use. Findings suggest that these groups produce similar rates of personal pronouns in the school-age years, while production of demonstratives differentiates these groups. This study contributes to the knowledge of the language phenotypes of idiopathic ASD and FXS +ASD, and provides implications for intervention targets for school-age children with these disorders.

Keywords

autism spectrum disorder; fragile X syndrome; demonstratives; personal pronouns; language

Introduction

Social referencing and using common ground is a challenge for individuals with autism spectrum disorder (ASD). Social referencing can be observed conversationally through

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Declaration of Interest

This research was supported by NIDCD grants R03 DC011616 and T32 DC05359, NICHD grant P30 HD03352, and start-up funds from the University of Wisconsin-Madison awarded to Audra Sterling. The authors have no additional declarations of interest to report.

demonstrative and personal pronoun use. Demonstratives and personal pronouns are early developing components of language. They use environmental referents to facilitate joint attention between the speaker and the listener (Diessel, 2006). Examples of demonstratives include: *this, that, these, those, here,* and *there*. Similarly, personal pronoun use requires recognition of a communication partner's perspective in order to produce correct referents (Charney, 1981). Joint attention can be initiated visually, through gaze, or linguistically, through words that label referents, such as demonstratives and personal pronouns (Diessel, 2006; Loveland & Landry, 1986).

School-age children with ASD commonly demonstrate deficits in demonstrative and personal pronoun production, in addition to deficits in joint attention and grammatical language production (Baltaxe & D'Angiola 1996; Hobson, García-Pérez, & Lee, 2010; Landry & Loveland, 1989). Children with fragile X syndrome (FXS), who often receive a co-diagnosis of ASD (FXS+ASD), demonstrate deficits in joint attention and grammar (Abbeduto, Brady, & Kover, 2007; Lewis et al., 2006; Losh, Martin, Klusek, Hogan-Brown, & Sideris, 2012; Sterling, Rice, & Warren, 2012), but no research to date has examined demonstrative use, and few studies have examined pronoun use in FXS (McDuffie, Thurman, Hagerman, & Abbeduto, 2015). Pragmatic language deficits persist beyond the early years for individuals with ASD and FXS+ASD (Losh et al., 2012; Roberts et al., 2007); however, the extent of demonstrative and personal pronoun impairment remains unknown. Characterizing these aspects of language in ASD and FXS+ASD can elucidate pragmatic and linguistic differences between these populations, as well as indicate targets for improving joint attention.

ASD and FXS

ASD is characterized by deficits in social communication along with restricted and repetitive interests and behaviours (American Psychiatric Association, 2013). ASD occurs in one in 59 children and is diagnosed behaviourally (American Psychiatric Association, 2013; Baio et al., 2018). FXS is the most common inherited cause of intellectual disability, and results from an expansion of the CGG trinucleotide repeat on the *FMR1* gene on the X chromosome (Verkerk et al., 1991). Given the X-linked nature of the disorder, FXS differentially impacts males and females. Thus, the current study only included males to control for sex differences. Comparing individuals with FXS and idiopathic ASD is valuable for multiple reasons. Between 27–75% of males with FXS receive a co-diagnosis of ASD, depending on the measures used (Clifford et al., 2007; Klusek, Martin, & Losh, 2014b). In addition, comparing FXS+ASD to idiopathic ASD will contribute to the understanding of the behavioural phenotype of FXS+ASD in relation to idiopathic ASD.

The language phenotypes of FXS and idiopathic ASD are variable, but boys in both groups often exhibit deficits in multiple domains. Boys with FXS demonstrate grammatical deficits, measured through mean length of utterance (MLU), compared to typically developing boys when controlling for non-verbal mental age (Finestack, Sterling, & Abbeduto, 2013; Price et al., 2008). Kover and Abbeduto (2010) found that boys with FXS+ASD and FXS-only matched on non-verbal mental age produced similar MLUs during conversation samples (Kover & Abbeduto, 2010). Boys with FXS also have lexical deficits (Abbeduto &

Hagerman, 1997; Finestack, Richmond, & Abbeduto, 2009). Alternatively, the language profile of idiopathic ASD is more variable. Some children with ASD have delays in grammar and vocabulary, but these deficits are not observed across all children with ASD (Tager-Flusberg et al., 2009). In addition, language development in ASD is often asynchronous; increases in MLU do not necessarily result in improved pragmatic language abilities (Tager-Flusberg & Anderson, 1991). Thus, it is unclear how MLU is related to demonstrative and pronoun use in individuals with ASD and FXS+ASD.

Demonstratives and personal pronouns in ASD

Typically developing children use demonstratives early in life; demonstratives are deictic terms that refer to the location of a common referent (Diessel, 2006). These words have a locative element to them, such that they facilitate or imply the need for joint attention (e.g., 'What's that' or 'Here it is'). In producing and comprehending demonstratives, one must take the perspective of the communication partner to create common ground. If a demonstrative refers to something tangible, the communication partners must engage in joint attention to understand the common referent. If the demonstrative refers to something intangible, such as a previously discussed topic, the communication partner must recognize that the speaker is referencing the formerly mentioned topic. Thus, demonstratives play a crucial role in language and social development (Diessel, 2006). Moreover, correct demonstrative use is necessary for successful daily interactions.

