Expressive Dominant Versus Receptive Dominant Language Patterns in Young Children: Findings from the Study to Explore Early Development

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Abstract

We examined language profiles of 2571 children, 30–68 months old, with autism spectrum disorder (ASD), other developmental disabilities (DD), and typical development from the general population (POP). Children were categorized as expressive dominant (ED), receptive dominant (RD), or nondominant (ND). Within each group, the ED profile was the least frequent. However, children in the ASD group were more likely to display an ED profile than those in the DD or POP groups, and these children were typically younger, had lower nonverbal cognitive skills, and displayed more severe social-affect symptoms of ASD compared to their peers with RD or ND profiles. These findings have research and clinical implications related to the focus of interventions targeting young children with ASD and other DDs.

Keywords Autism spectrum disorder · Expressive language · Receptive language · Mullen Scales of Early Learning

Introduction

The early language development skills of young children with autism spectrum disorder (ASD) have been the focus of attention over the last decade. Although much has been learned about these young children's language, one area that has remained unresolved by research is the nature of the relationship between children's receptive and expressive language skills. Children with typical language development generally understand much more vocabulary and more complex language structures than they express, but the gap between receptive language and expressive knowledge seems to be atypically small for at least some children with ASD, resulting in higher standard scores or age equivalent scores for expressive language than receptive language. A number of studies have reported such an atypical expressive-receptive language pattern in young children with ASD, contrasting with the language profiles seen in samples of children with typically developing language (TD) and/or those with other developmental disabilities (DD) (Barbaro and Dissanayake 2012; Charman et al. 2003; Davidson and Ellis Weismer 2017; Ellis Weismer et

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al. 2010; Hudry et al. 2010; Kjelgaard and Tager-Flusberg 2001; Kover et al. 2013, Luyster et al. 2008; Luyster et al. 2007; Volden et al. 2011; Woynaroski et al. 2016).

In their longitudinal study of initially nonverbal or low-verbal toddlers and preschoolers with ASD, Woynaroski et al. (2016) compared receptive and expressive vocabulary levels at repeated visits over 16 months. The children were between 24 and 48 months of age and said no more than 20 words at study entry, according to parent report. The results showed that expressive language age equivalent scores on the MacArthur-Bates Communicative Development Inventories: Words and Gestures checklist (MCDI) exceeded receptive language age equivalent scores at each of four time points. Although the differences tended to become smaller over time, this trend was nonsignificant (Cohen's ds = 0.37, 0.34, 0.24, 0.24, respectively, all $ps > 0.50$ for all comparisons of one time point against another). The data also revealed that early expressive vocabulary is more predictive of later receptive vocabulary size than early receptive vocabulary is predictive of later expressive vocabulary size for this group of initially minimally verbal children with ASD. This contributes to the notion that these children learn differently. These results indicate that an atypical expressive-receptive profile characterized the early stages of language development in this sample and was consistent with the hypothesis that expressive language was driving the development of later receptive language, contrary to evidence from other populations that receptive language development drives expressive development (Woynaroski et al. 2016). Consequently, the process of language acquisition may be different for some children with ASD (e.g., children who are relatively young and with significant language delays).

Although a number of studies, cited above, have found that expressive language scores exceed receptive language scores in individuals with ASD, a recently published meta-analysis of studies reporting both receptive and expressive language scores for individuals with ASD (Kwok et al. 2015) indicated that a number do not. Of the 12 studies cited in Kwok et al. (2015), that directly compared receptive and expressive skills in children with ASD, two reported findings of more advanced receptive than expressive language (but only on the Vineland Adaptive Behavior Scales; Luyster et al. 2008; Ellis Weismer et al. 2010), and three reported findings of equivalent receptive and expressive language levels (Loucas et al. 2008; Kjelgaard and Tager-Flusberg 2001; Jarrold et al. 1997). Nine of these studies reported evidence of more advanced expressive than receptive skills on at least one language measure. The Kwok et al. (2015) meta-analysis included a much larger number of students (n = 74), however, that reported both receptive and expressive language scores for children with ASD. Results of their meta-analysis did not support an unusual prevalence of a profile in which expressive skills exceed receptive skills among children with ASD.

