

Research Article

Effect of Speaker Gaze on Word Learning in Fragile X Syndrome: A Comparison With Nonsyndromic Autism Spectrum Disorder

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Purpose: This study examined use of a speaker's direction of gaze during word learning by boys with fragile X syndrome (FXS), boys with nonsyndromic autism spectrum disorder (ASD), and typically developing (TD) boys.

Method: A fast-mapping task with follow-in and discrepant labeling conditions was administered. We expected that the use of speaker gaze would lead to participants selecting as the referent of the novel label the object to which they attended in follow-in trials and the object to which the examiner attended in the discrepant labeling trials. Participants were school-aged boys with FXS ($n = 18$) or ASD ($n = 18$) matched on age, intelligence quotient, and

nonverbal cognition and younger TD boys ($n = 18$) matched on nonverbal cognition.

Results: All groups performed above chance in both conditions, although the TD boys performed closest to the expected pattern. Boys with FXS performed better during follow-in than in discrepant label trials, whereas TD boys and boys with ASD did equally well in both trial types. The type of trial administered first influenced subsequent responding. Error patterns also distinguished the groups.

Conclusion: The ability to utilize a speaker's gaze during word learning is not as well developed in boys with FXS or nonsyndromic ASD as in TD boys of the same developmental level.

With development, young children come to understand and infer meaning from the intentional actions of those around them (Baldwin & Markman, 1989; Carpenter, Nagell, & Tomasello, 1998). Among the most important skills gained during this developmental period is the ability to utilize the referential behaviors of communicative partners to support language acquisition. These social cues decrease ambiguity and guide the learner's focus of attention to the speaker's intended referent (Baldwin, 1993). A particularly significant social cue is provided by the speaker's direction of eye gaze (Brooks & Meltzoff, 2005). The relationship between gaze following and word learning is well documented for typically developing (TD) children as well as for children diagnosed with autism spectrum disorders (ASD; Carpenter, Pennington, &

Rogers, 2002). For other populations of children with neurodevelopmental disorders, including those with fragile X syndrome (FXS), the development of gaze following and subsequent effects on language development are not well understood. The current study examined how young boys with FXS use cues available from a speaker's direction of eye gaze to guide the acquisition of new words in a social interactive context.

Investigation of the process of language learning in FXS can add important information to our theoretical understanding of the psychological mechanisms that support or hinder language learning in this neurodevelopmental disorder as well as guide intervention planning. Given the well-documented overlap in behavioral symptoms between FXS and nonsyndromic ASD (i.e., cases of ASD in which a genetic etiology has been ruled out), the current study utilized a comparison group of children with nonsyndromic ASD matched on chronological age, intelligence quotient (IQ), and nonverbal developmental level as well as a comparison group of TD children matched on developmental level. Such comparisons can provide insight into the nature of the learning processes that are uniquely associated with or shared between neurodevelopmental disorders relative to typical development.

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When Social Cues Guide Word Learning

By late toddlerhood, TD children use a variety of social cues (e.g., gaze shifts, head turns, and pointing gestures) to guide word learning in word-learning situations in which there is no overt pairing between label and object (Baldwin, 1991, 1993; Baldwin et al., 1996; Tomasello, Strosberg, & Akhtar, 1996). The speaker's direction of gaze toward an intended referent is a particularly critical cue that decreases ambiguity and enables an accurate label-object pairing. The construct of gaze following, also referred to as attention following (McDuffie, Yoder, & Stone, 2006) or responding to joint attention (Mundy et al., 2007), signals the emergence of a child's ability to be an active contributor to the word-learning process.

Use of Social Cues by Children With Nonsyndromic ASD

A number of studies have used fast-mapping paradigms to examine the ways in which children with nonsyndromic ASD use social cues to learn words when there is ambiguity about the referent of a novel label (Luyster & Lord, 2009; McDuffie et al., 2006; McGregor, Rost, Arenas, Farris-Trimble, & Stiles, 2013; Walton & Ingersoll, 2013). *Fast mapping* is the term for an associative learning process, based on temporal contiguity, in which two units of information (e.g., a label and an object) are linked in memory after one or very few exposures (Carey & Bartlett, 1978). Fast-mapping studies have provided evidence of difficulty in learning new words when the speaker's direction of gaze is the only available cue to referential intent (Akechi et al., 2011; Baron-Cohen, Baldwin, & Crowson, 1997). Baron-Cohen et al. (1997) adapted a paradigm previously used with TD toddlers (Baldwin, 1991) to determine the extent to which children with nonsyndromic ASD could use speaker direction of gaze to learn words in a discrepant labeling context (i.e., a context in which two potential object referents are available to the child at the moment the verbal label is provided and the object being labeled is not the focus of the child's attention). Relative to younger TD children, children with nonsyndromic ASD are less able to use direction of gaze and more likely to make mapping errors when objects outside of their attentional focus are labeled (Baron-Cohen et al., 1997). Other studies have shown that word learning under conditions of discrepant labeling can approach the level demonstrated by TD participants when the language abilities of individuals with nonsyndromic ASD are age appropriate or when additional cues and word repetitions are present (Bani Hani, Gonzalez-Barrero, & Nadig, 2013; de Marchena, Eigsti, Worek, Ono, & Snedeker, 2011).

Although individuals with nonsyndromic ASD may demonstrate learning under particular variations of discrepant labeling, use of a matching strategy in which groups of participants are comparable on language abilities may obscure potential relationships between the phenotypic characteristics of a given neurodevelopmental disorder and word-learning tasks performance. Thus, the present study

was designed to examine the ways in which children with FXS or nonsyndromic ASD, matched on nonverbal cognition and chronological age, learned a novel word during conditions of follow-in labeling (i.e., a context in which two potential object referents are available to the child at the moment the verbal label is provided when the object being labeled is in the child's focus of attention) and discrepant labeling.

