

# Investigating the Receptive-Expressive Vocabulary Profile in Children with Idiopathic ASD and Comorbid ASD and Fragile X Syndrome

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**Abstract** Previous work has noted that some children with autism spectrum disorder (ASD) display weaknesses in receptive vocabulary relative to expressive vocabulary abilities. The current study extended previous work by examining the receptive-expressive vocabulary profile in boys with idiopathic ASD and boys with concomitant ASD and fragile X syndrome (ASD+FXS). On average, boys with ASD+FXS did not display the same atypical receptive-expressive profile as boys with idiopathic ASD. Notably, there was variation in vocabulary abilities and profiles in both groups. Although we did not identify predictors of receptive-expressive differences, we demonstrated that non-verbal IQ and expressive vocabulary positively predicted concurrent receptive vocabulary knowledge and receptive vocabulary predicted expressive vocabulary. We discuss areas of overlap and divergence in subgroups of ASD.

**Keywords** Autism spectrum disorder · Fragile X syndrome · Language phenotype · Vocabulary

## Introduction

Children with autism spectrum disorder (ASD) present with complex behavioral phenotypes (Charman et al. 2011; Thurman et al. 2014). The manifestation of core ASD

symptoms can vary widely and are often related to cognitive and structural language skills. Therefore, research has turned to investigations of more homogeneous subgroups within ASD based on language abilities (e.g., Brady et al. 2015; Kjelgaard and Tager-Flusberg 2001; Roberts et al. 2004; Tager-Flusberg and Joseph 2003). This work is important because studying subgroups within ASD may help in advancing our understanding of phenotypes associated with ASD and potentially enhance our understanding of the etiology (Tager-Flusberg and Joseph 2003; Belmonte and Bourgeron 2006). Additionally, examination of subgroups may allow for advancements in our understanding of underlying mechanisms related to the phenotypic characteristics seen in individuals with ASD.

In addition to examining within-syndrome similarities and differences in language, research has included cross-syndrome comparisons including individuals with similar behavioral phenotypes (e.g., Finestack et al. 2013; Martin et al. 2013; McDuffie et al. 2013; Tager-Flusberg and Calkins 1990). One interesting cross-syndrome comparison group is fragile X syndrome (FXS). FXS is the leading cause of inherited intellectual disability (Hagerman and Hagerman 2002). It is X-linked and caused by a trinucleotide expansion on the *FMRI* gene (Verkerk et al. 1991), which shuts down production of a protein that plays a crucial role in brain development and functioning (Bassell and Warren 2008; Rogers et al. 2001). Males with FXS have a number of cognitive, social, and linguistic deficits, including intellectual disability, language impairment, ASD, and ADHD (Bailey et al. 2001; Roberts et al. 2005).

Notably, individuals with FXS have a high comorbidity of ASD. It is estimated that 50–75 % of males with FXS have a co-diagnosis of ASD (Harris et al. 2008; Hernandez et al. 2009; Klusek et al. 2014b). Furthermore, 50–90 % of boys with FXS display autistic-like

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behaviors, such as hand flapping, hand biting, sensory defensiveness, poor eye contact, restricted interests, and repetitive behaviors (Bailey et al. 2001; Hatton et al. 2006; Rogers et al. 2001). Importantly, the risk of comorbid FXS and ASD is higher than that associated with the majority of known ASD risk factors (Persico and Bourgeron 2006).

Given that FXS is a single-gene disorder that is associated with several phenotypic characteristics of ASD, it has been suggested that studying FXS may provide important information about the etiology of idiopathic ASD, which is genetically more complex and diagnosed based on behavioral assessments (Belmonte and Bourgeron 2006; Jiang et al. 2014). However, it is debated whether a co-diagnosis of FXS and ASD reflects “true” ASD (Abbeduto et al. 2014; McDuffie et al. 2015). Abbeduto et al. (2014) suggest that important differences between individuals with idiopathic ASD and individuals with comorbid ASD and FXS may be masked by a categorical ASD diagnosis rather than a symptom-based description. Also, Abbeduto and colleagues argue that intellectual disability in FXS confounds ASD symptomatology given that ASD symptom severity is associated with IQ in individuals with FXS (Hatton et al. 2006). Conversely, it also has been argued that children with FXS and a co-diagnosis of ASD demonstrate a pattern of behaviors and symptoms more closely related to children with idiopathic ASD compared to children with FXS-only (Budimirovic and Kaufmann 2011; Hernandez et al. 2009), and therefore represent a clinically meaningful group.

One way to explore ASD in FXS is to examine behavioral or language profiles that have been reported in ASD. The language deficits noted in FXS, in terms of rate of acquisition and deficits in syntax and morphology, are strikingly similar to some children with ASD (Roberts et al. 2004). The current study builds on previous work examining a particular language phenotype, discrepancies in receptive and expressive language, in children with ASD (Charman et al. 2003; Hudry et al. 2010; Kover et al. 2013). By comparing children with idiopathic ASD and comorbid ASD and FXS, we can better understand language phenotypes within ASD. Such comparisons also can shed light on subgroups within ASD that demonstrate distinct or overlapping language characteristics. Furthermore, comparing specific behavioral characteristics that are associated in idiopathic ASD between these two groups can provide information that may influence conceptual models of ASD in FXS. Therefore the current study aimed to examine receptive and expressive vocabulary in boys with idiopathic ASD and boys with a comorbid diagnosis of ASD and FXS (ASD + FXS).

## Receptive-Expressive Language Profile in Children with ASD.

Several studies have found an atypical receptive-expressive language profile in some children with ASD, with a reduced receptive language advantage relative to expressive language abilities (e.g., Charman et al. 2003; Ellis Weismer et al. 2010; Woynaroski et al. 2015; see “Appendix” for a comprehensive outline of research on this topic). This atypical receptive-expressive language profile has been noted in omnibus assessments that measure broad language abilities across different domains (i.e., vocabulary and syntax). Hudry et al. (2010) found that at least one-third of the preschoolers with ASD demonstrated an atypical receptive-expressive profile on the Preschool Language Scale (PLS; Zimmerman et al. 1997), the Vineland Adaptive Behavior Scales (VABS; Sparrow et al. 2005), and/or the Communicative Development Inventory (CDI; Fenson et al. 1993). Ellis Weismer et al. (2010) found the same pattern in preschoolers with autism, but this pattern was not present in children with PDD-NOS (based on DSM-IV classifications) or children with developmental delays without ASD. Lastly, Kjelgaard and Tager-Flusberg (2001) found that 57% of their sample (ages 4–14 years) with ASD had significantly higher expressive than receptive language age-equivalent scores on the Clinical Evaluation of Language Fundamentals (CELF-III; Semel et al. 1995; CELF-Preschool; Wiig et al. 1992). Notably though, only about half of their sample was able to complete the CELF.