A few studies have examined within-sample variables—child age, nonverbal cognitive level, and ASD symptom severity—to determine whether they associated with the size of the gap between receptive and expressive language levels in children with ASD. Outcomes of two studies examining children in the toddler to preschool age range suggested that as children with ASD get older, fewer children show a relative weakness in receptive language skills compared to expressive skills (Davidson and Ellis Weismer 2017; Seol et al. 2014). In contrast, another study of preschool children (Hudry et al. 2010) as well as a study of children aged 4 to 11 years (Kover et al. 2013) found that the receptive disadvantage in vocabulary relative to expressive vocabulary was more prominent in older children. These same two studies (Hudry et al. 2010; Kover et al. 2013) reported that higher nonverbal cognitive skills were associated with a greater receptive disadvantage compared to the expressive scores in children with ASD, whereas Volden et al. (2011) found that as nonverbal mental age scores increased, the degree to which expressive scores exceeded receptive scores diminished; adding to the lack of consistency in findings, Davidson and Ellis Weismer found no association between nonverbal cognitive scores and the expressive-receptive language discrepancy among young children with ASD. Finally, there has been some interest in whether severity of ASD symptoms is associated with the discrepancy between expressive and receptive language. Kover et al. (2013) found no association between the expressive-receptive language discrepancy and the ADOS total calibrated severity scores (computed per Gotham et al. 2009), whereas Hudry et al. (2010) found that children who showed relatively stronger expressive than receptive skills had lower total calibrated severity scores. Looking specifically at a behavior in the social affect symptom domain, McDaniel and colleagues (McDaniel et al. 2018) found that greater amount of time spent looking at a speaker was associated with a more normative profile of receptive and expressive vocabulary. This finding suggests that a greater severity of social affect domain symptoms may be associated with a greater relative expressive advantage/receptive disadvantage among young children with ASD.

The goal of this study was to examine patterns of receptive and expressive language in a population-based sample of children aged 2–5 years in three well-characterized groups of children: those with autism spectrum disorder (ASD, n = 695), non-ASD related developmental disabilities (DD, n = 987), and those sampled from the general population (POP, n = 889). Based on a procedure, similar to that used by Seol et al. (2014), we classified children in each group according to one of three profiles based on their expressive and receptive language age equivalent scores: expressive dominant, receptive dominant, and nondominant.

The aims of this study were to determine (1) whether and how the proportions of expressive dominant and receptive dominant children differ by case status (ASD, DD, POP), (2) whether children who are expressive language dominant will have lower receptive and expressive language age equivalent scores than children who are receptive language dominant, and (3) for the ASD group, whether the degree of the discrepancy between receptive and expressive language scores will vary according to child chronological age (CA), severity of ASD social affect symptoms, and/or nonverbal cognitive level, as well as child sex, and maternal education. The literature cited above suggests that child CA, ASD symptom severity, and nonverbal cognitive level might be associated with language profiles among children with ASD. Additionally, child sex and maternal education were included due to their widely reported associations with preschool language skills in other populations (e.g., Lange et al. 2016; Rice and Hoffman 2015; Rowe et al. 2016) and potential for confounding the group comparisons in the current study; studies of language among children with ASD have rarely included enough females or sufficient variability in parental education to provide the power to examine these factors.

For aim (1), we hypothesize that some children with ASD are more likely to have an ED profile than are children in DD or POP groups. For aim (2) we hypothesize that children who are ED will have lower receptive and expressive language age equivalent scores than children who are RD, and for aim (3) that children who are younger and have more severe ASD social affect symptoms and lower cognitive levels will tend to have the largest gaps between receptive and expressive language scores with expressive scores higher than receptive.

Methods

Participants

The Study to Explore Early Development (SEED) Phase I was a multi-site, observational case–control study of the causes and correlates of ASD that asked children ages 2 to 5 years and their parents to complete multiple data collection components, including developmental assessments. Details of the recruitment and enrollment processes and data collection components for SEED are reported elsewhere (Schendel et al. 2012). Briefly, the study included catchment areas within six states: California, Colorado, Georgia, Maryland, North Carolina, and Pennsylvania. Eligible children were born between September 1, 2003 and August 31, 2006 and were age 30 to 68 months at the time of assessment. Participants were born and residing in the catchment areas at the time of study participation, had a legal guardian available to consent, and a consistent caregiver since 6 months of age (or younger) who spoke English (all sites) or Spanish (California and Colorado only). Families with a child suspected of having an ASD or other DD were identified from clinical, and educational sources within each site's catchment area. Population controls were identified from birth records in the catchment areas. Data collection occurred between 2007 and 2011.