Findings vary regarding demonstrative use in children with ASD. Multiple studies of children with ASD ages four to 12 report no group differences in the number of demonstratives produced among children with ASD, children with developmental language delay (DLD), and typically developing children matched on mental age and MLU (Landry & Loveland, 1988; Loveland & Landry, 1986). Other studies suggest that individuals with ASD have impairments in demonstrative use compared to peers without ASD, specifically finding that young children with ASD produced fewer demonstratives in adult-directed or spontaneous interactions when compared to language- and age-matched children with Down syndrome (DS), language-matched typically developing children, and language- and cognitive-matched children with DLD (Baltaxe & D'Angiola, 1996; Landry & Loveland, 1989; Tager-Flusberg et al., 1990). In comparison to language- and age-matched children and adolescents with intellectual disability, individuals with ASD made more errors with deictic 'here/there' demonstratives in an experimental object-oriented task with an examiner, although participants in both groups incorrectly said 'that' or 'there' (Hobson et al., 2010). Furthermore, children with ASD ages five to 14 years have difficulty comprehending demonstratives (Hobson et al., 2010), which is likely related to difficulties with both language comprehension and joint attention. These findings suggest that demonstrative use may be impaired in idiopathic ASD beyond early childhood, yet it is unclear whether these deficits are related to ASD severity or overall language ability.

Like demonstratives, personal pronouns are an early developing component of language. Personal pronouns are complex; they require linguistic, pragmatic, and cognitive skills for correct use (Baltaxe & D'Angiola, 1996; Lee et al., 1994). Personal pronoun errors and reversal are commonly observed in individuals with ASD and are argued to be associated

with social communication deficits (Arnold, Bennetto, & Diehl, 2009; Charney, 1981; Lee et al., 1994; Naigles et al., 2016). Studies have found that children with ASD produce fewer personal pronouns overall, and when pronouns are used, they are more prone to errors compared to peers with similar language abilities (Baltaxe & D'Angiola, 1996). In terms of specific pronouns, children and adolescents with ASD use fewer instances of 'you' when referring to an examiner, and more use of their own name when referencing themselves (Lee et al., 1994). These errors directly impact communication success.

Prior research has also examined personal pronoun use in children with ASD throughout development. Naigles et al. (2016) examined personal pronoun reversal longitudinally in young children with ASD (mean age=31.64 months) and typical development (mean age=20.5 months), who were similar on vocabulary and receptive language scores. They coded joint attention from mother-child play sessions, and found that both joint attention and early language ability were related to the amount of pronoun reversal produced by children with ASD. They also suggested that children with ASD who had more advanced language development compared to social development demonstrated an increase in personal pronoun reversals (Naigles et al., 2016). Arnold et al. (2009) found an age effect on pronoun production in ASD. Specifically, younger children with ASD (9.8–12.9 years) produced fewer pronouns during a narrative re-tell compared to older children with ASD (13.1–17.8 years) and typically developing peers matched on age, IQ, and receptive language ability (Arnold et al., 2009). Thus, it may be the case that individuals with ASD no longer produce many pronoun errors in adolescence. However, the context, such as a narrative versus a conversation, may impact pronoun use. The current study will use a reciprocal conversation sample which will provide a unique perspective on pronoun use in adolescence during a semi-naturalistic context.

Demonstratives and personal pronouns in FXS

Prior research indicates that boys with ASD exhibit deficits in their demonstrative and personal pronouns use; however, our knowledge is limited for boys with FXS. McDuffie and colleagues (2015) used a parent interview, the Autism Diagnostic Interview-Revised (ADI-R; Rutter, Le Couteur, & Lord, 2003), to examine behavioural differences between 4–10 year-old boys with FXS and idiopathic ASD. Based on parent report, boys with FXS+ASD produced less pronoun reversal than boys with ASD. However, when the groups were matched on ASD severity, the two groups were not significantly different on pronoun reversal, indicating the importance of ASD severity (McDuffie et al., 2015). These findings suggest that there is not a unique genetic contribution of FXS on pronoun reversal, and pronoun reversal may be a feature associated with ASD severity.

Studies of lexical, grammatical, and pragmatic skills in boys with FXS indicate that ASD severity is linked to language skills (Martin et al., 2013; McDuffie, Kover, Abbeduto, Lewis, & Brown, 2012). Since boys with idiopathic ASD and FXS+ASD are phenotypically similar in their behaviour and language profiles, it is likely that both groups have similar difficulty using demonstratives and personal pronouns. Perhaps these aspects of language are closely associated with language ability or ASD severity, as suggested in previous research (Baltaxe & D'Angiola, 1996; Martin et al., 2013; McDuffie et al., 2012; McDuffie et al., 2015;

Naigles et al., 2016). As ASD severity is associated with language skills, we controlled for this by matching on ASD severity. This allowed for examination of the relationship between grammar and demonstrative and personal pronoun use in FXS+ASD and idiopathic ASD, as well as examination of potential differences and overlap between phenotypes.

Research questions

Demonstratives and personal pronouns are important in developing language and referencing common ground during a conversation, yet are notably impaired in children with ASD. It is likely that demonstratives and personal pronouns are impaired in FXS as well, given the high co-morbidity between FXS and ASD, but research has not yet explored demonstratives in FXS, and findings on personal pronouns are limited for boys with FXS+ASD. Given the critical role that demonstratives and personal pronouns play in language development, understanding these impairments in FXS+ASD and idiopathic ASD can help create intervention targets during the school-age years. The present study addressed the following questions:

1. Are demonstrative and personal pronoun use related to grammatical complexity (MLU), ASD severity, or nonverbal IQ for boys with FXS+ASD or boys with idiopathic ASD?
2. Are there quantitative or qualitative differences in demonstrative and personal pronoun use between boys with FXS+ASD and boys with idiopathic ASD matched on ASD severity?