### Vocabulary

A relative weakness in receptive language has been observed when examining receptive and expressive vocabulary as well. Charman et al. (2003) studied vocabulary abilities in a sample of preschoolers with ASD using the CDI. Compared to the CDI normative data, preschoolers with ASD had a relative advantage in expressive over receptive vocabulary. That is, although children with ASD understood more words than they produced, they produced more words than typically developing preschoolers at the same comprehension level (see also Hudry et al. 2010; Luyster et al. 2007, 2008).

This profile also has been examined in children at heightened risk of having ASD (i.e., having a sibling with an ASD diagnosis). Hudry et al. found that 14-month-old toddlers at heightened risk (HR) of ASD demonstrated a relative weakness in receptive vocabulary abilities. Notably, only HR children who went on to receive an ASD diagnosis or to have atypical developmental outcomes persisted in this atypical receptive-expressive vocabulary profile at 24 months of age (Hudry et al. 2014). Furthermore, receptive and

expressive vocabulary abilities have been examined in older children with ASD with concomitant intellectual disability (ID) between 3 and 11 years of age (Maljaars et al. 2012). Maljaars et al. found that 36 % of the children with comorbid ASD and ID in their sample had higher expressive than receptive age-equivalent vocabulary scores. This profile significantly differed from the children in the other groups without ASD. On average, children with ID without ASD and children with typical development had higher receptive age-equivalent vocabulary scores, a profile that only 10 % of the children with ASD and ID demonstrated.

Although studies have found that some children with ASD have relative weaknesses in receptive vocabulary, this pattern is not consistent for all children with ASD. Kjelgaard and Tager-Flusberg (2001) examined receptive and expressive vocabulary in older children with ASD between 4 and 14 years of age. They found that, on average, receptive and expressive vocabulary standard scores did not significantly differ. Only 4 % of the children with ASD had expressive standard scores that were more than 1 standard deviation above receptive vocabulary standard scores, which was in contrast to their findings of the receptive and expressive higher-order language profile. In addition, Kover et al. found that on average, children with ASD had a greater discrepancy between receptive and expressive vocabulary abilities relative to typically developing children. However, when examining the trajectory of vocabulary development, differences between receptive and expressive vocabulary development were not significant after controlling for nonverbal cognitive abilities (Kover et al. 2013).

Given the mixed findings, it has been unclear whether the atypical receptive-expressive language profile is a significant feature of ASD. A meta-analysis of 74 studies found that, although some children with ASD have relative weaknesses in receptive language, this atypical receptive-expressive pattern was not a robust language feature across children with ASD. As such, the authors concluded that the receptive-expressive language profile is not a useful marker of ASD at the group level (Kwok et al. 2015). However, it could also be argued that the receptive-expressive profile could provide meaningful information about subgroups within ASD and may reveal important information about processing mechanisms. Careful examination of the receptive-expressive language profile also is important because it can speak to the conversation concerning ASD in FXS. Notably, it has been suggested that receptive language weaknesses in children with FXS may provide important information about comorbid ASD features (Lewis et al. 2006; Philofsky et al. 2004).

### Expressive and Receptive Language Profiles in ASD and Comorbid FXS.

Given the debate about the nature of ASD in FXS (Abbeduto et al. 2014; Klusek et al. 2014b), and the research suggesting that receptive language weaknesses may be particularly associated with ASD features in individuals with FXS (Philofsky et al. 2004), careful examination of the receptive-expressive language profile in children with ASD+FXS is warranted. Although some studies have examined receptive and expressive language in children with ASD+FXS, only two studies directly compared the language domains (Philofsky et al. 2004; Roberts et al. 2001).

Roberts et al. (2001) examined whether patterns of global receptive and expressive language development differed according to ASD symptoms in a sample of 39 boys with FXS between the ages of 20 and 81 months. Eight of the 39 boys with FXS met criteria for a comorbid ASD classification. Roberts and colleagues found that, despite substantial variability, overall receptive and expressive language levels did not significantly differ in children with FXS or children with ASD+FXS. They also found that the rate of expressive language growth was significantly slower than the rate of receptive language growth. Moreover, ASD characteristics were not predictive of receptive-expressive discrepancies over time.

Philofsky et al. (2004) examined whether boys with ASD+FXS and boys with FXS who did not meet criteria for ASD (FXS-only) had a similar pattern of early linguistic and cognitive skills relative to young boys with idiopathic ASD. They examined whether receptive language was particularly impaired relative to nonverbal cognition in 18 boys with idiopathic ASD, 10 boys with FXS-only, and 8 boys with ASD+FXS between the ages of 22 and 45 months. Although boys with FXS had higher global receptive language abilities than expressive abilities, regardless of ASD status, children with FXS-only had significantly higher receptive language abilities than children with ASD and ASD+FXS. Philofsky et al. (2004) suggested that receptive language weaknesses may serve as a marker of autism symptoms in FXS.

Relatedly, Lewis et al. (2006) compared global language abilities in adolescents and young adults with FXS-only and ASD+FXS ( $n_s = 10$ ), who were matched on nonverbal cognition and age. Males with ASD+FXS had significantly poorer global receptive language abilities relative to males with FXS-only; however, no group differences were observed on global expressive language abilities. Importantly, other studies have failed to find differences in receptive language abilities in boys with ASD+FXS and FXS-only after controlling for nonverbal cognitive abilities (McDuffie et al. 2012; Price et al. 2007). Notably, Lewis

et al. (2006), McDuffie et al. (2012), and Price et al. (2007) did not compare receptive and expressive language abilities within groups.

While the studies we reviewed provide preliminary insight into receptive-expressive language profiles in males with ASD+FXS and FXS-only, they have notable limitations. First, the previous studies had limited sample sizes, which could have affected the statistical power to detect group differences. Second, only Philofsky et al. (2004) included an idiopathic ASD group with which to compare the ASD+FXS group. Third, the previous studies examined language abilities in either early childhood or adolescence, thereby missing school-age years. Importantly, linguistic skills that are developed in the school-age years are critical to academic success, such as reading (Adlof et al. 2015; Klusek et al. 2015).