FXS

FXS is an X-linked genetic disorder and the most common inherited cause of intellectual disability (Crawford, Acuna, & Sherman, 2001). FXS is caused by an expansion of a CGG trinucleotide repeat on the *fragile X mental retardation 1 (FMR1)* gene on the X chromosome, which results in a decrease in or absence of *FMR1* protein (FMRP), the protein typically produced by the *FMR1* gene (Loesch, Huggins, & Hagerman, 2004). FMRP is critical for synaptic plasticity and experience-dependent learning (Bassell & Warren, 2008). Although FXS can affect both sexes, it is more prevalent in males, who are, on average, more severely affected than females (Hagerman, 2008). For males, the behavioral phenotype of FXS is characterized by intellectual disability (Hessl et al., 2009), language delays (Abbeduto, Brady, & Kover, 2007), repetitive behaviors (Wolff et al., 2012), hyperarousal and inattention (Cornish & Wilding, 2010), and social anxiety (Hessl, Glaser, Dyer-Friedman, & Reiss, 2006). In addition, as many as 90% of males with FXS display behaviors typically associated with nonsyndromic ASD, such as gaze aversion, stereotyped behaviors, and repetitive language (Hessl et al., 2006). Last, up to 60% of males with FXS have symptoms that are frequent and severe enough to warrant a comorbid diagnosis of ASD (Rogers, Wehner, & Hagerman, 2001). However, the exact nature of ASD in FXS continues to generate controversy (Hall, Lightbody, Hirt, Rezvani, & Reiss, 2010; McDuffie, Thurman, Hagerman, & Abbeduto, 2014; Rogers et al., 2001; Thurman, McDuffie, Hagerman, & Abbeduto, 2014; Wolff et al., 2012). Although there is some indication of neurobiological overlap between nonsyndromic ASD and FXS (e.g., Fatemi & Folsom, 2011), there is not yet consensus that this overlap leads to the same behavioral symptoms (Hall et al., 2010).

Accounts of the social development of young children with FXS often describe eye-gaze avoidance (Cohen, Vietze, Sudhalter, Jenkins, & Brown, 1991; Hessl et al., 2006), suggesting that these children are likely to miss behavioral indicators of the attentional focus of other people. It has been argued that the underlying cause of social avoidance in children with FXS is social anxiety; that is, gaze-aversive behaviors in FXS may function to minimize the social anxiety caused by direct gaze sharing (Dalton, Holsen, Abbeduto, & Davidson, 2008; Hessl et al., 2006). Regardless of its developmental etiology, gaze aversion may decrease the likelihood that a child will respond appropriately to the social cues of communicative partners, thereby indirectly affecting word learning. In addition, other aspects of the behavioral phenotype of boys with FXS are likely to

interfere with the ability to effectively respond to the referential cues of communicative partners during early language learning (Cornish, Cole, Longhi, Karmiloff-Smith, & Scerif, 2012). In the current study, we examined one aspect of social communication that is of primary importance during early word learning: the ability to utilize the direction of gaze of a communicative partner as a cue to referential intent when disambiguating the meaning of a novel word.

Approaches to the Study of Language Development in Boys With FXS

Investigations into the language abilities of boys with FXS generally have used global measures of expressive or receptive language or have investigated how language abilities are concurrently or longitudinally related to the development of other skills (Abbeduto et al., 2003). This line of research has provided evidence that boys with FXS have receptive vocabulary skills that, although delayed relative to age, are commensurate with nonverbal cognitive level and that may even be a strength relative to expressive language ability (Abbeduto et al., 2003; McDuffie, Kover, Abbeduto, Lewis, & Brown, 2012). Moving beyond an understanding of language as a product of development and examining the process by which language is acquired by boys with FXS can further both clinical and theoretical knowledge of the FXS phenotype.

Word Learning in FXS

Using a task that included ostensive labeling of novel objects, McDuffie, Kover, Hagerman, and Abbeduto (2013) examined fast mapping in 4- to 10-year-olds with either FXS or nonsyndromic ASD who were matched on chronological age. In this paradigm, successfully establishing an associative link between the target object and a novel label did not require attention to the speaker's direction of gaze or understanding of the speaker's referential intent because only one object was presented at a time and only one object in each pair of stimulus objects was labeled. Given the way this fast-mapping task was structured, the findings of McDuffie et al. (2013) can be considered to provide a baseline assessment of associative word-learning ability. Boys with FXS and those with nonsyndromic ASD responded to the task at above-chance levels; however, participants with FXS significantly outperformed participants with nonsyndromic ASD despite having lower levels of nonverbal cognition. The current study was designed to extend the work of McDuffie et al. (2013) by testing whether boys with FXS demonstrate a relative advantage or weakness in the use of a single critical social cue—the direction of a speaker's gaze—to learn words in a nonostensive context. A comparison with performance by individuals with nonsyndromic ASD has the potential to add to our understanding of how symptoms of ASD relate to specific learning competencies.

Research Questions

Using an experimental fast-mapping paradigm, the current study was designed to investigate the ability of young boys with FXS to use a speaker's direction of gaze as a guide to word learning. We adapted the paradigm originally introduced by Baldwin (1991) and later adapted by Baron-Cohen et al. (1997) to examine the process of word learning under conditions of follow-in and discrepant labeling. A group of same-aged boys with nonsyndromic ASD matched on IQ and nonverbal developmental level served as a comparison group, as did a group of young TD boys matched on nonverbal developmental level. To generate hypotheses regarding causal mechanisms involved in word learning, we also examined relationships with several phenotypic characteristics that might affect word-learning performance. The following research questions were addressed:

1. Do children with FXS, children with nonsyndromic ASD, or TD children learn a novel word under conditions of follow-in labeling (i.e., when the examiner labels the object in the child's focus of attention)?
2. Do children with FXS, children with nonsyndromic ASD, or TD children learn a novel word under conditions of discrepant labeling (i.e., when the examiner labels the object not in the child's focus of attention)?
3. Within each diagnostic group, is there a relative difference in the ability to learn a novel word across follow-in and discrepant labeling conditions?
4. Within each diagnostic group, is task performance influenced by the type of trial (follow-in vs. discrepant labeling) administered first to each participant?
5. Do patterns of errors vary as a function of labeling condition, trial type, or diagnostic group?
6. How does word-learning performance relate to child phenotypic characteristics (i.e., autism symptoms, vocabulary level, cognitive level) in FXS, nonsyndromic ASD, or typical development?

Method

Participants

Participants in the current study were drawn from a larger project investigating early word learning in FXS and nonsyndromic ASD. The larger sample included 57 boys with FXS, 61 boys with nonsyndromic ASD, and 59 TD boys. Participants were recruited and tested at two university sites, one on the West Coast and one in the Midwest. TD participants were recruited locally at each site, whereas participants with nonsyndromic ASD and FXS were recruited nationally. A boy was included if speech was his primary means of communication, English was the primary language spoken at home, and he lived with his biological mother. For all participants, parents indicated that their sons would be able to respond to simple instructions, such as "Give me the toy," and had used at least 10 different

spoken words over the previous month. According to parent report, no participant had any uncorrected motor or sensory impairments that would preclude participation in the experimental tasks. Hearing was screened during the research visit to rule out hearing loss. Families of participants with FXS provided documentation of a molecular test to confirm the diagnosis of FXS, and participants with nonsyndromic ASD were enrolled in the study only if they had a pre-existing community diagnosis of ASD or pervasive developmental disorder—not otherwise specified as well as a negative molecular test for FXS. TD participants were not receiving any special education services at the time of study enrollment.