Assessment Measures

All children who were seen in person were administered the Mullen Scales of Early Learning (MSEL) (Mullen 1995) to assess basic cognitive development. The MSEL is validated for children from birth to 68 months of age. Research reliable clinicians with advanced degrees administered scales for four developmental domains: Expressive Language, Receptive Language, Visual Reception, and Fine Motor (Schendel et al. 2012). Of particular interest for this analysis are the Receptive and Expressive Language Scales. The specific items administered to a child depend on the child's level of language functioning, following the test's basal and ceiling rules. The Receptive Language Scale measures auditory comprehension and memory by, for example, asking children to point to named items ("point to the car" or "point to the stick that is longer") or follow verbal commands of varying complexity ("give me the ball," "put the bear beside the table," or "give me the crayon, then put the car on the chair"). A few items require one- to three-word vocalizations such as "What is your name?" or "What do we use to wash our hands?" (soap and water). The Expressive Language Scale measures speaking ability and language formation. Some items are scored based on the examiner's observations of the child's verbalizations—for example, is s/he making sounds like "dada" or engaging in pat-a-cake or using multi-word sentences? Other items require the child to name objects—for example, "What is this?" and show them a key or a picture of a house. At higher skill levels, items require children to answer questions such as "What do you do when you are sleepy?" or to complete verbal analogies such as "During the day it is light; at night it is _____." (dark). If children obtained an overall standard score of less than 78 standard points on the MSEL, the Vineland Adaptive Behavior Scales-Second Edition (VABS-II) (Sparrow et al. 2005) was administered to the parent. The VABS-II is administered to a parent or caregiver in a semi-structured interview format and is standardized for individuals aged birth through 90 years. It includes the domains of receptive and expressive language, daily living skills, socialization, and motor skills.

Children with a previous diagnosis of ASD or who scored 11 or higher on the Social Communication Questionnaire (SCQ) (Rutter et al. 2003), used to screen for ASD during the initial telephone recruitment call, were scheduled for a further evaluation for ASD. This included the Autism Diagnostic Observation Schedule (ADOS) (Lord et al. 1999) and Autism Diagnostic Interview-Revised (ADI-R) (Lord et al. 1994). The ADOS is a semi-structured assessment for individuals suspected of having autism. It assesses the areas of communication, social interaction, restricted and repetitive behaviors/interests, and play. The ADI-R is a structured interview that can be conducted with parents or caregivers of individuals with a mental age of 18 months or higher who are suspected of having autism. It focuses on the areas of communication, reciprocal social interaction, and patterns of repetitive behavior.

Clinicians who administered the ADI-R and ADOS were required to establish and maintain ongoing reliability, as well as to verify administration fidelity. Overall, quarterly inter-site reliability among SEED clinicians was 99% on first-pass ADI-R and ADOS exercises, and 100% on second-pass ADI-R and ADOS exercises for those who did not achieve reliability on the first pass. Quarterly intra-site reliability among SEED clinicians was 87% on first-pass ADI-R exercises, 99% on first-pass ADOS exercises, and 100% on second-pass ADI-R and ADOS exercises for those who did not achieve reliability on the first pass.

Final Study Classification

Based on results from the in-person assessment, the SEED final classification algorithm was used to classify each child into one of four study groups: ASD, Suspected ASD but incomplete data (i.e., Incomplete Classification), DD, and POP. The SEED final classification algorithm was based on best practice guidelines, review of the literature, and clinical experience, as described in Wiggins et al. (2015) (PMD: 25348175). The study was approved by each participating site's IRB.