Lastly, the previous studies did not test whether children with ASD+FXS have a relative weakness in receptive vocabulary compared to expressive vocabulary. Specific information about vocabulary profiles are needed given that males with FXS have delayed onset of first words and demonstrate other word learning deficits in later years (Abbeduto and Hagerman 1997; McDuffie et al. 2013; Roberts et al. 2007; Sterling and Warren 2008). Although there is a growing literature examining the language phenotype in boys with FXS-only and boys with ASD+FXS, very few studies have focused specifically on receptive and expressive vocabulary. Roberts et al. (2007) examined receptive and expressive vocabulary, along with other speech and language abilities, in 3.5–14 year old children with ASD+FXS and FXS-only. Their study did not specifically test whether receptive and expressive vocabulary abilities differed from one another. However, they reported descriptive data demonstrating that group averages of age-equivalent scores for receptive vocabulary were well above age-equivalent averages for expressive vocabulary in children with ASD+FXS and FXS-only (Roberts et al. 2007). Therefore, more information is needed to substantiate claims that receptive language abilities may be particularly impaired in males with ASD+FXS, relative to males with FXS-only (Lewis et al. 2006; Philofsky et al. 2004).

### Predictors of Language

The documented, yet inconsistent, findings of relative weaknesses in receptive language in children with ASD and limited information for children with ASD+FXS highlight the importance of examining whether child characteristics are associated with vocabulary knowledge. Within the ASD literature, previous work has found that nonverbal cognition is positively associated with receptive and expressive language abilities (e.g., Ellis Weismer et al. 2010; Luyster et al. 2008; but see; Maljaars et al. 2012). In

addition, expressive vocabulary is associated with receptive vocabulary (e.g., Kover et al. 2013) and receptive vocabulary is associated with expressive vocabulary (Luyster et al. 2007). Within the FXS literature, nonverbal cognition also is positively associated with language abilities (McDuffie et al. 2012; Price et al. 2007). Additionally, ASD symptoms have been found to be negatively associated with receptive vocabulary abilities (McDuffie et al. 2012).

Given that nonverbal cognition is a significant predictor of receptive vocabulary for boys with ASD and boys with FXS with and without comorbid ASD, it is important to note a methodological concern when comparing these populations. It is estimated that more than 90% of males with FXS have nonverbal cognitive abilities that are in the intellectual disability range (Hessl et al. 2009). In addition, males with ASD+FXS have been found to have more severe cognitive impairments than males with FXS without a comorbid ASD diagnosis (Hatton et al. 2006; Hernandez et al. 2009; McDuffie et al. 2010). In contrast, it has been estimated that 38% of individuals with idiopathic ASD have ID (Prevalence of Autism Spectrum Disorders 2012). This difference is not only important because cognitive abilities are associated with structural language abilities within ASD and FXS, but also because nonverbal cognition may influence social communication (Abbeduto et al. 2014; Bottcher 2010). Therefore, the relationship between nonverbal cognition and structural language in males with idiopathic ASD and ASD+FXS should be carefully considered.

### *Predictors of receptive-expressive language differences*

In addition to exploring predictors of language abilities in ASD and ASD+FXS, it is important to examine predictors of receptive and expressive language differences. Only one previous study has explored whether child characteristics predicted differences in receptive and expressive vocabulary abilities in children with ASD+FXS. Roberts et al. (2001) found that neither cognition nor ASD characteristics predicted language discrepancies in boys with FXS, regardless of ASD status.

Only three previous studies have examined predictors of receptive-expressive language differences in children with idiopathic ASD. Kover et al. (2013) found that nonverbal cognition correlated with receptive and expressive vocabulary difference scores; however, chronological age and autism severity scores did not. They observed that greater difference scores were associated with higher nonverbal cognitive scores in children with ASD. Hudry et al. (2010) also compared child characteristics between subgroups of preschoolers with ASD who had typical and atypical receptive-expressive profiles and found differences in age, nonverbal cognition, adaptive functioning abilities, and autism severity. Children with



relative weaknesses in receptive language were older, had higher nonverbal cognitive scores, higher adaptive functioning skills, and lower autism severity scores. In contrast, findings from a meta-analysis reported that age, cognition, and language domain failed to predict differences in receptive-expressive language abilities (Kwok et al. 2015). Although Kwok et al. (2015) suggest that inconsistent findings in the literature cannot be explained by chronological age, Woynaroski et al. (2015) proposed that a child's developmental level may influence findings. Therefore, the extent to which child characteristics predict differences in receptive and expressive language abilities has only begun to be explored and has thus far yielded conflicting findings.

### Current Study

The current study extends on the previous literature by examining the receptive-expressive vocabulary profile and predictors of vocabulary in two subgroups of children with ASD: idiopathic ASD and ASD+FXS. We explored this in a slightly older group of children (9–16 years of age), many of whom display significant developmental delays. We asked:

Do boys with idiopathic ASD and ASD+FXS demonstrate differences in receptive and expressive vocabulary abilities and do profiles differ between the groups?

What child characteristics predict receptive and expressive vocabulary differences in the two groups?

What child characteristics predict receptive and expressive vocabulary abilities and do predictors differ between groups?

We predicted that, on average, boys with idiopathic ASD would demonstrate differences between receptive and expressive vocabulary abilities; however, we did not expect all boys with idiopathic ASD to have receptive-expressive vocabulary differences. Additionally, we predicted that the ASD+FXS group would be less likely to demonstrate the atypical receptive-expressive language profile, relative to the idiopathic ASD group. Furthermore, we predicted that child characteristics may be associated with receptive-expressive differences; however, our predictions were tentative given the inconsistency of predictive child characteristics in the literature. Lastly, we predicted that autism severity would be negatively associated with vocabulary, and nonverbal cognition abilities would be positively associated with vocabulary skills.

## Methods

### Participants

A total of 50 children participated in the current study (idiopathic ASD  $n=22$ ; ASD+FXS  $n=28$ ). Due to the

variability seen in females with FXS, we only included boys in the current study. Participants came from a larger study examining syntactic language in boys with ASD and boys with FXS (Haebig et al. 2016). FXS was confirmed via previous molecular genetic testing and all boys with idiopathic ASD completed genetic testing to rule out FXS. All boys with idiopathic ASD had a community diagnosis of ASD. Participants were monolingual English speakers, per parent report.

Of the 22 boys with idiopathic ASD, 18 were reported to be Caucasian, one was reported to be other, and two were reported to be more than one race (one participant's data was not available). Additionally, 18 of the boys with idiopathic ASD were reported to not be of Hispanic or Latino ethnicity, one was Hispanic or Latino. Ethnicity was not reported for three participants with idiopathic ASD. Ten mothers of the boys with idiopathic ASD had a graduate degree or some graduate training, six had a Bachelor's degree, four mothers had some college or technical training, and one mother had a high school degree. Maternal education was missing for one mother. Of the 28 boys with ASD+FXS, 22 were reported to be Caucasian, four were African-American, one was reported to be other, and one was reported to be more than one race. In addition, 24 were reported to be non-Hispanic, two Hispanic or Latino, and ethnicity was not provided for three boys with ASD+FXS. Eight mothers of boys with ASD+FXS had a graduate degree or graduate training, nine had a Bachelor's degree, eight had an Associate's degree or some college training, and three mothers had a high school degree.