Participants with FXS or nonsyndromic ASD from the larger sample were included in the current study if they had a nonverbal IQ below 85, as measured by the Brief IQ subtests of the Leiter International Performance Scale–Revised (LIPS-R; Roid & Miller, 1997). In addition, participants with nonsyndromic ASD were required to receive (a) a diagnostic classification of autism on the Autism Diagnostic Interview–Revised (ADI-R; Lord, Rutter, & Le Couteur, 1994) and (b) an autism severity score of at least 4 (Gotham, Pickles, & Lord, 2009) on the Autism Diagnostic Observation Schedule (ADOS; Lord, Rutter, DiLavore, & Risi, 1999). TD participants were included if they scored below a cutoff of 12 on the Social Communication Questionnaire (SCQ; Rutter, Bailey, Berument, Lord, & Pickles, 2003). These criteria resulted in a subsample of 51 boys with FXS, 37 boys with nonsyndromic ASD, and 58 TD boys.

Remaining participants were considered for inclusion in the current study only if all six trials of the experimental word-learning task were valid as determined by task administration guidelines. A trial could be invalid due to child noncompliance, examiner error, or other interruption such as the need for a bathroom break. This criterion resulted in a subsample of 27 boys with FXS, 21 boys with nonsyndromic ASD, and 34 TD boys. Last, 18 participants with either nonsyndromic ASD or FXS were matched on chronological age ($p = .83$, $d = 0.07$), nonverbal IQ ($p = .84$, $d = 0.07$), and LIPS-R growth scores ($p = .61$, $d = 0.18$). Using LIPS-R growth scores, 18 TD participants were then matched on nonverbal developmental level to those with nonsyndromic ASD ($p = .62$, $d = 0.17$) and FXS ($p = .94$, $d = 0.03$), yielding a final sample size of 54 (18 boys with FXS, 18 boys with nonsyndromic ASD, and 18 TD boys). Growth scores were used in addition to IQ as an absolute measure of cognitive ability to match participants with FXS and nonsyndromic ASD to TD participants. Characteristics of participants are presented in Table 1.

Standardized Assessments

SCQ

The SCQ, a parent-report questionnaire based on the ADI-R, was developed to serve as a screening tool for autism symptoms. It was collected only for the TD participants, as described previously (Corsello et al., 2007).

ADOS

Participants with nonsyndromic ASD and FXS were administered the ADOS, a play-based, semistructured assessment that is seen as a gold-standard measure of autism symptomatology. In the present study, the ADOS was used to verify diagnosis for participants with nonsyndromic ASD as well as to create a continuous metric of autism symptom severity based on the algorithms introduced by Gotham et al. (2009). The ADOS was administered by research-reliable examiners.

LIPS-R

Brief IQ subtests of the LIPS-R, a nonverbally administered measure of nonverbal cognition, were administered. The Brief IQ subtests include Figure Ground, Form Completion, Sequential Order, and Repeated Patterns. Standard scores, which indicate a child's ability relative to same-aged peers, and growth scores, which indicate absolute ability levels, were used to provide measures of IQ and nonverbal developmental level, respectively.

Peabody Picture Vocabulary Test–Fourth Edition

The Peabody Picture Vocabulary Test–Fourth Edition (PPVT-4; Dunn & Dunn, 2007) is a measure of receptive vocabulary. Both standard scores and growth scores were recorded for this measure. Growth scores were used for analyses.

Expressive Vocabulary Test–Second Edition

The Expressive Vocabulary Test–Second Edition (Williams, 2007), which is conormed with the PPVT-4, is a measure of expressive vocabulary. Both standard scores and growth scores were recorded for this measure. Growth scores were used for analyses.

Word-Learning Task

Overview

The protocol for the larger study included four fast-mapping tasks, which were administered to the majority of participants over a 2-day visit. The word-learning task used in the current study was the second word-learning task administered to participants. As with the original version (Baldwin, 1991; Baron-Cohen et al., 1997), the current task was designed to determine if the participant was able to use direction of eye gaze as a nonverbal referential cue when mapping a novel label to a novel object. The task was introduced as the Mine and Yours game, with participants being told that it was a game in which both the child and examiner would be playing with objects. The task included three trials in each of two labeling conditions: follow-in labeling and discrepant labeling. Regardless of condition, each of the trials consisted of a teaching phase, during which the novel label was presented, and a test phase, during which comprehension and generalization were assessed. A different novel label was used for each of the six trials, and the novel objects and labels were randomly assigned to participants. In addition, the order of trials was counterbalanced and randomly assigned to participants such that half of the

Table 1. Demographic characteristics and word-learning task performance in boys with fragile X syndrome (FXS; $n = 18$), boys with nonsyndromic autism spectrum disorders (ASD; $n = 18$), and typically developing (TD; $n = 18$) boys.

Variable	FXS		Nonsyndromic ASD		TD	
	<i>M</i>	<i>SD</i>	<i>M</i>	<i>SD</i>	<i>M</i>	<i>SD</i>
Chronological age ^a	7.09	2.41	7.24	1.81	3.66	1.08
Nonverbal cognition (LIPS-R)						
IQ ^b	66.56	12.73	65.72	12.43	110.44	14.60
Growth score	454.78	12.47	456.89	12.16	455.06	9.89
Receptive vocabulary (PPVT-4)						
Standard score ^b	75.06	14.22	63.17	19.99	122.87	8.00
Growth score ^c	121.00	26.62	106.67	28.57	129.29	18.28
Expressive vocabulary (EVT-2)						
Standard score ^b	75.16	15.09	62.44	25.56	119.00	10.37
Growth score	129.11	21.24	116.83	30.07	132.94	13.80
Autism symptomatology (ADOS)						
Calibrated severity score ^d	5.17	2.09	8.28	1.41		
Word-learning task						
Follow-in labeling condition ^e	4.44	1.58	3.33	1.85	4.28	1.49
Discrepant labeling condition ^f	2.50	1.76	2.44	1.65	2.83	2.23

Note. Unless otherwise noted, no between-groups scores significantly differ. LIPS-R = Leiter International Performance Scale-Revised (Roid & Miller, 1997); IQ = intelligence quotient; PPVT-4 = Peabody Picture Vocabulary Test-Fourth Edition (Dunn & Dunn, 2007); EVT-2 = Expressive Vocabulary Test-Second Edition (Williams, 2007); ADOS = Autism Diagnostic Observation Schedule (Lord et al., 1999).