For question one, we hypothesized that both ASD severity and grammatical complexity would be related to demonstrative and pronoun use in boys with idiopathic ASD based on the relationships identified between language ability and pronoun use in the study by Naigles et al. (2016). We hypothesized that grammatical complexity, ASD severity, and IQ scores, would also be related to demonstrative use and personal pronoun use in boys with FXS+ASD given the overlap often seen between boys with idiopathic ASD and FXS+ASD. For question two, we hypothesized a lack of differences in quantitative and qualitative demonstrative use between groups given the phenotypic overlap between these populations.

Methods

Participants

Boys with FXS+ASD and idiopathic ASD were selected from a larger study examining language abilities in these populations (Friedman, Sterling, & Barton-Hulsey, 2018; Haebig & Sterling, 2017; Sterling, 2018). Forty-four boys participated in the current study (FXS +ASD: n=24, idiopathic ASD: n=20). Participants were Standard American English speakers. All boys with FXS met criteria for ASD using both the Autism Diagnostic Observation Schedule (ADOS; Lord et al., 2012; Lord, Rutter, DiLavore, & Risi, 1999), and the expanded research cut-off for ASD outlined in Risi et al., 2006 for the Autism Diagnostic Interview – Revised (ADI-R; Rutter et al., 2003). Groups were matched on ASD severity as measured by the ADOS, $t(42)=-0.43$, $p = .671$, $d = 0.13$, variance ratio = 1.01, and Childhood Autism Rating Scale – Second Edition (CARS-2; Schopler, Van Bourgondien, Wellman, & Love, 2010), $t(42)=-0.01$, $p = .989$, $d = 0.01$, variance ratio =

1.17. Groups were also similar on chronological age, $t(42)=-1.51$, $p = .139$, $d = 0.46$, variance ratio = 1.03. Group matching criteria followed suggestions by Kover and Atwood (2013). Group means, standard deviations and group differences are presented in table 1.

To determine whether differences in demonstrative and pronoun use were associated with ASD or the genetic contribution of FXS, we matched on ASD severity, measured by the ADOS and CARS. Moreover, given the similarities in phenotypes between ASD and FXS, we matched on ASD severity to better understand the unique contribution of FXS on pronoun and demonstrative use.

Procedures

The institutional review board approved the study protocol. A legal parent or guardian provided written consent, and the participant provided oral assent. The visits were completed in one day at the Waisman Center at the University of Wisconsin-Madison. Participants completed ASD, nonverbal IQ, and language assessments. Participants were given breaks throughout the session as needed.

Measures

ASD assessments.—All boys completed either the first or second edition of the ADOS (Lord et al., 2012, Lord, Rutter, DiLavore, & Risi, 1999). The ADOS is a semi-structured assessment measuring ASD symptoms and severity. An examiner who was research-reliable or training to be research-reliable (with a research-reliable examiner present for live scoring) administered the appropriate ADOS module based on the child's language level; thus, all participants received a module 1, 2 or 3. We allowed module to vary given that our two groups were matched on ASD severity. Severity was calculated using the severity scoring algorithm (Gotham, Pickles, & Lord, 2009). One boy with ASD and two boys with FXS received a module 1, four boys with ASD and 14 boys with FXS+ASD received a module 2, and 15 boys with ASD and nine boys with FXS+ASD received a module 3. After the visit, the primary examiner completed the CARS, which is an observational measure with scores ranging from 15 to 60, where higher scores indicate more ASD symptoms (Schopler et al., 2010). The CARS was based on observations from the entire visit.

Nonverbal IQ.—The Leiter International Performance Scale – Revised (Roid & Miller, 1997) was used to measure nonverbal cognitive ability. Boys completed four subscales: Figure Ground, Form Completion, Sequential Order, and Repeated Patterns, which were all necessary for computing a brief IQ score (mean of 100, SD of 15).

Language.—All boys completed a semi-structured conversation language sample. The samples were 10 minutes long, and were completed by a trained examiner who followed a list of conversation topics and employed language elicitation techniques, such as asking open-ended questions. Parents gave the examiners a list of topics of interest for their sons, and the examiner asked about these topics at the beginning and end of the conversation. For the remainder of the conversation, the examiner followed a list of topics, which included questions such as, 'Tell me about your favourite part of school' and 'Tell me about your pets'. The language samples followed the same procedures as outlined in Berry-Kravis et al.

(2013). Conversation language samples are commonly used in language research as well as clinical practice, and provide a valid context for examining language ability in children with FXS (Berry-Kravis et al., 2013). Language samples were transcribed using the Systematic Analysis of Language Transcript conventions (SALT; Miller, Andriacchi, & Nockerts, 2011). We used complete and intelligible utterances to calculate morpheme-level mean length of utterance (MLU), which is a measure of grammatical complexity. Four out of the 20 files for ASD (20% of sample) and five out of the 24 files for FXS+ASD (21% of sample) were randomly selected and transcribed by a second transcriber for reliability. Reliability was calculated line-by-line. Agreement for SALT variables were: utterance segmentation=83.22%, unintelligible utterances=94.53%, number of morphemes=84.03%, number of words=85.18%, and word identification=88.10%.

Demonstrative coding

Transcribed conversation samples were coded for demonstrative use by two trained coders. Demonstrative coding was adapted from a published coding system (Baltaxe & D'Angiola, 1996). First, coders identified all demonstratives: *this, these, that, those, here, there.* Next, coders determined if the referent was concrete or abstract. Concrete referents were defined as demonstratives that referred to something tangible, such as a specific object or activity. Abstract demonstratives referred to an intangible idea or characteristic. Lastly, coders watched videos of the conversation language samples to determine if the demonstrative was used appropriately. Reliability was calculated for 20% of the files using Cohen's kappas. Cohen's kappa for the total number of demonstratives was 0.94. Demonstratives coded as concrete and abstract were 0.96 and 0.50 respectively. Cohen's kappas for appropriate or inappropriate use were 0.94 and 0.19 respectively. Values between 0.81 and 1.0 indicate near perfect agreement and values between 0.61 and 0.80 indicate substantial agreement (Hallgren, 2012). Demonstratives coded as abstract or inappropriate were both less frequent, which likely contributed to the lower kappa values (i.e. 0.50 for abstract indicating moderate agreement and 0.19 for inappropriate indicating slight agreement). All instances of inappropriate use and abstract demonstratives were then consensus coded for all participants due to low agreement.