### Group Matching

Groups were matched on ADOS severity scores  $t(48)=0.17$ ,  $p=.86$ ,  $d=0.05$  variance ratio=1.20. In addition, the groups did not statistically differ in chronological age  $t(48)=1.53$ ,  $p=.13$ ,  $d=0.44$  variance ratio=1.01 (idiopathic ASD: 9.42–16.75 years; ASD+FXS: 9.25–16.42 years). However, the idiopathic ASD group had significantly higher nonverbal IQ scores than the ASD+FXS group,  $t(48)=6.48$ ,  $p<.001$ ,  $d=1.75$  variance ratio=2.88.

### Procedure

Participants came to the Waisman Center, at the University of Wisconsin-Madison and participated in a 1-day session. Parents provided written informed consent and children provided verbal or written assent. Study procedures were approved by the institutional review board. Children completed cognitive and linguistic assessments and an ASD assessment. Children were given breaks as needed.

### Cognitive and Linguistic Assessments

Nonverbal cognitive ability was measured using the Leiter—Revised (Roid and Miller 1997). Subtests within the Brief IQ Composite measure nonverbal visualization and reasoning abilities. Raw scores, growth scores, age-equivalents, and standard scores were calculated.

The Peabody Picture Vocabulary Test—4th edition (PPVT-4; Dunn and Dunn 2007) was used to measure receptive vocabulary knowledge. Children were presented with four colored images on a page and asked to point to the image that depicts the word the examiner provided. The Expressive Vocabulary Test—2nd edition (EVT-2; Williams 2007) measured expressive vocabulary abilities. On each item, a colored picture was shown to the child and the child was asked to label the image or to identify a synonym for the depicted image. Both assessments yield raw scores, growth scores, age-equivalents, and standard scores. Additionally, the PPVT-4 and EVT-2 were co-normed, which allows for direct comparisons of standard scores. Standard scores and age-equivalent scores are susceptible to floor effects in children with developmental disabilities. Two boys with FXS were at floor on both the PPVT-4 and EVT-2 for standard scores, but not age-equivalents or growth scores. Growth scores are derived from a transformation of the raw scores and form an equal-interval scaled value. Because growth scores are on an equal-interval scale, they reflect more continuous incremental changes in absolute levels of ability and are considered to be more psychometrically sound relative to age-equivalent and raw scores (Dunn and Dunn 2007). Also, they have been noted to be especially useful when studying populations with significant delays (Hooper et al. 2000).

### ASD Assessment

Autism symptoms were measured using the Autism Diagnostic Observation Schedule (ADOS; Lord et al. 1999, 2012), which is a semi-structured assessment through which social communication and restricted and repetitive behaviors are evaluated. One of four modules is administered according to child age and language level. In the ASD group, one child completed the module 1, six children completed the module 2, and 15 children completed the module 3. In the ASD+FXS group, two children completed the module 1, 15 children completed the module 2, and 11 completed the module 3. The ADOS examiner was either research reliable or training for research reliability with a research reliable examiner present for coding. All boys in the ASD group exceeded cut-off scores on the relevant module, confirming the community diagnosis. Similarly, all boys with FXS exceeded ADOS cut-off scores on the relevant module and were determined to meet criteria for ASD.

The ADOS has been used as the sole method of classifying ASD+FXS and FXS-only for several studies of FXS (e.g., Martin et al. 2013; Price et al. 2008). Calibrated severity scores, which facilitate ASD symptom level comparisons across modules, also were recorded to assess associations with ASD symptom severity with other child characteristics. Table 1 provides group characteristics.

### Analysis Plan

To determine whether boys with idiopathic ASD and ASD+FXS demonstrate a similar receptive-expressive profile, we conducted two different sets of analyses. First, we conducted a mixed-effect regression model comparing PPVT-4 and EVT-2 growth scores across the groups. Then, to further examine receptive-expressive profiles, differences in PPVT-4 and EVT-2 standard scores were examined. Difference scores were calculated by subtracting PPVT-4 from EVT-2 standard scores for each child (Kover et al. 2013; Williams 2007). Negative values indicated higher expressive vocabulary than receptive scores. Next, we identified whether differences between receptive and expressive standard scores were significantly meaningful at the 0.05 level relative to the norming sample (Williams 2007, Appendix B7). Normative data reflecting meaningful

**Table 1** Participant characteristics

	Idiopathic ASD	ASD+FXS	Group comparisons <sup>1</sup>
	Mean (SD)	Mean (SD)	
Chronological age	13.39 (1.93)	12.56 (1.91)	<i>p</i> = .134
ADOS severity score <sup>2</sup>	7.68 (1.67)	7.61 (1.40)	<i>p</i> = .867
Nonverbal IQ <sup>3</sup>			
Standard score	71.09 (20.80)	43.79 (7.23)	<i>p</i> < .001
Age equivalent	8.68 (2.75)	4.94 (1.05)	<i>p</i> < .001
Growth score	487.68 (12.00)	461.57 (11.85)	<i>p</i> < .001
Receptive vocabulary <sup>4</sup>			
Standard score	78.45 (21.03)	53.46 (17.66)	<i>p</i> < .001
Age equivalent	9.75 (3.55)	5.98 (2.03)	<i>p</i> < .001
Growth score	170.00 (23.19)	137.18 (24.05)	<i>p</i> < .001
Expressive vocabulary <sup>5</sup>			
Standard score	81.14 (19.47)	54.57 (16.98)	<i>p</i> < .001
Age equivalent	9.74 (3.75)	5.52 (1.77)	<i>p</i> < .001
Growth score	163.82 (15.39)	139.36 (17.12)	<i>p</i> < .001

<sup>1</sup>Paired *t*-tests

<sup>2</sup>Leiter International Performance Scale—Revised, Brief IQ (Roid and Miller 1997)

<sup>3</sup>Autism Diagnostic Observational Schedule (Lord et al. 1999, 2012)

<sup>4</sup>Peabody Picture Vocabulary Test—4th edition (Dunn and Dunn 2007)

<sup>5</sup>Expressive Vocabulary Test—2nd edition (Williams 2007)

receptive-expressive differences are only available for standard score differences. We tested for group differences in meaningful receptive-expressive discrepancies by conducting a series of Fisher's exact tests.

Our second research question asked whether child characteristics predict receptive and expressive vocabulary differences in the two groups. We conducted correlation analyses on receptive-expressive difference scores, age, nonverbal-IQ, and autism severity score. Lastly, our third research question asked whether child characteristics predicted receptive and expressive vocabulary skills and whether predictors differed between groups. Therefore, we conducted two linear regression models examining predictors of vocabulary.