Statistical Analysis

Addressing our research aims required a strategy for classifying children according to language dominance "patterns" representing their relative performance on expressive versus receptive language scales. We adopted a procedure similar to that used by Seol et al. (2014) for this purpose. This procedure included determining the language dominance pattern by dividing the MSEL Receptive Language age equivalent (RLAE) score by the Expressive Language age equivalent (ELAE) score. We then used the distribution of this ratio in the POP group to derive standard cut offs, based on the mean $\pm$ one standard deviation, for our entire sample. Thus, participants with a ratio less than $1.04 - 0.13 = 0.91$ were categorized as expressive dominant (ED), those with a ratio greater than $1.04 + 0.13 = 1.17$ were categorized as receptive dominant (RD), and those with a ratio between 0.91 and 1.17 were categorized as non-dominant (ND). We also performed two sensitivity analyses to assess the stability of results using different cut offs for the language dominance categories. In the first sensitivity analysis, we derived the standard cut offs in the POP group according to tertiles of child chronological age. In the second sensitivity analysis, we excluded all participants with an ELAE score of 70 or a RLAE of 69 (these AE scores are at the ceiling of expressive language and receptive language, respectively).

Although symptoms in both the Social Affect (SA) and Restricted and Repetitive Behaviors (RRB) domains are required for a diagnosis of ASD, the correlations among symptoms in these two broad domains are modest (Dworzynski et al. 2009; Mandy and Skuse 2008). Related to this, Hus et al. (2014) point out that measuring severity based on a combination of SA and RRB domains will obscure very different profiles of symptom severity. For example, one child may show more extreme repetitive and restricted behaviors with relatively less social-communication impairment, whereas another child with the same severity score may have relatively less severe symptoms in the repetitive and restricted behavior domain but substantial social-communication difficulties. Based on the importance of the distinction between the two domains and considering the implicit link between language (as the focus of this study) and social communication (Thurm et al. 2015), the SA domain rather than total calibrated symptom severity was chosen as the symptom severity measure for this study. Social affect severity scores were calculated based on the ADOS raw domain total using the algorithm provided in Hus et al. (2014). This process standardizes participant scores, so they can be compared across different ADOS modules based on the same scale.

Among the children with ASD and DD, a large proportion (41% and 10%, respectively) had Visual Reception scores on the MSEL at 20, the lowest possible standard score on this measure. Therefore, we derived a nonverbal IQ proxy score that would avoid floor effects by computing the ratio of the Visual Reception age equivalent (VRAE) in months to the child's chronological age (CA) in months (VRAE:CA).

Tests of differences in proportions between ED and RD participants for each case status were conducted using Chi square tests. An overall test to examine the association between case status and language dominance was conducted using the Cochran-Mantel–Haenszel Chi square statistic, and a generalized logit model was fit to produce odds ratios and 95% Wald confidence intervals.

Means (averaging over levels of other covariates) for receptive and expressive language raw scores and age equivalent (AE) scores by case status were generated from a linear regression model, adjusting for child age at clinic visit, language dominance, VRAE:CA ratio, sex, maternal age, and maternal education. Case status interactions with language dominance and sex were also included. Two-way comparisons of differences in means of receptive and expressive language raw scores and AE scores for each case status and language dominance combination were computed using a Tukey adjustment method for multiple comparisons.

To evaluate the degree of the discrepancy between receptive and expressive language scores, linear regression models were utilized to assess the association of the following characteristics with the difference between a child's ELAE and RLAE score—social affect severity, maternal education, maternal age, child chronological age (CA) at clinic visit, child sex, VRAE:CA ratio, and interactions between social affect severity and education, child CA, and child sex. Interaction terms that were not statistically significant were removed from the model using a backward elimination model selection method.

As a sensitivity analysis to account for ineligible participants (i.e., missing ELAE or RLAE scores), we used an inverse probability weighting (IPW) model. The IPW model weighted study participants by the inverse of the probability that they were ineligible, estimated using a logistic regression model, to compensate for underrepresentation of persons with characteristics associated with ineligibility. The logistic regression model included child year of birth, maternal age at child birth, child sex, maternal education, maternal ethnicity, and maternal race, as well as all two-way interactions between variables. Forward selection was used with a p value threshold of $p=0.15$ to select the terms to be added to the model. Model fit was evaluated using Hosmer–Lemeshow goodness-of-fit statistic, and weights were examined for stability and were normalized to the sample size by eligibility status.