Personal pronoun coding

Following demonstrative coding, the SALT transcripts were coded for personal pronouns. The authors created a coding scheme for identifying personal pronouns. First, coders identified any production of a personal pronoun and coded whether it was a first (i.e. *I, me, we, us)*, second (i.e. *you*), or third person (i.e. *them, they*) pronoun. The pronoun was then identified as correctly used or used in substitution for a different pronoun (e.g. 'you' instead of 'I'). If substituted, the coders identified which pronoun should have been produced. The coders also identified if pronouns were omitted. Reliability was calculated for 20% of the files using Cohen's kappas. Several variables demonstrated almost perfect agreement: total personal pronouns=0.97, proportion of correct personal pronouns=0.97, proportion of errors=0.86, first person pronouns=0.99, second person pronouns=0.99, third personal pronouns=0.96. Cohen's kappa for the number of omitted pronouns was 0.72, indicating substantial agreement (Hallgren, 2012).

Data analysis plan

Question 1.—The first research question used Pearson correlations to examine whether demonstrative and pronoun use were related to grammatical complexity (MLU), ASD severity, or IQ in boys with idiopathic ASD and FXS+ASD.

Question 2.—The second research question examined group differences in demonstrative and pronoun use. We considered including MLU, ASD severity or IQ as covariates; these variables were not correlated with one another, aside from a significant correlation between MLU and ASD severity in the boys with FXS+ASD (r=−0.43, p=0.038). Given that MLU was correlated with demonstrative and personal pronoun codes, we selected this as a covariate. Additionally, there were significant differences in MLU between groups, and given our focus on expressive language, it was important to control for grammatical complexity in the language samples. We considered using nonverbal IQ as a covariate; however, it was not correlated with any of the dependent variables measured in the study. Additionally, Dennis et al. (2009) outlined several important methodological and theoretical concerns with using nonverbal IQ as a covariate, and suggested that adjusting for nonverbal IQ in clinical populations such as FXS, where intellectual disability is a key part of the disorder, makes interpretation of results difficult.

To examine group differences, we first conducted a multivariate analysis of covariance (MANCOVA) to examine the effect of group on demonstrative use, controlling for MLU. Diagnostic group was a fixed factor. Dependent variables in our MANCOVA for analysis of demonstratives included proportion of concrete demonstratives, proportion of appropriate demonstratives, and the number of total demonstratives. Proportions of abstract demonstratives and inappropriate demonstratives were not used in the model, as these values were proportional to the values for concrete and appropriate demonstratives, respectively, and if added, would result in multicollinearity. Since this study looked at both the quantity (i.e. total number of demonstratives) and quality (i.e. concrete/abstract and appropriateness), frequency (i.e. count) was used to analyse the total number of demonstratives and proportions were used to analyse qualitative use. A main effect of group was followed up with planned pairwise comparisons, keeping the selected covariate consistent. A Holm-Bonferroni correction was used to allow for multiple comparisons (Holm, 1979).

We also conducted a MANCOVA to examine the effect of group on pronoun use, controlling for MLU. Dependent variables included total number of pronouns produced, the proportions of errors (including omissions and substitutions) for first, second, and third person pronouns, and the proportion of substitutions and proportion of omissions. Quantitative use was measured through total number of pronouns, while qualitative use was measured through proportions. Proportions were used to account for differences in the production of pronouns; moreover, participants were neither penalized nor advantaged when analysing error patterns based on their quantitative use of pronouns. Once again, a main effect of group was followed up with planned pairwise comparisons with our selected covariate based on research question one and Holm-Bonferroni corrections.

Assumptions for both MANCOVAs were checked. The total numbers of demonstratives and personal pronouns demonstrated a normal distribution and linearity. Constant variance was

assessed with spread-level plots; residuals showed similar variance. Proportions of concrete and appropriate demonstratives, and the proportion of personal pronoun errors, did not meet all assumptions. However, we restricted our alpha level to be 0.010 instead of 0.050. Additionally, using a Holm-Bonferroni correction adjusted for Type I error from multiple comparisons or violation of assumptions.

Results

Question 1: Relationships between demonstrative and personal pronoun use and MLU, ASD severity, and IQ

Separate Pearson correlation analyses were conducted for boys with FXS+ASD and boys with idiopathic ASD (see table 2). For the boys with FXS+ASD, MLU was significantly correlated with the total number of personal pronouns and demonstratives. There were significant relationships between ASD severity and total number of personal pronouns, as well as the proportion of omitted pronouns for the boys with FXS+ASD, such that an increase in ASD severity was related to a decrease in the number of personal pronouns produced and a higher proportion of omitted personal pronouns. For the boys with ASD, MLU was significantly correlated with the proportion of appropriate demonstratives, the total number of demonstratives, the proportion of first person errors, the proportion of third person errors, the proportion of omitted pronouns, and the total number of personal pronouns. There were no significant relationships between ASD severity and demonstrative or personal pronoun use for boys with idiopathic ASD. Additionally, IQ was not significantly correlated with any variables for both groups. Figure 1 depicts relationships between total number of demonstratives and total number of personal pronouns with MLU, and figure 2 depicts relationships between total number of demonstratives and total number of personal pronouns with ASD severity scores.