## Results

### Research Question 1: Receptive-Expressive Profile

First, we examined whether boys with ASD+FXS and boys with idiopathic ASD demonstrate a similar receptive-expressive profile. A mixed-effect model was a better fitting model than a regular linear regression model  $p < .001$ . The dependent variable in the mixed-effect model consisted of growth value scores for the PPVT-4 and EVT-2. Growth value scores have sound psychometric properties that are sensitive to small incremental differences in absolute levels of ability and were therefore used instead of age-equivalent scores. Fixed-effect predictors included: group (ASD vs. ASD+FXS), task (PPVT-4 vs. EVT-2), and an interaction between group and task. Participant vocabulary scores were allowed to vary randomly (i.e., random effect). There was a significant effect of group ( $Estimate = -28.64$ ,  $SE = 5.51$ ,  $t = -5.20$ ,  $p < .001$ ), indicating that the boys with idiopathic ASD had significantly higher receptive and expressive vocabulary growth scores than the boys with ASD+FXS. There was a non-significant effect of vocabulary task, ( $Estimate = -2.00$ ,  $SE = 1.82$ ,  $t = -1.10$ ,  $p = .27$ ). However, there was a significant interaction between group and task, ( $Estimate = 8.36$ ,  $SE = 3.64$ ,  $t = 2.29$ ,  $p = .02$ ), indicating that, on average, boys with ASD had greater discrepancies in growth scores between the PPVT-4 and EVT-2 than the boys with ASD+FXS.

We also considered the potential influences of nonverbal IQ in our idiopathic ASD and ASD+FXS group comparison. Nonverbal IQ was positively associated with receptive and expressive vocabulary ( $r_s > 0.6$ ). Previous work has indicated that nonverbal IQ may influence differences in receptive and expressive vocabulary development (Kover et al. 2013). Therefore, we conducted a second mixed-effect regression that included nonverbal IQ growth scores as a covariate to ensure that nonverbal IQ did not

influence the relationship between group and task. Nonverbal IQ was a significant predictor of vocabulary ( $Estimate = 1.20$ ,  $SE = 0.16$ ,  $t = 7.56$ ,  $p < .001$ ). With nonverbal IQ in the model, the effect of group was non-significant ( $Estimate = 2.73$ ,  $SE = 5.58$ ,  $t = 0.49$ ,  $p = .63$ ). Additionally, there was a non-significant effect of vocabulary task ( $Estimate = -2.00$ ,  $SE = 1.82$ ,  $t = -1.10$ ,  $p = .27$ ). However, the group by task interaction remained significant ( $Estimate = 8.36$ ,  $SE = 3.64$ ,  $t = 2.29$ ,  $p = .02$ ).

In addition to assessing differences in PPVT-4 and EVT-2 growth scores, we examined whether boys demonstrated clinically meaningful receptive-expressive differences. Specifically, we tested whether there were group differences in the proportion of boys who met criteria for significantly poorer receptive vocabulary scores than expressive and significantly poorer expressive than receptive vocabulary scores by conducting a series of Fisher's exact tests.

First, we tested whether the proportion of boys with idiopathic ASD who met criteria for significantly poorer receptive vocabulary scores than expressive differed from that of the boys with ASD+FXS. Of the 22 boys with idiopathic ASD, 8 (36.36%) has significantly lower standard scores on the PPVT-4 than the EVT-2. This profile was seen in 4 (14.29%) of the 28 boys with ASD+FXS. The difference between groups in the proportion of boys with relative weaknesses in receptive vocabulary failed to reach significance, Fisher's exact test,  $p = .099$ , two-tailed. In contrast, two (9.09%) of the boys with idiopathic ASD and one (3.57%) boy with FSX+ASD had significantly higher standard scores on the PPVT-4 than the EVT-2. There were no significant group differences, Fisher's exact test,  $p = .576$ , two-tailed.

### Research Question 2: Predictors of Receptive and Expressive Vocabulary Differences

Next we examined if chronological age, ADOS severity score, and nonverbal-IQ were associated with receptive-expressive vocabulary difference scores for each group. None of the predictors were significantly correlated with receptive-expressive difference scores ( $r_s$  ranged from 0.24 to  $-0.20$ ). This pattern was consistent for both groups.

### Research Question 3: Predictors of Receptive Vocabulary

Finally, in addressing our third research question, we identified predictors of vocabulary in two steps, first with correlation and then with linear regression analyses. Despite our prediction, autism severity scores did not predict vocabulary scores. Therefore, autism severity scores were excluded from the linear regressions because our sample

size limited the number of predictor variables that could be included. In our first linear regression model, PPVT-4 growth scores served as the dependent variable and predictor variables included mean-centered nonverbal IQ growth scores, mean-centered EVT-2 growth scores, group, and interactions between group and the other predictor variables. Our model revealed that nonverbal IQ and expressive vocabulary skills were uniquely and positively associated with receptive vocabulary skills. Additionally, the main effect of group and interactions between group and nonverbal IQ and expressive vocabulary were not significant (see Table 2). Figure 1 depicts the relationship between receptive and expressive vocabulary.

We completed a similar linear regression model to assess predictors of expressive vocabulary. Nonverbal IQ did not significantly predict expressive vocabulary scores,  $p = .088$ . However, receptive vocabulary was a significant predictor of expressive vocabulary. Group was not a unique predictor. Additionally, nonverbal IQ and receptive vocabulary did not predict expressive vocabulary differently between the groups (see Table 3).

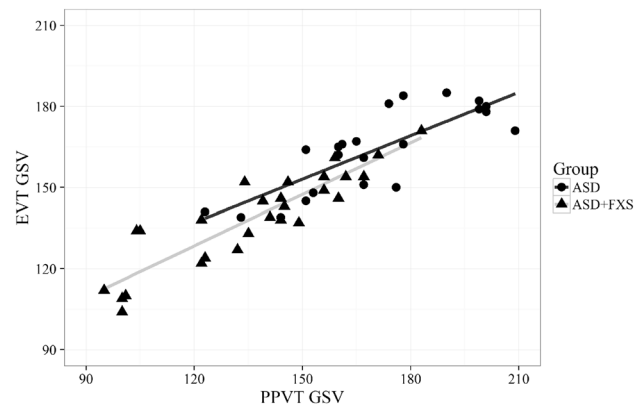
**Discussion**

The current study sought to examine the receptive-expressive language profile that has been discussed in the ASD literature. We did so in two subgroups within ASD: idiopathic ASD and ASD+FXS. Through our comparison, we also aimed to examine the relationship of a comorbid ASD and FXS classification with receptive language weaknesses as a method of exploring the nature of ASD in FXS through the language lens. We found that there was a significant difference in receptive-expressive vocabulary profiles between the groups. On average, the boys with idiopathic ASD had greater discrepancies in growth scores between the PPVT-4 and EVT-2 than boys with ASD+FXS. This remained true even when nonverbal IQ was controlled; therefore, differences in profiles were not fully explained by nonverbal IQ. These findings agree with previous descriptive receptive and expressive vocabulary data from boys

with FXS-only and ASD+FXS presented by Roberts et al. (2007). Although boys with ASD+FXS had similar levels of autism symptomatology as the boys with idiopathic ASD, their vocabulary profile differed.