^aTD participants significantly younger than participants with FXS and nonsyndromic ASD. ^bScores for TD participants significantly greater than those for participants with FXS and nonsyndromic ASD. ^cScores for TD participants significantly greater than those for participants with nonsyndromic ASD. ^dScores for participants with nonsyndromic ASD significantly greater than those for participants with FXS. ^eCorrect response = child's object. ^fCorrect response = examiner's object.

participants in each group received a follow-in labeling trial first and half received a discrepant labeling trial first. Within the current task, no more than two trials of the same condition were presented sequentially to any participant.

Teaching the Novel Word

Each trial began with the examiner presenting two novel objects to the child. In both conditions, the examiner placed the examiner's object on the table first while saying, "I'll play with *this* one!" and then placed the child's object in front of the child while saying, "You can play with *this* one!" The rationale for providing the child's object second in both experimental conditions was twofold: to increase the likelihood that the child would notice the novel object that was placed in front of the examiner and to increase the likelihood that the child would be focused on his own object when the novel label was provided.

In the follow-in labeling condition, the examiner picked up his/her own object (i.e., the distractor object for this experimental condition) but directed his/her gaze to the child's object (i.e., the target object for this experimental condition) and provided the labeling phrase (e.g., "It's a *modi!* A *modi!* It's a *modi!*"). The examiner did not present the novel label until the child was visually focused on his own object. If the child was not focused on his own object, the examiner would start that trial from the beginning by presenting the two novel objects again.

The discrepant labeling condition followed the same procedure except that the examiner directed his/her gaze to the examiner's object (i.e., the target object for this

experimental condition) when providing the novel label. As in the follow-in labeling condition, the novel label was not presented until the examiner had confirmed that the child's gaze was directed toward the child's own object (i.e., the distractor object for this experimental condition). For each trial and for each condition, the examiner removed both objects from the table after completion of the labeling process and began the test phase.

Testing Comprehension and Generalization

Teaching of the novel label was followed immediately by a test phase that assessed learning of the novel label using a forced-choice paradigm. The test phase consisted of two types of learning probes: a comprehension probe and a generalization probe. Both probes consisted of the presentation of four stimulus objects, each placed in an individual clear plastic compartment arranged in a line and fixed in place on a 1-in. by 6-in. board. This fixed-compartment response array allowed the examiner to make a clear determination of the child's response to each comprehension or generalization probe. In the comprehension probe, the stimulus items in the response array were the two novel objects from the immediately preceding teaching phase and two familiar objects that had been identified by the parent as objects for which the child knew a label (e.g., a toy boat, a miniature shirt, a toy chair). In the generalization probe, two of the stimulus items were new exemplars of the novel objects used for that trial's comprehension probe (differing in color, size, or embellishment) and two were additional familiar items. Familiar objects were included in comprehension

and generalization probes to provide a metric for behavioral compliance during the task and to make certain that the child understood the task demands.

During both the comprehension and generalization probes, the examiner asked the participant for both the labeled target object and one of the familiar objects by extending his/her upturned palm and asking the child, "Where's the [novel/familiar label]? Give me the [novel/familiar label]!" The left-to-right order of the objects in the tray as well as the order in which the examiner asked for the target or familiar object was counterbalanced across conditions. For all trials, the examiner and child were seated at a small table diagonally across from one another. After each individual probe, the examiner placed the selected object back into the tray such that the child was always picking from four objects.

Materials

Novel objects for this task were primarily store-bought items that were unlikely to be familiar to children. The objects were bright in color and roughly 5 to 7 in. in size. Each novel object used in a comprehension probe also had a matched object that was altered in some way (e.g., color, size, embellishment), which was used in the generalization probe.

Novel Words

The novel words used in this task were consonant–vowel–consonant–vowel nonsense syllables that were screened to make sure they did not resemble any English words that would be familiar to participants. The words for each task in the testing protocol were rotated and assigned sequentially from a list of 68 novel words with the constraints that (a) the child would not be exposed to the same word more than once across all tasks from the larger study and (b) that no two words within a task began with the same phoneme or differed by only one phoneme.

Familiar Objects

Each child's parents chose familiar objects from a group of more than 200 common household items, toys, play foods, or animal figures on the basis of the child's familiarity with the objects. For this task, parents were asked to select 24 familiar objects that the parent was confident the child could select if the examiner asked the child to find that object. The selection of 24 familiar objects allowed for two familiar objects to be used in the comprehension and generalization probes for each of six trials without repeated use of a familiar object for a given participant.

Recording Responses

The examiner recorded all responses on a protocol sheet that was designed to allow rapid notation of the object selected. After the testing session was completed, all videos of the experimental task were watched by the examiner who administered the task as well as by a different trained examiner to ensure that scoring decisions were accurate and that trial administrations were valid. In order for a trial to be valid, the child had to be looking at his own object during the onset of the labeling statement, regardless of the condition.

Analysis Plan

The distribution of word-learning task scores violated the parametric assumptions of normality for all three participant groups; thus, nonparametric statistics were used in all analyses. Comparisons of performance between comprehension and generalization probes revealed no significant differences for any group; therefore, performance was collapsed across these test probe types for all analyses. In addressing the first and second research questions, we first compared the number correct for each participant group in each experimental condition with chance using Wilcoxon signed-ranks tests, thereby evaluating the extent to which object–label pairings were learned. Next, we compared the frequencies with which the child's own object and the examiner's object were chosen by each participant group in each condition using Wilcoxon signed-ranks tests. This analysis provided insight into whether the participants were able to disengage from their own object to map the examiner's object to the novel label. We then sought to identify diagnostic group differences in learning the object–label pairings in the two experimental conditions. We did this by comparing the number of correct object choices for each participant group in each of the two experimental conditions using Mann–Whitney *U* tests and the Serlin–Harwell aligned-rank procedure (Serlin & Harwell, 2004), with the latter controlling for group differences in autism symptom severity. In addressing the third research question, we used Wilcoxon signed-ranks tests to compare the two experimental conditions, separately for each participant group, regarding the number of correct object choices, the number of child object choices, and the number of examiner object choices. These analyses provided insight into the extent to which the accuracy of performance and the tendency to choose the child's own (or the examiner's) object were influenced by condition. In addressing the fourth research question, we used Mann–Whitney *U* tests to examine the extent to which the difference in the number of correct object choices in the two experimental conditions varied according to which condition was experienced first, thereby addressing the issue of context, or priming, effects. In addressing the fifth research question, chi-square analyses were used to evaluate whether the distribution of participant errors differed as a function of group (FXS, nonsyndromic ASD, or TD), labeling condition (follow-in vs. discrepant), or test probe type (comprehension vs. generalization). Last, the sixth research question was addressed by computing Spearman's rho correlations between word-learning task performance and nonverbal cognitive ability, receptive vocabulary, and autism symptoms for each participant group.