Question 2: Differences in demonstrative and personal pronoun use between groups

Means and standard deviations for demonstrative variables for each group are presented in table 3. Only one variable from each category (e.g., concrete from the concrete and abstract codes) was used in the MANCOVA to reduce multicollinearity in the model. MLU was included as a covariate. The main effect of group was significant, $F(3,38)=8.36$, Wilk's $\Lambda = 0.60$, $p = .000$, $\eta_p^2 = .40$. The effect of MLU was also significant, $F(3,38) = 9.83$, Wilk's $\Lambda = 0.56$, $p = .000$, $\eta_p^2 = .44$. Follow-up univariate comparisons were completed; p-values were adjusted using a Holm-Bonferroni correction, and adjusted p-values are presented (Holm, 1979). The proportion of concrete demonstratives, $F(1,40)=8.30$, $p = 0.012$ $\eta_p^2 = 0.17$, and proportion of appropriate demonstratives, $F(1,40)=9.67$, $p = 0.009$, $\eta_p^2 = 0.19$, differed significantly between groups. The groups produced a significantly different number of total demonstratives, $F(1,40)=5.15$, $p = 0.029$, $\eta_p^2 = 0.14$. Specifically, the group with FXS+ASD produced greater proportions of concrete and appropriate demonstrates, and more demonstratives overall than the group with idiopathic ASD. Pairwise comparisons for each dependent variable are presented in table 3. Figure 3 depicts group differences on these three dependent variables.

We also completed a MANCOVA for personal pronouns. Group was a fixed factor and MLU was a covariate. The main effect of group was not significant, $F(6,35)=1.64$, Wilk's $\Lambda = 0.78$, $p = 0.164$, $\eta_p^2 = 0.22$. The main effect of MLU was significant, $F(6,35)=10.83$, Wilk's $\Lambda = 0.35$, $p = 0.000$, $\eta_p^2 = 0.65$. Due to the lack of a significant group effect, follow-up comparisons were not completed. Means and standard deviations are presented in table 3.

Discussion

This study examined demonstrative and personal pronoun production in boys with FXS +ASD and boys with idiopathic ASD, matched on ASD severity. We also examined the relationship between grammatical ability, ASD severity, and IQ scores in the production of these skills. Our findings indicate that MLU is related to several components of demonstrative and personal pronoun production, and when controlling for differences in MLU, boys with FXS+ASD and idiopathic ASD are similar in their personal pronoun production, but not their use of demonstratives. These findings contribute to the literature in several important ways. This study provides insight on linguistic differences between boys with idiopathic ASD and boys with FXS+ASD unrelated to ASD status and provides useful information on factors that may contribute to the successful use of demonstratives and personal pronouns. Lastly, the findings can help inform intervention targets for these populations.

The correlational analyses indicated that grammatical complexity was related to several aspects of demonstrative and personal pronoun production in participants with idiopathic ASD. Higher MLUs were related to increased demonstrative and personal pronoun use, fewer errors in demonstratives, first and third person personal pronouns, and fewer omitted personal pronouns. Literature on early personal pronoun production in children with ASD suggested that language ability played an important role for this skill (Naigles et al., 2016) and our findings extend this relationship into the school-age years for boys with idiopathic ASD. However, MLU in boys with FXS+ASD was only related to the total number of demonstratives and personal pronouns. Errors in demonstratives or personal pronouns were not related to MLU in FXS+ASD as they were in idiopathic ASD, indicating that language ability may not play the same role in these referencing skills for both groups.

We also examined relationships between demonstrative and personal pronoun production and both IQ scores and ASD severity. There were no significant correlations between IQ scores and demonstrative or personal pronoun production in either group. Thus, intellectual ability may not be an important factor in the use of these aspects of language. In addition, we did not find any correlations between ASD severity and demonstrative and personal pronoun production for the boys with idiopathic ASD. However, ASD severity was positively correlated with the proportion of omitted personal pronouns and negatively correlated with the total number of personal pronouns for the boys with FXS+ASD. Based on previous work, the relationship between ASD severity and language is mixed, depending on the language domain (e.g., pragmatics, grammar, vocabulary), and if the task tests language comprehension or production (De Marchena & Eigsti, 2016; Klusek, Martin, & Losh, 2014a; Martin et al., 2013; McDuffie et al., 2012).

The present study only analysed expressive grammatical complexity through MLU, and not other aspects of language production or comprehension. However, grammatical complexity appears to be associated with demonstrative and personal pronoun production in boys ASD more so than in boys with FXS+ASD. In boys with FXS+ASD, demonstrative use and personal pronoun production may be differentially impacted by ASD severity and grammatical language ability. In the boys with FXS+ASD, demonstrative use was related to MLU but not ASD severity, yet pronoun use was related to both MLU and ASD severity. Previous research has indicated that ASD severity and language ability may be related in FXS (for a review, see Abbeduto, McDuffie, & Thurman, 2014), and the results from this study mirror those findings specifically for pronoun use. It may be important to consider both language ability and ASD severity when evaluating pronoun use in this group.