**Meaningful Receptive-Expressive Vocabulary Differences**

When examining receptive and expressive differences, it is important to provide meaningful characterizations of difference scores. A small difference between the receptive and expressive vocabulary assessments does not necessarily indicate clinically meaningful receptive-expressive vocabulary differences. As such, it is necessary to more thoroughly examine receptive and expressive scores in order to speak to clinical as well as theoretical questions. In the current study, some but not all boys presented with meaningful differences in receptive and expressive vocabulary, according to normative data provided in the EVT-2 manual. Although differences in receptive-expressive profiles differed between groups in the mixed-effect regression models, there was only a marginally higher proportion of



**Fig. 1** Association between expressive and receptive vocabulary

*Note:* This figure illustrates the relationship between expressive and receptive vocabulary for the two groups

**Table 2** Predictors of receptive vocabulary

	Estimate	Std. error	t-value	p-value
Nonverbal IQ	0.42	0.20	2.03	0.049
Expressive vocabulary	1.03	0.15	6.83	<0.001
Group	3.61	5.16	0.70	0.489
Group X nonverbal IQ	-0.56	0.41	-1.37	0.178
Group X expressive vocabulary	0.31	0.30	1.02	0.313

$F(5, 44) = 48.21, p < .001, R^2 = 0.846$

**Table 3** Predictors of expressive vocabulary

	Estimate	Std. error	t-value	p-value
Nonverbal IQ	0.25	0.15	1.74	0.088
Receptive vocabulary	0.49	0.07	6.68	<0.001
Group	-2.01	3.61	-0.56	0.580
Group X nonverbal IQ	0.26	0.29	0.90	0.372
Group X receptive vocabulary	0.01	0.15	0.06	0.949

$F(5, 44) = 49.82, p < .001, R^2 = 0.850$



boys with idiopathic ASD who had meaningful receptive vocabulary weaknesses.

In the current study, 36% of the boys with idiopathic ASD had a meaningful relative weakness in receptive vocabulary. This percentage is relatively similar to previous studies. For example, an atypical receptive-expressive language profile was identified in roughly 33% of Hudry et al. (2010) sample, 36% of Maljaars et al. (2012) sample, and 18% of Kover et al. (2013) sample. Additionally, the percentage of children with idiopathic ASD who had meaningful receptive vocabulary advantages was similar between the current findings and those presented by Kover et al. (respectively, 9 and 10%). Similar to previous studies, a notable number of children with idiopathic ASD demonstrate receptive-expressive differences; however, the majority did not. Although receptive-expressive vocabulary differences do not appear to be a reliable predictor of ASD (Kwok et al. 2015), it is important to understand that children with ASD are at heightened risk of this atypical language profile from early development to school-age and adolescent years. As such, children with ASD who have an atypical receptive-expressive language profile form an interesting subgroup that should be examined carefully across development. Such studies would enhance our understanding of the nature of language subgroups within ASD and allow us to tailor interventions to fit this atypical profile of language development (Pickles et al. 2014).

In addition to examining receptive-expressive vocabulary differences in an older group of boys with ASD, the current study was the first to compare receptive-expressive vocabulary differences in boys with ASD+FXS. Meaningful relative receptive vocabulary weaknesses, according to the EVT-2 manual, were only marginally lower for the ASD+FXS group than the idiopathic ASD group in the current study. Previous studies in children with FXS and ASD+FXS have not reported the percentage of children who demonstrate atypical receptive-expressive language differences. Therefore, the current study provides much needed preliminary information on this language profile. The percentage observed in the current study for the boys with ASD+FXS (14%), appears to be slightly higher than the percentage of typically developing children who have been found to have meaningfully higher expressive than receptive vocabulary abilities (8%, Kover et al. 2013). Additionally, fewer boys with ASD+FXS (3.6%) demonstrated meaningful receptive vocabulary advantages relative to Kover et al. (2013) typically developing group (25%). This pattern is especially interesting given that, judging from descriptive data provided by Kover et al. (2013), receptive and expressive vocabularies appear to be at a similar level for the ASD+FXS and typically developing groups. Therefore, a difference in receptive-expressive profile does not appear to be due to vocabulary level, at least

in this example. However, fewer children with ASD+FXS in the current study demonstrated receptive-expressive vocabulary differences relative to other studies in the ASD literature, including children with ASD+ID (e.g., 33%, Hudry et al. 2010; 36%; Maljaars et al. 2012). It is possible that boys with ASD+FXS may demonstrate smaller receptive advantages than typically developing children, but less severe discrepancies than children with idiopathic ASD. This would support the suggestion that receptive vocabulary weaknesses may be associated with ASD symptoms in boys with FXS (Lewis et al. 2006; Philofsky et al. 2004). As such, it was particularly important to examine whether child characteristics predict receptive-expressive language differences in boys with idiopathic ASD and ASD+FXS.

### Predictors of Receptive-Expressive Vocabulary Differences

In the current study, we examined whether age, nonverbal IQ, and autism severity predicted receptive-expressive difference scores. Unlike previous work (Hudry et al. 2010; Kover et al. 2013), none of the child characteristics were significant predictors. This was true for both the idiopathic ASD group and the ASD+FXS group. Thus far, there is a limited amount of support for predictors of this atypical receptive-expressive profile in the literature, other than having an ASD diagnosis (Ellis Weismer et al. 2010; Maljaars et al. 2012). It is possible that the significant predictors reported by Hudry et al. (2010) represent developmental influences. Perhaps, child characteristics early in development are stronger predictors for receptive-expressive differences than child characteristics later in development. Roberts et al. (2001) also failed to find an association between nonverbal IQ and ASD symptoms in children with FXS-only and ASD+FXS. The current findings, along with Roberts et al. (2001) provide evidence that nonverbal IQ is not the sole and driving factor in the ASD symptoms reported in FXS, despite arguments in the literature supporting this theory (Abbeduto et al. 2014). While the current study does not answer this debate, it provides evidence that continued inquiry is necessary in order to understand the impact of ASD on FXS specifically and the language system more broadly.