Results

Preliminary Analyses

The number of correct responses to familiar object probes was compared with chance using Wilcoxon signed-ranks tests. Participants in all groups selected the familiar objects at levels significantly greater than chance: FXS, $T = 171.00$, $z = 4.06$, $p < .001$, $r = .98$; nonsyndromic ASD,

$T = 171.00$, $z = 3.78$, $p < .001$, $r = .89$; TD, $T = 171.00$, $z = 3.87$, $p < .001$, $r = .91$; all large effect sizes. These results provide assurance that participants understood and successfully complied with the demands of the experimental task.

Experimental Task Performance

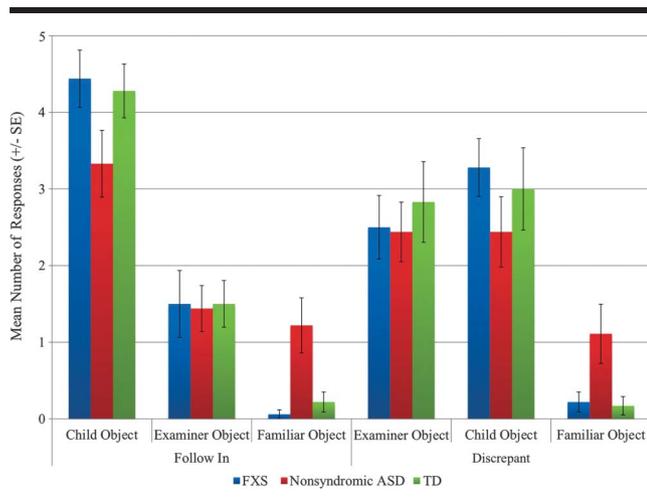
Follow-In Labeling Condition

The first research question addressed whether participants with FXS, participants with nonsyndromic ASD, or TD participants were able to learn new words during the follow-in labeling condition. In this condition, participants had six opportunities to choose the target object (i.e., the child's object) from the response array during test probes. Descriptive statistics for follow-in labeling trials are presented in Table 1, and task performance is summarized in Figure 1.

Comparisons relative to chance. Each group of participants performed at a level that significantly exceeded chance regarding the number of correct object choices: FXS, $T = 168.50$, $z = 3.65$, $p < .001$, $r = .86$; nonsyndromic ASD, $T = 157.50$, $z = 3.17$, $p = .002$, $r = .75$; TD, $T = 171.00$, $z = 3.74$, $p < .001$, $r = .88$; all large effect sizes.

Within-group comparisons. Participants in all three groups chose their own (target) object significantly more often than the examiner's (distractor) object: FXS, $T = 12.00$, $z = 2.77$, $p = .006$, $r = .65$; nonsyndromic ASD, $T = 22.00$,

Figure 1. Word-learning task performance by labeling condition and participant group. The child's object was the target object in the follow-in labeling condition, whereas the examiner's object was the target object in the discrepant labeling condition. There were no significant between-groups differences in performance within either condition. However, participants with fragile X syndrome (FXS) and typically developing (TD) participants chose the child's object more frequently during follow-in trials relative to discrepant labeling trials, whereas participants with nonsyndromic autism spectrum disorders (ASD) and TD participants selected the examiner's object more frequently during discrepant labeling trials relative to follow-in trials. The odds of selecting a familiar object when asked for a target object were significantly greater for participants with ASD relative to both other groups.



$z = 2.40$, $p = .02$, $r = .57$; TD, $T = 4.00$, $z = 3.07$, $p = .002$, $r = .72$.

Between-groups comparisons. In examining the number of correct object choices, we found that participants with FXS had marginally higher scores than participants with nonsyndromic ASD ($U = 221.00$, $z = 1.90$, $p = .06$, $r = .45$). However, this between-groups difference was not significant after controlling for severity of ASD symptomatology, $F(1, 35) = 0.89$, $p = .35$. TD participants did not significantly differ from participants with either nonsyndromic ASD ($U = 112.00$, $z = 1.61$, $p = .11$, $r = .38$) or FXS ($U = 177.00$, $z = 0.49$, $p = .63$, $r = .12$).

Discrepant Labeling Condition

The second research question addressed whether participants with FXS, participants with nonsyndromic ASD, or TD participants were able to learn new words during the discrepant labeling condition. As was the case for follow-in labeling, participants had six opportunities to choose the target object (i.e., the examiner's object) from the response array during test probes. Descriptive statistics for discrepant labeling trials are presented in Table 1, and task performance is summarized in Figure 1.

Comparisons relative to chance. Each group of participants performed at a level that significantly exceeded chance regarding the number of correct object choices: FXS, $T = 132.00$, $z = 2.06$, $p = .04$, $r = .49$; nonsyndromic ASD, $T = 134.00$, $z = 2.14$, $p = .03$, $r = .50$; TD, $T = 133.00$, $z = 2.08$, $p = .04$, $r = .49$; all large effect sizes.

Within-group comparisons. The frequency with which the examiner's (target) object was chosen did not differ from the frequency with which the child's own (distractor) object was chosen for any participant group: FXS, $T = 74.00$, $z = 0.81$, $p = .42$, $r = .19$; nonsyndromic ASD, $T = 59.00$, $z = 0.06$, $p = .95$, $r = .01$; TD, $T = 72.00$, $z = 0.21$, $p = .83$, $r = .05$.

Between-groups comparisons. In examining the number of correct object choices, we found no significant differences in performance between any of the participant groups: FXS versus TD, $U = 151.00$, $z = 0.35$, $p = .72$, $r = .17$; FXS versus nonsyndromic ASD, $U = 158.50$, $z = 0.11$, $p = .91$, $r = .03$; nonsyndromic ASD versus TD, $U = 146.50$, $z = 0.50$, $p = .62$, $r = .12$. In addition, the difference between participants with FXS and those with nonsyndromic ASD remained nonsignificant after controlling for severity of ASD symptomatology, $F(1, 35) = 1.75$, $p = .19$.

Follow-In Versus Discrepant Labeling Performance

The third research question examined whether performance within each group differed as a function of labeling condition. The number of correct object choices by participants with FXS was significantly greater during follow-in labeling than during discrepant labeling ($T = 14.00$, $z = 2.98$, $p = .003$, $r = .70$). In contrast, participants with nonsyndromic ASD ($T = 43.50$, $z = 1.28$, $p = .20$, $r = .30$) and TD participants ($T = 35.50$, $z = 1.69$, $p = .09$, $r = .40$) performed similarly in both conditions (see Figure 1).