We compared boys with FXS+ASD and idiopathic ASD in demonstrative and personal pronoun use during a semi-structured conversation sample. When controlling for MLU, boys with ASD produced a smaller proportion of appropriate demonstratives, and fewer demonstratives overall. No group differences were hypothesized given the overlap in phenotypes between groups; however, the presence of differences may indicate that demonstrative production difficulty is a deficit unique to idiopathic ASD. No previous published work investigated demonstrative use in FXS+ASD, yet preliminary findings show that overall use and appropriate use of demonstratives may be better than what one would expect compared to individuals with idiopathic ASD. In addition, these findings contribute to our knowledge of impairments in demonstrative production in ASD relative to typically developing children as well as peers with DS (Baltaxe & D'Angiola, 1996; Hobson et al., 2010; Landry & Loveland, 1989; Tager-Flusberg et al., 1990), by extending these findings to an ASD symptom severity-matched group of boys with FXS+ASD. Specifically, the finding that boys with idiopathic ASD produced a larger proportion of inappropriate demonstratives adds to the study by Hobson et al. (2010), which included individuals of similar ages and found that the group with ASD produced more errors on some types of demonstratives compared to children with intellectual disability. While the use of demonstratives develops early for typically developing individuals (Diessel, 2006), boys with idiopathic ASD in particular demonstrate incorrect demonstrative use into later childhood and adolescence.

Despite group differences in demonstrative use, boys with FXS+ASD and boys with idiopathic ASD were similar in personal pronoun production. Our findings complement prior work, including reports of similar rates of pronoun reversal between ASD severity-matched boys with FXS+ASD and idiopathic ASD (McDuffie et al., 2015). The present study examined pronoun production during a conversation, while McDuffie and colleagues measured pronoun reversal using the ADI-R (McDuffie et al., 2012; Rutter et al., 2003). Notably, the boys in the present study produced few personal pronoun errors. This finding is consistent with Arnold et al. (2009), who found an age effect on pronoun use, such that younger children with ASD produced fewer pronouns than older children with ASD. They hypothesized that the strengthening of theory of mind through development may be responsible for this change (Arnold et al., 2009). Specifically, correct use of personal pronouns indicates that the speaker is aware of the listener's knowledge, such that the listener will understand the use of a pronoun instead of a more specific reference to a person or group. This awareness of knowledge may be connected to theory of mind; as theory of

mind develops, children produce more personal pronouns (Arnold et al., 2009), and they produce less errors, as indicated by the current findings. Although neither the present study nor Arnold et al., (2009) included a theory of mind task, it is a potential mechanism that underlies the complementary findings.

Limitations and future directions

This study has several limitations. The sample sizes were small for both groups. FXS is a relatively rare neurodevelopmental disorder, and the sample was in-line with published studies (McDuffie et al., 2012; Sterling et al., 2012). To account for this, we were cautious in our choice of analyses, and reported effect sizes to augment interpretation of our findings, but larger sample sizes would allow for more sophisticated analyses. There was no group of boys with FXS-only. Future work should include this group to elucidate the impact of ASD and the genetic contribution of FXS on these two components of language. Furthermore, this study excluded girls. Girls with ASD and FXS are important, understudied clinical groups. We only measured expressive use of demonstratives and personal pronouns through a conversation language sample; future work should include standardized or experimental tasks targeting demonstrative and personal pronoun production as well as comprehension. Although a conversation sample is an ecologically valid tool when examining language, the boys in our study did not produce many errors during this interaction. Manipulative control provided by experimental tasks, in conjunction with a conversation sample, could potentially provide a more robust examination of demonstrative and personal pronoun production. Additionally, longitudinal studies examining theory of mind and personal pronoun and demonstrative production simultaneously would aid in understanding the development of social referencing skills.

Conclusion

This study contributes to our understanding of language phenotypes in idiopathic ASD and FXS+ASD, and suggests that although both groups produce few errors in personal pronouns in the school-age years, demonstrative production differentiates these groups. Additionally, grammatical language ability is related to some aspects of demonstrative and pronoun production in boys with idiopathic ASD, while grammatical language does not play a significant role for boys with FXS+ASD. ASD severity in boys with FXS+ASD was related to the number of personal pronouns produced, as well as the proportion of omitted personal pronouns. Better understanding of demonstrative and personal pronoun strengths and challenges in these populations and in this age range can help inform intervention targets throughout the school-age years, as demonstratives and personal pronouns are critical for language development, social referencing, and communication success.

Acknowledgments

We thank the families that participated in this research. We also thank Susen Schroeder for her help with personal pronoun coding development, Kellie Willis and Shelby Gunderson for their assistance with coding, as well as the rest of the Research in Developmental Disabilities Language Lab for assistance with recruitment and data collection.

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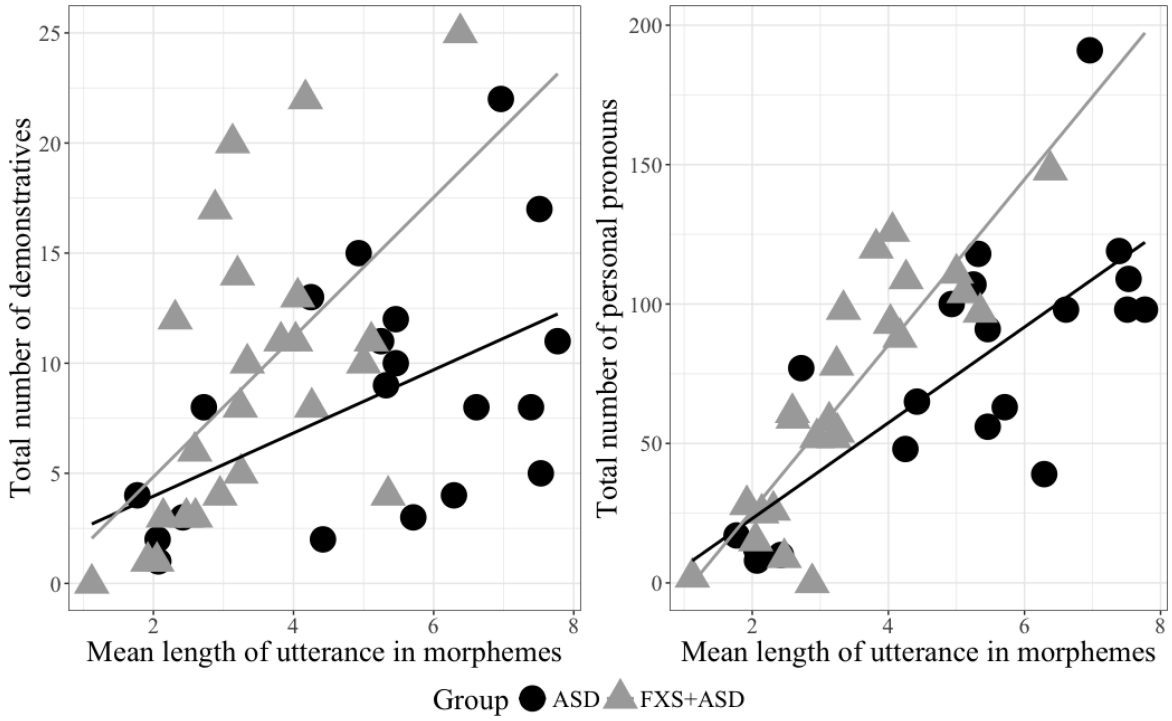