### Predictors of Receptive and Expressive Vocabulary

In addition to examining whether child characteristics were associated with receptive-expressive difference scores, we assessed whether child characteristics differentially predicted receptive and expressive vocabulary knowledge in boys with ASD and ASD+FXS. We found that concurrent nonverbal IQ and expressive vocabulary abilities each uniquely predicted concurrent receptive vocabulary

knowledge in both groups. Children with higher nonverbal IQ scores and with higher expressive word knowledge also had higher receptive vocabularies. Additionally, we found that receptive vocabulary was a unique predictor of expressive vocabulary, but that nonverbal IQ was only marginally associated with expressive vocabulary. Given the atypical receptive-expressive vocabulary knowledge profile that some children display, one may have expected expressive vocabulary to play a weak or nonsignificant predictive role in receptive vocabulary than normally observed. This, however, was not the case. The highly positive association between expressive and receptive vocabulary may indicate that children with higher expressive vocabulary abilities may have stronger word representations and therefore a more advanced knowledge base to support word learning opportunities. Additionally, children with larger expressive vocabulary knowledge may be able to more effectively interact with others and therefore gain additional word learning opportunities through such interactions. In fact, a recent study by Woynaroski et al. (2015) revealed that expressive vocabulary was more strongly associated to later receptive vocabulary than early receptive vocabulary to later expressive vocabulary in a group of minimally verbal toddlers with ASD.

Although the current study only examines concurrent predictors, our findings provide support for the idea that expressive vocabulary size may drive or have an atypically strong influence on receptive vocabulary size in some children with ASD. Future work is needed to understand the mechanistic role of expressive vocabulary in supporting receptive vocabulary development. For example, such work could explore whether interventions that emphasize expressive language goals also positively influence receptive language development (Woynaroski et al. 2015). Additionally, studies could examine whether child characteristics such as age, developmental level, language level, or diagnostic classification (e.g., idiopathic ASD, ASD+FXS) impact the efficacy of targeting expressive language to improve receptive language.

In line with studies of younger children with ASD and children with FXS (Ellis Weismer et al. 2010; Luyster et al. 2008; Roberts et al. 2007), nonverbal cognitive abilities were positively associated with receptive vocabulary knowledge in the current study. Of note, the positive relationship between nonverbal IQ and receptive vocabulary was seen in the boys with ASD+FXS, who all had an intellectual disability. Our sample of boys with idiopathic ASD had greater variation in nonverbal IQ scores. In contrast to the ASD+FXS group, about half (10 of the 22) of the boys with idiopathic ASD had an intellectual disability. Despite the differing range of nonverbal cognitive abilities, both groups demonstrated a positive association between nonverbal IQ and receptive

vocabulary. Our study suggests that receptive vocabulary knowledge is supported by abilities both within and outside of the language domain, even in individuals with intellectual disability. However, the unique relationship between nonverbal IQ and expressive vocabulary only approached significance. Unlike receptive vocabulary, linguistic abilities may drive expressive vocabulary abilities more than broader cognitive abilities. Future studies should focus on the role of targeting global cognitive abilities and the influence it may have on more domain-specific skills, such as receptive and expressive language and if this relationship differs between production and comprehension.

Furthermore, it was particularly interesting that ADOS symptom severity was not correlated with receptive or expressive vocabulary. McDuffie et al. (2012) generally found that language abilities did not differ between boys with FXS-only and ASD+FXS once nonverbal IQ was controlled; however, findings were mixed depending on the language measure and the way in which ASD features were measured. Findings by McDuffie et al. (2012) and Price et al. (2007) go against the suggestion that receptive language abilities may be a marker for ASD in FXS (Lewis et al. 2006; Philofsky et al. 2004). The inclusion of a FXS-only group would help disentangle this question. At present, our findings indicated that the severity of receptive language weaknesses is not related to ASD symptoms within children with ASD+FXS.

### Study Limitations and Future Directions

Although the current study contributed needed information to further characterize language impairments and language profiles across these two populations, we were limited in our ability to discuss the causative mechanisms that lead to atypical receptive-expressive language profiles. In addition, our limited sample size restricted the power to assess receptive-expressive language profiles in our two groups and the predictors of receptive-expressive differences. Therefore, our analysis examining predictors of receptive-expressive difference scores served as a preliminary step in identifying potential developmental and cognitive domains that influence receptive-expressive differences. Other domains of language, such as receptive and expressive grammatical skills also should be examined. Future work also would benefit from examining additional predictors and examining longitudinal associations of predictive child characteristics.

Furthermore, we acknowledge that the PPVT-4 and the EVT-2 place some different demands on children, which may influence child performance. Although both tasks require rather minimal social interaction, the PPVT-4 requires the child to point to a picture whereas the EVT-2

requires the child to think of a word that correctly answers the examiner's question and to communicate it vocally to the examiner. Given these task differences, one could argue that it may be more difficult to identify receptive vocabulary weaknesses relative to expressive vocabulary weaknesses especially in this population. Indeed, previous work has found that the PPVT tends to be less sensitive to vocabulary deficits compared to the EVT (Gray et al. 1999; McGregor et al. 2012). Nevertheless, the difference in task demands should rightfully be noted and may possibly point to additional predictors that may influence receptive-expressive language differences that should be explored in future work. Lastly, although the assessments used in the current study are extensively used in research and clinical settings, standardized assessments that are linked, like the PPVT and EVT, should yield more even receptive-expressive profiles in typically developing children, which has not always been the case (e.g., Kover et al. 2013) and potentially raises questions about the assessments.

The current study included boys with FXS who met ADOS cut-off scores for an ASD classification. The ADOS is part of the gold standard assessment measures designed to describe the symptoms of ASD. A significant number of previous studies have used the ADOS in order to classify ASD in FXS (e.g., Hall et al. 2008; Klusek et al. 2014a; Price et al. 2008). The Autism Diagnostic Interview—Revised (Rutter et al. 2003) and the DSM-V also could have been used to categorize boys with FXS as having comorbid ASD, and a comprehensive ASD assessment should be used in future studies.

To more clearly address questions of the nature of ASD in FXS and the influence of ASD symptoms on receptive language, future studies should examine receptive-expressive vocabulary profiles in children with FXS who do and do not have a comorbid ASD diagnosis. The current study only included boys with ASD+FXS as an initial step in examining language phenotypes in subgroups of children with ASD. Given the extent of behavioral overlap that is even seen in children with FXS without an ASD diagnosis and children with idiopathic ASD, it may be interesting to extend work conducted by Roberts et al. (2001) and Philofsky et al. (2004) and examine receptive-expressive vocabulary profiles in a broader sample of children with FXS. Also, an examination of receptive-expressive vocabulary profiles in younger children with FXS would allow for a more complete developmental understanding of skills. Such a study also could be related to the previous studies exploring this question in young children with idiopathic ASD.