Selection of the child's object. Participants with FXS ($T = 30.00, z = 2.00, p = .046, r = .47$) and TD participants ($T = 6.00, z = 2.79, p = .005, r = .66$) chose their own object more often during follow-in labeling than during discrepant labeling. This difference was not significant for participants with nonsyndromic ASD ($T = 16.50, z = 1.81, p = .07, r = .43$).

Selection of the examiner's object. TD participants ($T = 6.00, z = 2.79, p = .005, r = .66$) and participants with nonsyndromic ASD ($T = 8.00, z = 2.29, p = .02, r = .54$) selected the examiner's object significantly more often during discrepant labeling than during follow-in labeling. This difference was not significant for participants with FXS ($T = 49.00, z = 1.61, p = .11, r = .38$).

Effect of Initial Trial Type on Performance

To address the fourth research question, participants within each group were categorized according to the trial type (follow-in, discrepant) that they received first during administration of the experimental task. A difference score was computed using the following formula: number of correct follow-in labeling trials minus number of correct discrepant labeling trials. The effect of initial trial type was not significant for participants with FXS ($U = 23.50, z = -1.38, p = .17, r = .33$) or TD participants ($U = 31.00, z = -0.69, p = .49, r = .16$). However, a significant difference did emerge for participants with nonsyndromic ASD ($U = 14.50, z = -2.33, p = .02, r = .55$), for whom the advantage of follow-in relative to discrepant labeling was attenuated when a discrepant labeling trial was the initial trial administered during the experimental task (see Figure 2).

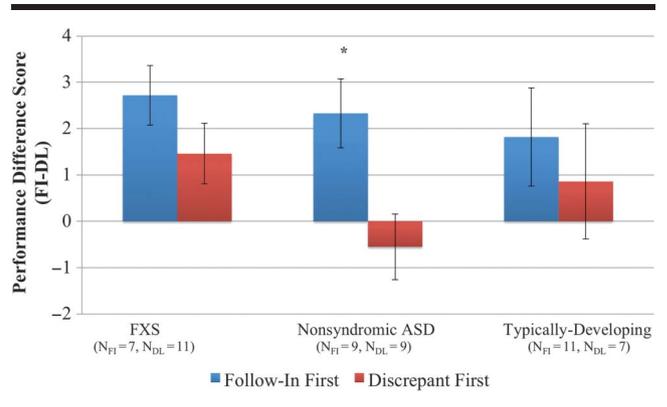
Error Analyses

The fifth research question addressed error patterns. An error could be made during test probes in one of two ways. A novel object error would occur if the child chose the examiner's object during follow-in labeling or the child's object during discrepant labeling. A familiar object error would occur if the child chose a familiar object instead of the novel target object.

Error type as a function of participant group. A significant difference was observed between participants with FXS and those with nonsyndromic ASD, $\chi^2[1] = 28.91, p < .001, \phi = .38$, odds ratio = 10.3, indicating that the odds of choosing a familiar object when asked for a target object were 10.3 times greater for participants with nonsyndromic ASD. A similar pattern of findings was observed when comparing participants with nonsyndromic ASD and TD participants, $\chi^2[1] = 23.26, p < .001, \phi = .34$, odds ratio = 6.94. Error patterns did not differ when participants with FXS were compared with TD participants, $\chi^2[1] = 0.43, p = .51, \phi = .05$.

Error type as a function of labeling condition. Within each group, patterns of errors did not differ based on whether the participant was responding to a follow-in or a discrepant labeling trial: FXS, $\chi^2[1] = 0.29, p = .59, \phi = .06$; nonsyndromic ASD, $\chi^2[1] = 2.49, p = .12, \phi = .15$; TD, $\chi^2[1] = 0.69, p = .41, \phi = .09$.

Figure 2. Word-learning performance as a function of initial trial type. Due to the counterbalancing of trial presentation order, participants received either a follow-in (FI) trial or a discrepant (DL) trial as their initial trial during task administration. A difference score was used to compare task performance between FI and DL trials within each group according to the type of trial that was administered first to each participant. Unlike the participants with fragile X syndrome (FXS) or typically developing participants, participants with nonsyndromic autism spectrum disorders (ASD) who received a DL trial as their initial trial type showed a significant decrement in performance for FI trials, as evidenced by the negative difference score obtained when subtracting the number of correct DL trials from the number of correct FI trials. * $p < .05$.



Error type as a function of test probe type. Within each group, patterns of errors did not differ based on whether the participant was responding to a comprehension or a generalization probe: FXS, $\chi^2[1] = 0.19, p = .66, \phi = .05$; nonsyndromic ASD, $\chi^2[1] = 0.54, p = .46, \phi = .07$; TD, $\chi^2[1] = 3.19, p = .07, \phi = .19$.

Correlates of Task Performance

The last research question examined whether word-learning performance was concurrently correlated to any of the child characteristics measured.

Follow-in labeling condition. For participants with FXS, the number of correct object choices in follow-in labeling was significantly correlated with both PPVT-4 growth scores ($r_s = .56, p = .02$) and LIPS-R growth scores ($r_s = .57, p = .01$). Significant correlations between the number of correct object choices and PPVT-4 growth scores ($r_s = .57, p = .01$) and LIPS-R growth scores ($r_s = .57, p = .01$) were also observed for participants with nonsyndromic ASD. These correlations did not reach significance for TD participants (see Table 2).

Discrepant labeling condition. The number of correct object choices in the discrepant labeling condition was not significantly correlated with any child characteristic for any participant group (see Table 2).

Discussion

The ability to respond to the attentional focus of a conversational partner by following his or her line of regard is critical for establishing joint reference within ambiguous

Table 2. Correlations between word-learning task performance and child characteristics in boys with fragile X syndrome (FXS), boys with nonsyndromic autism spectrum disorder (ASD), and typically developing (TD) boys.

Variable	1	2	3
FXS			
Follow-in labeling	.56*	.57*	-.13
Discrepant labeling	-.13	-.23	-.34
1. Receptive vocabulary	—	.86**	.05
2. Cognitive level	—	—	.09
3. ADOS severity	—	—	—
Nonsyndromic ASD			
Follow-in labeling	.57*	.57*	-.14
Discrepant labeling	.12	-.18	-.31
1. Receptive vocabulary	—	.54*	-.18
2. Cognitive level	—	—	-.08
3. ADOS severity	—	—	—
TD			
Follow-in labeling	.14	.06	-.37
Discrepant labeling	-.27	.03	.18
1. Receptive vocabulary	—	.55*	.31
2. Cognitive level	—	—	-.03
3. SCQ total	—	—	—

Note. ADOS = Autism Diagnostic Observation Schedule (Lord et al., 1999); SCQ = Social Communication Questionnaire (Rutter et al., 2003).