Figure 1.
Relationships between mean length of utterance (MLU) and outcome variables, total numbers of demonstratives and personal pronouns.

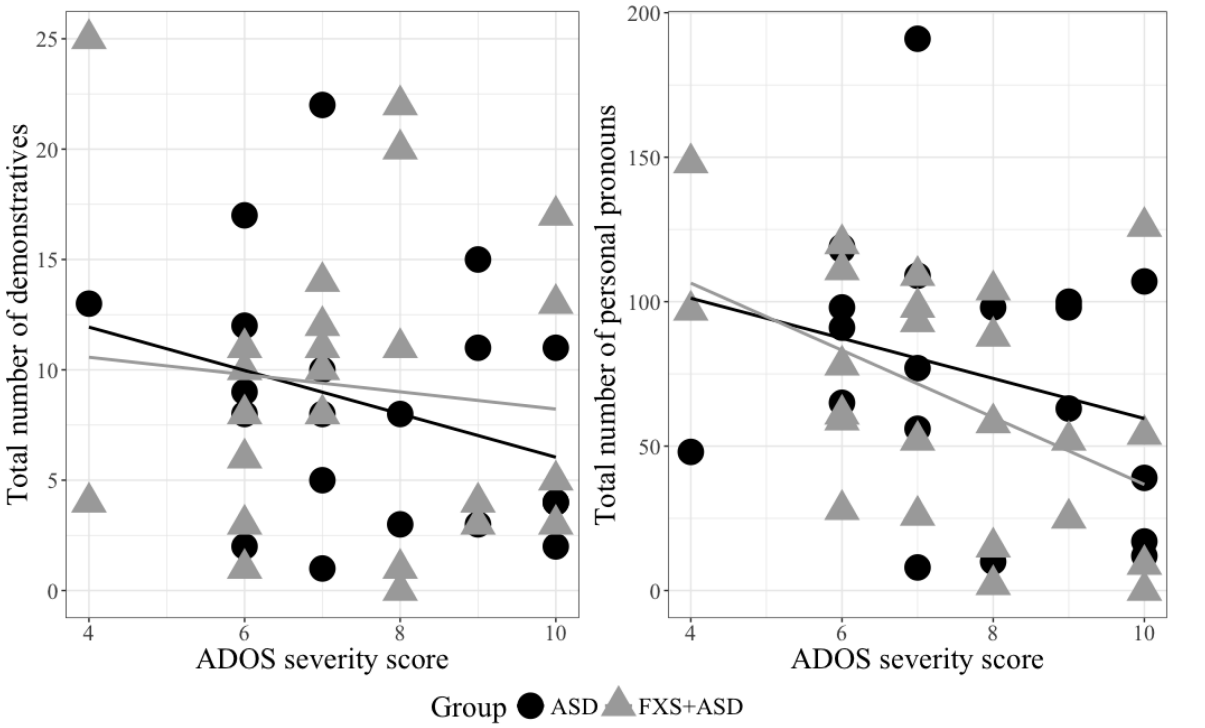

Figure 2.
Relationships between ADOS severity score and outcome variables, total number of demonstratives and total number of personal pronouns.