## Clinical Implications

The current work points to important clinical implications for clinicians, educators, and parents. First, clinicians should pay particular attention to receptive vocabulary knowledge during assessment and intervention. Often there is an emphasis on increasing spoken vocabularies. Although expressive vocabulary is positively associated with receptive vocabulary, clinicians should carefully examine whether children with ASD and FXS evidence relative receptive weaknesses and monitor growth in both domains in response to language intervention. Additionally, clinicians should work with educators and caretakers in creating a language rich environment to enhance depth and breadth of lexical knowledge.

## Conclusions

The current study demonstrated that, on average, boys with ASD+FXS do not display the same atypical receptive-expressive profile as boys with idiopathic ASD. Boys with ASD+FXS are more likely to have more even receptive-expressive vocabulary abilities. Although meaningful relative receptive vocabulary weaknesses appeared in both groups, the majority of the children did not display this profile. Beyond these group comparisons, we did not identify clear predictors of receptive-expressive differences for either group. However, we demonstrated that nonverbal IQ and expressive vocabulary are positively associated with receptive vocabulary knowledge in boys with ASD as well as boys with ASD+FXS. Additionally, receptive vocabulary predicted expressive vocabulary, but nonverbal IQ was only a marginally significant predictor of expressive vocabulary. Through the comparison of subgroups (i.e., idiopathic ASD and ASD+FXS), the current study highlights areas of overlap and divergence in language profiles within the broader ASD umbrella.

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**Informed Consent** Written informed consent was obtained from all parents and all children provided verbal or written assent.

**Compliance with Ethical Standards**

**Conflict of Interest** EH has received F31 DC013485 that supported the work on this project. AS received a research Grant from the R03 DC011616 that supported this study. The authors have no other conflict of interest to report.

**Appendix**

See Table 4.

**Table 4** Studies directly comparing receptive and expressive language in children with ASD and children with FXS

Population	Receptive assessment	Expressive assessment	Age (years)	Sample size	Cohen's <i>d</i> <sup>a</sup>
<b>ASD</b>					
More advanced expressive than receptive language					
Charman et al. (2003)	CDI-infant form	CDI-infant form	1.5–7.33	134	NA
Luyster et al. (2007)	CDI (vocabulary)	CDI (vocabulary)	~2.5 (range not available)	93	NA
Woynarowski et al. (2015)	CDI (vocabulary)	CDI (vocabulary)	2–4	87	-.24 to -.37
Hudry et al. (2010)	CDI (vocabulary), PLS-3-UK, VABS-II	CDI (vocabulary), PLS-3-UK, VABS-II	2–4.9	152	CDI = -.40 PLS = -.13 VABS = -.33
Luyster et al. (2008)	CDI (vocabulary), MSEL	CDI (vocabulary), MSEL	1.5–2.75	164	NA
Hudry et al. (2014)	CDI, VABS-II, MSEL	CDI, VABS-II, MSEL	~0.62 (range not available)	54 high ASD risk (ASD <i>n</i> = 17)	NA
Maljaars et al. (2012)	Reynell/Dutch-CDI	Schlichting/Dutch-CDI	3.3–11.3	ASD+ID <i>n</i> = 36	.13
Ellis Weismer et al. (2010)	MSEL (1989 or 1995), SICD	MSEL (1989 or 1995), SICD	2–3	257	MSEL = -.38 SICD = -.32
Barbaro and Dissanayake (2012)	MSEL	MSEL	~1.1 (range not available)	125	NA
Pickles et al. (2014)	VABS/VABS-II, MSEL	VABS/VABS-II, MSEL	2–19	192	NA
Kover et al. (2013) <sup>b</sup>	PPVT-III	EVT	4–11	49	-.34
Jarrold et al. (1997) <sup>c</sup>	BPVS	WFT	5.5–19.58	120	NA
Volden et al. (2011)	PLS-IV	PLS-IV	2–4.92		-.31
Kjelgaard and Tager-Flusberg (2001)	CELF (preschool or III)	CELF (preschool or III)	4–14	44	-.21
More advanced receptive than expressive language					
Luyster et al. (2008)	VABS	VABS	1.5–2.75	164	NA
Ellis Weismer et al. (2010)	VABS/VABS-II	VABS/VABS-II	2–3	257	.34
Equivalent receptive and expressive language					
Jarrold et al. (1997)	TROG	APTG	5.5–19.58	120	NA
Kjelgaard and Tager-Flusberg (2001)	PPVT-III	EVT	4–14	81	.07
Philofsky et al. (2004)	MSEL	MSEL	1.83–3.75	18	.02
Kwok et al. (2015)	Meta-analysis	Meta-analysis	1–20	74 studies	
<b>FXS</b>					
Equivalent receptive and expressive language, but lower receptive language in the ASD+FXS group relative to the FXS-only group					
Roberts et al. (2001)	Reynell	Reynell	1.58–6.75	FXS-only <i>n</i> = 31 ASD+FXS <i>n</i> = 8	NA



**Table 4** (continued)

Population	Receptive assessment	Expressive assessment	Age (years)	Sample size	Cohen's $d^a$
Philofsky et al. (2004)	MSEL	MSEL	1.83–3.75	FXS-only $n=10$ ASD+FXS $n=8$	FXS-only = 1.39 ASD+FXS = 1.44

<sup>a</sup>Cohen's  $d$  values were included if they were provided in the article or calculated if means and standard deviations for the ASD group were provided. NAs signify that the information was not available to calculate Cohen's  $d$

<sup>b</sup>After controlling nonverbal IQ the trajectory of receptive and expressive vocabulary abilities would be categorized "Equivalent receptive and expressive language"

<sup>c</sup>After applying a Bonferroni multiple comparison correction, expressive vocabulary was no longer significantly higher than receptive

PPVT peabody picture vocabulary test (Dunn and Dunn 1997), EVT expressive vocabulary test, PLS preschool language scale (Zimmerman et al. 1997), Reynell Reynell developmental language scales (Reynell and Gruber 1990), CDI MacArthur-Bates communicative development inventory (Fenson et al. 1993), Schlichting Schlichting test for Dutch language production (Schlichting et al. 1995), MSEL Mullen scales of early learning (Mullen 1995), SICD sequenced inventory of communication development (Hendrick et al. 1984), VABS vineland adaptive behavior scales (Sparrow et al. 1984, 2005), CELF clinical evaluation of language fundamentals (CELF-III: Semel et al. 1995; CELF-III-UK: Semel et al. 2000; CELF-P; Wiig et al. 1992), BPVS British picture vocabulary scale (Dunn and Dunn 1997), TROG test for reception of grammar (Bishop 1983), WFT Renfrew word finding vocabulary test (Renfrew 2010), APTG action picture test grammar scale (Renfrew 1988)

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