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

word-learning situations. In naturalistic settings, for example, adults frequently label objects and events that do not represent the child's current focus of attention (Harris, Jones, & Grant, 1983). In such a situation, the child must attend to the speaker's face, recognize the speaker's direction of gaze as relevant, follow the speaker's line of regard, and associate a spoken label with the novel object that is the speaker's intended referent (Baldwin, 1993). Although many variations of fast-mapping paradigms have been used to assess novel word learning in children with nonsyndromic ASD, only two published studies have examined the process of fast mapping in young boys affected by FXS (McDuffie et al., 2013; Thurman et al., 2014). McDuffie et al. (2013) demonstrated that boys with FXS outperformed boys with nonsyndromic ASD in a fast-mapping task requiring only associative learning despite having lower levels of nonverbal cognitive ability. Thurman et al. (2014) used a fast-mapping task in which the emotional reaction of the examiner (excitement vs. disappointment) indicated whether the search for a novel object—and therefore the accuracy of the label-object pairing—was successful or unsuccessful. Performance for all groups exceeded chance in both search conditions. In the successful search condition, participants with nonsyndromic ASD performed similarly to participants with FXS after controlling for severity of ASD. In the unsuccessful search condition, participants with FXS performed significantly worse than participants with nonsyndromic ASD after controlling for severity of ASD. The current study sought to extend the literature by (a) examining the ability of young boys with FXS to use a speaker's direction of gaze

as a cue for learning new words and (b) comparing patterns of word-learning performance between boys with FXS and boys with nonsyndromic ASD.

In the current study, above-chance levels of performance were observed for all groups in both conditions of the experimental fast-mapping task. A direct comparison of performance between participants with FXS and those with nonsyndromic ASD failed to reveal any significant between-groups differences after controlling for severity of ASD symptomatology. In addition, TD participants performed similarly to both participants with FXS and those with nonsyndromic ASD in both experimental conditions. Last, although participants in all groups chose the child's object significantly more often than the examiner's objects during follow-in trials, there was no difference in the category of object chosen for discrepant labeling trials (i.e., the examiner's object was not chosen significantly more often than the child's object). Despite these similarities across groups, the results of the current study also revealed subtle differences in word-learning performance within each group as a function of labeling condition.

TD participants demonstrated comparable levels of accuracy across the follow-in and discrepant labeling conditions. Furthermore, they were the only participant group to demonstrate the ability to adapt or shift their response pattern depending on the examiner's direction of gaze, as evidenced by selecting the child's object significantly more often during the follow-in condition and selecting the examiner's object significantly more often during the discrepant labeling condition. Thus, as expected, TD participants were highly competent in this task, confirming that the task operated as intended for typical language learners.

In contrast, the response accuracy of participants with FXS was significantly better in follow-in labeling trials than in discrepant labeling trials. Participants with FXS also chose the child's object significantly more often during follow-in trials than they did during discrepant trials. However, unlike TD participants, participants with FXS did not choose the examiner's object significantly more often during discrepant trials than during follow-in trials. Thus, the findings of the present study support the findings of McDuffie et al. (2013) that word learning in FXS is more successful when the participant is able to make a direct link, facilitated by temporal contiguity, between the label and the object. The findings of Thurman et al. (2014) additionally suggest that, compared with performance by individuals with nonsyndromic ASD, performance by individuals with FXS is negatively affected by the need to use more sophisticated inferencing skills to support word learning. More advanced reasoning skills are likely to be required by the unsuccessful search condition presented by Thurman et al. and the discrepant labeling condition used in the current study. The findings of the current study suggest that associative learning may be a relative strength for children with FXS of this cognitive and language level, whereas their ability to use, monitor, and interpret direction of eye gaze as a referential cue may not yet be fully developed. It also seems possible that challenges in the effective use of the speaker's eye gaze

for word learning by children with FXS may not be related to social indifference but rather to other behavioral difficulties observed in FXS, such as poor sequential processing and executive control (e.g., Kemper, Hagerman, & Altshul-Stark, 1988; Wilding, Cornish, & Munir, 2002). Future studies should continue to examine the association between phenotypic characteristics such as these and word learning in FXS.

Like the younger TD participants, participants with nonsyndromic ASD demonstrated comparable levels of accuracy across the two experimental conditions; that is, they did not demonstrate the relative advantage for follow-in trials observed in participants with FXS. In addition, although participants with nonsyndromic ASD were significantly more likely to select the examiner's object during discrepant labeling trials, they unexpectedly did not favor selection of their own object during follow-in labeling trials. Further insights into the word-learning performance of children with nonsyndromic ASD were provided by examination of error response patterns across both experimental conditions as well as by examination of the effect of the initial type of trial presented during the experimental task.

Participants with nonsyndromic ASD showed very high levels of correct performance on familiar-item probes, suggesting that they understood and complied with the performance demands of the experimental task. However, when they responded in error during comprehension or generalization probes requesting a novel target object, participants with nonsyndromic ASD demonstrated a significantly greater likelihood of choosing a familiar object relative to the other participant groups. That is, when the examiner used a novel label to request the target object during test probes, the expected error would be to select the novel distractor object, not a familiar object. One possibility is that children with nonsyndromic ASD in the current study were not as strongly constrained by mutual exclusivity (Markman & Wachtel, 1988; Mather & Plunkett, 2011) as were the children in the other two groups. This line of reasoning may be unlikely given the strong bias shown by children with nonsyndromic ASD to avoid mapping a novel label to an object that has already been labeled (de Marchena et al., 2011). However, it should be noted that participants with nonsyndromic ASD in the de Marchena et al. study had language skills in the average to above-average range, whereas the children with nonsyndromic ASD in the current study all had moderate to severe language and cognitive delays. Given the evidence that young children with nonsyndromic ASD have a stronger interest in familiar relative to novel objects (Adamson, Deckner, & Bakeman, 2010; Brucker & Yoder, 2007), one additional possibility is that children with nonsyndromic ASD in the present study may have had difficulty inhibiting their interest in objects with which they were already familiar.