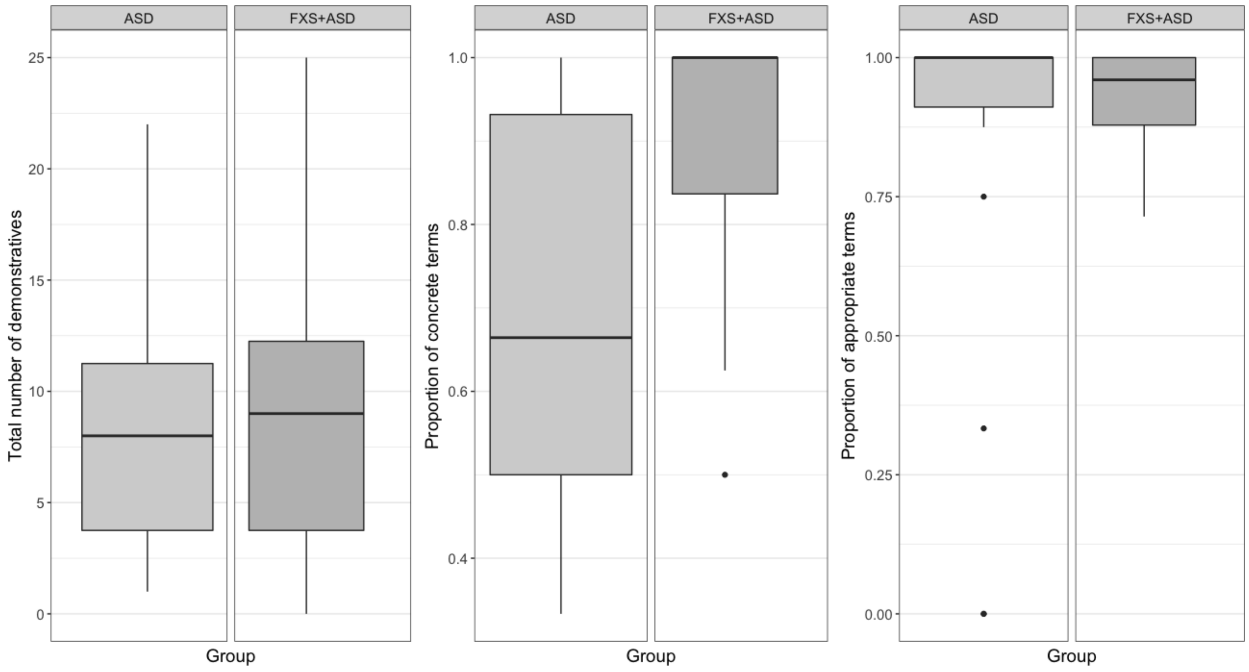

Figure 3.

Boxplots depicting group differences for total number of demonstratives, proportion of appropriate demonstratives, and proportion of concrete demonstratives

Table 1.

Descriptive data for each group and group comparison information

	ASD (*n* = 20) mean (SD)	FXS+ASD (*n* = 24) mean (SD)	*t* (df)	*p*	*Cohen's d*
Chronological age	13.61 (1.78)	12.79 (1.81)	−1.51 (42)	.139	0.46
MLU*	5.09 (1.99)	3.39 (1.24)	−3.47 (42)	.001	1.02
ADOS	7.60 (1.73)	7.38 (1.74)	−0.43 (42)	.671	0.13
CARS	30.38 (4.63)	30.35 (5.01)	−0.01 (42)	.989	0.01
IQ*	71.75 (21.24)	44.75 (6.92)	−4.88 (42)	.000	1.71

Note. MLU = mean length of utterance in morphemes; ADOS = Autism Diagnostic Observation Schedule severity score; CARS = Childhood Autism Rating Scale; *p* = exact *p* value.

* indicates a significant difference between groups.

Table 2.

Pearson correlation coefficients for mean length of utterance and ASD severity with personal pronoun and demonstrative codes. Correlations are presented for both boys with idiopathic ASD and boys with FXS+ASD.

	ASD			FXS+ASD		
	MLU	**ASD severity**	**IQ**	**MLU**	**ASD severity**	**IQ**
Proportion concrete demonstratives	−0.16	−0.11	0.44	0.00	0.07	−0.02
Proportion appropriate demonstratives	0.65**	−0.21	−0.01	0.08	−0.16	−0.17
Total demonstratives	0.51*	−0.30	−0.09	0.58**	−0.10	0.28
Proportion first person errors	−0.52*	0.28	0.12	−0.29	0.27	0.08
Proportion second person errors	−0.26	−0.14	0.11	−0.06	−0.04	0.00
Proportion third person errors	−0.48*	−0.01	0.30	−0.17	0.31	−0.14
Proportion incorrect-omitted	−0.64**	0.21	0.25	−0.31	0.42*	−0.07
Proportion incorrect- substituted	−0.33	−0.10	0.01	0.07	−0.26	−0.05
Total personal pronouns	0.74**	−0.26	−0.20	0.87**	−0.48*	0.28

* $p < 0.050$

** $p < 0.010$

Table 3.

Demonstrative and personal pronoun data for each group and group comparison information, controlling for MLU

Variable	ASD mean (SD)	FXS+ASD mean (SD)	*F*	p [a]	η_p^2
Demonstratives					
Total demonstratives	8.40 (5.61)	9.65 (6.67)	$F(1,40) = 5.15$	0.029	0.11
Proportion concrete	0.70 (0.14)	0.91 (0.14)	$F(1,40) = 8.30$	0.012	0.17
Proportion appropriate	0.84 (0.33)	0.93 (0.09)	$F(1,40) = 9.67$	0.009	0.19
Personal pronouns					
Total personal pronouns	76.20 (46.19)	70.13 (40.93)			
Proportion of first person errors[b]	0.07 (0.11)	0.09 (0.12)			
Proportion of second person errors[b]	0.01 (0.02)	0.01 (0.01)			
Proportion of third person errors[b]	0.04 (0.06)	0.04 (0.05)			
Proportion of omitted personal pronouns[b]	0.06 (0.07)	0.06 (0.08)			
Proportion of substituted personal pronouns[b]	0.01 (0.03)	0.01 (0.02)			

[a] Holm-Bonferroni correction was used to account for multiple comparisons; adjusted p-values are presented (Holm, 1979).

[b] Proportions of first, second, and third person pronoun errors are calculated out of the total number of personal pronouns produced. The proportions of omitted and substituted personal pronouns are also calculated out of the the total number of personal pronouns produced; the proportion of correct personal pronouns was not included in the model due to potential issues with multicollinearity.

Note. One participant with FXS+ASD did not produce demonstratives during their conversation sample, thus impacting the degrees of freedom. Additionally, one participant with FXS+ASD did not produce personal pronouns during their conversation sample. Group was not a main effect of personal pronoun use; no post hoc group comparisons were completed.