The analysis examining the effect of initial trial type additionally suggests that participants with nonsyndromic ASD in the current study may have had difficulties with set shifting. In this analysis, participants with nonsyndromic ASD who received a follow-in labeling trial as the initial

trial type showed a significant difference in overall task performance relative to participants with nonsyndromic ASD who received a discrepant labeling trial first. That is, the difference score (i.e., number of correct follow-in labeling trials minus number of correct discrepant labeling trials) for participants who received a follow-in trial first was positive, on average, whereas the same difference score for participants with nonsyndromic ASD who received a discrepant trial first was negative (see Figure 2). It is therefore possible that participants with nonsyndromic ASD adopted the style of the first trial of the experimental task to guide their functional interpretation of how to play the game rather than actively utilizing the examiner's changing gaze during each trial as a source of information. This pattern of responding was not observed in either of the other two participant groups.

Associations between word-learning performance and cognitive ability and receptive language ability suggest some fruitful directions for investigating causal mechanisms. Word-learning performance for the follow-in labeling trials was significantly correlated with general cognitive and language ability for participants with FXS and nonsyndromic ASD. These significant correlations, in addition to the pattern of performance demonstrated by TD participants, support the utility of the fast-mapping task as a construct valid measure of word-learning ability for these groups of children (Cronbach & Meehl, 1955). Discrepant labeling performance tended to be lower relative to follow-in labeling performance for all groups of participants. This restricted variability may have contributed to the lack of significant associations observed for the discrepant labeling condition.

Due to the behavioral features shared between the FXS and nonsyndromic ASD phenotypes, researchers have been interested in determining whether similar neurocognitive mechanisms underlie these phenotypes. In the present study, comparable levels of accuracy were observed between boys with FXS and those with nonsyndromic ASD during both follow-in and discrepant labeling conditions. In addition, similar patterns were observed between the two groups in terms of the correlates of word-learning performance in either condition. There is, however, some evidence that for individuals with nonverbal IQs less than 85, boys with FXS earn significantly higher receptive vocabulary standard scores than do boys with nonsyndromic ASD. The results from the present study add to this emerging picture by suggesting that the ability to utilize eye gaze as a referential cue is not a primary mechanism by which boys with FXS are acquiring larger receptive vocabularies. It is interesting that a number of subtle differences between the participants with FXS and those with nonsyndromic ASD were noted on the word-learning task (i.e., effect of initial trial type on performance and selection of familiar objects as error responses), highlighting the importance of further understanding the complex interplay of genetic, developmental, and experiential factors that are manifested through observable behaviors.

A major difference between the current study and previous studies of children with nonsyndromic ASD (Baron-Cohen et al., 1997; Luyster & Lord, 2009) is the

number of experimental trials administered. The current study included three trials in each condition, with each trial consisting of both a comprehension probe and a generalization probe. This yielded a total of six opportunities for each participant to respond in each condition. Most previous studies using this paradigm administered only one trial of each condition across many participants. We reasoned that additional trials, as well as the inclusion of generalization probes, would provide a more stringent test of word learning than would a single trial. In addition, TD toddlers who participated in the original Baldwin (1991) study did not see the examiner's object during presentation of the novel word in either condition because that object was kept in a container throughout the trial. Failure to see the examiner's object may have differentially influenced response patterns. Furthermore, there are differences across studies in the number of times the novel object was labeled as well as the number of distractors that were presented during comprehension probes. Last, in the current study the objects were arranged systematically in a divided tray during test probes, whereas in other studies all of the objects were placed in a bag or other container and participants were asked to retrieve the target object from the container. We reasoned that such a response format would have interfered with correct responding for children with FXS, who benefit from environmental structure to display what they know. Thus, our test probes varied from previous studies in a number of ways.

Limitations

The word-learning task used in the current study required compliance and cooperation on the part of the participants. Our stringent requirement that only data from participants who completed six valid trials be included in the analyses left us with a reduced sample size and may have resulted in a less representative sample of children with FXS, children with nonsyndromic ASD, or even TD children. Those children who contributed six valid trials may have been more compliant and willing or able to engage with an examiner, follow instructions, and respond to task parameters.

Although the current results suggest that participants did, in fact, utilize the examiner's direction of gaze to guide word learning in the discrepant labeling condition, the use of this strategy would be further supported by an observational analysis of whether participants shifted their gaze away from their own object and checked in with the examiner during discrepant labeling trials. Last, the matching process utilized in the current study resulted in the elimination of participants with nonsyndromic ASD whose nonverbal cognitive ability exceeded 85, as few boys with FXS achieve this level of cognitive functioning. Thus, participants with nonsyndromic ASD in the current study do not constitute a representative sample of individuals with nonsyndromic ASD. Because of this, conclusions from our between-syndromes comparisons may be applicable only to lower functioning individuals with nonsyndromic ASD.

Conclusions and Future Directions

Findings from the present study contribute to our understanding of the ways in which children with FXS are able to learn new words in an interactive context. These findings also add to the growing body of evidence suggesting that important social processes may differ between FXS and nonsyndromic ASD (McDuffie et al., 2014; Thurman et al., 2014; Wolff et al., 2012). Given that social anxiety, challenging behaviors, and inattention are frequent phenotypic characteristics of boys with FXS, it is interesting that participants with FXS showed subtle advantages over participants with nonsyndromic ASD during follow-in labeling trials. This advantage was noted despite closely matched levels of nonverbal cognition and chronological age. These findings add to the evidence that the behavioral phenotype of boys with FXS may differ in important ways from the behavioral phenotype of boys with nonsyndromic ASD and that these differences may result in distinctions in the ways in which these individuals engage in social learning. Future research should examine a wider range of phenotypic characteristics.

One extension of the current study that would provide additional insight into the process of word learning in nonostensive contexts for children with either FXS or nonsyndromic ASD would involve the behavioral coding of looks to the examiner during both follow-in and discrepant trials. These data would allow us to determine whether the frequency with which a child "checks on" the examiner's direction of gaze is related to levels of task performance. Furthermore, such behavioral coding would enable the determination of whether above-chance levels of word learning in the discrepant labeling conditions is in fact a result of the participant shifting gaze to the speaker and using the speaker's direction of gaze to guide learning. Future studies should additionally examine how children with FXS and nonsyndromic ASD use other types of social cues to support word learning.

Understanding differential patterns and correlates of word-learning performance can help more clearly elucidate the unique behavioral phenotypes of FXS and nonsyndromic ASD and can lead to a better understanding of whether overt behaviors that are shared between neurodevelopmental disorders truly represent shared cognitive mechanisms. The scientific community is currently developing targeted interventions that will treat the core symptoms of nonsyndromic ASD and FXS. Understanding the different behavioral and learning manifestations of underlying mechanisms and how they may differ between disorders is essential for informing the efficient application of targeted interventions, both behavioral and pharmacological.

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