All analyses were conducted at the alpha = 0.05 significance level and performed using SAS/STAT software, Version 9.3 of the SAS System for Windows. Copyright © 2010 SAS Institute Inc. SAS and all other SAS Institute Inc. product or service names are registered trademarks or trademarks of SAS Institute Inc., Cary, NC, USA.

Results

Of the 3899 children enrolled in SEED, we excluded 130 siblings, 569 who could not be clearly classified into one of the three study groups (ASD, DD, or POP) due to incomplete MSEL or a mental age that was too low for administration of the ADOS and/or ADI (rendering them invalid), and four whose chronological age (CA) was older than 68 months at the time of the developmental assessment. Additionally, participants who were not seen in person for the developmental evaluation were ineligible for this analysis (n = 625). While the 625 participants did not differ significantly in child CA ($p=0.83$) or maternal ethnicity ($p=0.31$) compared to those included in the current analysis, eligible participants were more likely to be male ($p<0.01$), white maternal race ($p=0.02$) and higher maternal education ($p<0.01$) (Table 1).

The observed differences between the eligible and ineligible participants on each characteristic were eliminated by using IPWs to adjust the sample; this indicates that the IPW model worked well and demonstrates balance between the eligible analytical group and the ineligible group on these key characteristics (Table 1). In sensitivity analyses, IPW-adjusted results were not substantively different from those that were unadjusted (Supplemental Tables 1–4). Additionally, results of sensitivity analyses using different cut offs for language dominance categories (including tertiles by chronological age and exclusion of ELAE or RLAE scores at the ceiling) were not appreciably different from the results of the primary analysis (Supplemental Tables 5–8). Thus, results of the unadjusted analyses using language dominance patterns derived from the POP group are presented in the remainder of this section.

The 2571 remaining eligible children included 695 with ASD, 987 with DD, and 889 in the POP group. No significant differences in child CA at clinic visit were present by case status ($p=0.79$), nor were there significant differences in maternal age ($p=0.19$) or maternal ethnicity ($p=0.09$) (Table 2). The distribution of sex differed significantly ($p<0.01$) by case status. Males represented 82% of children with ASD, 67 percent of children with DDs, and 53 percent of children from the POP group. The distribution of maternal education also differed significantly by case status ($p<0.01$). Seventeen and 18 percent of children with ASD and DD, respectively, had a mother with a high school diploma, while 11 percent of POP mothers had a high school diploma. Alternatively, 28 and 27% of mothers of children in the ASD and DD groups, respectively, had an advanced degree, compared to 37% of POPs. Statistically significant racial differences are present among the case statuses ($p<0.01$), with white maternal race representing 68, 74, and 80% of children classified as ASD, DD, and POP, respectively.

Clinical characteristics of the study participants differed significantly by case status ($p<0.01$). Those in the ASD group had lower mean expressive and receptive language raw scores and AE scores, lower VRAE scores, and lower VRAE:CA ratios on average, compared to those in the DD and POP groups.

Comparison of Case Status by Language Dominance

Figure 1 shows that the pattern of the distribution of language dominance is not homogeneous across the case status groups ($p<0.01$). A higher proportion of participants in the ASD group were ED (24%) and RD (30%) and a lower proportion were ND (47%), compared to the DD and POP

Table 1 Characteristics of study participants by eligibility status

	Eligibility status[a]			Unadjusted *p* value**	IPW adjusted *p* value**
	Overall (N = 3196)	Eligible (N = 2571)	Not eligible (N = 625)		
Child CA (mo) at clinic visit	59.4 ± 7.1	59.4 ± 7.1	59.0 ± 6.5	0.830	0.83
Maternal age (year) at child birth	31.6 ± 5.7	31.8 ± 5.6	30.7 ± 6.3	< 0.001	0.90
Sex				0.003	0.70
Female	1122 (35%)	872 (34%)	250 (40%)		
Male	2069 (65%)	1699 (66%)	370 (59%)		
Not specified[b]	5 (0%)		5 (1%)		
Maternal education				< 0.001	0.92
8th grade or less	50 (2%)	39 (2%)	11 (2%)		
9th–12th grade	158 (5%)	113 (4%)	45 (7%)		
High school	526 (16%)	395 (15%)	131 (21%)		
Some college, no degree	650 (20%)	517 (20%)	133 (21%)		
Bachelor's degree	799 (25%)	672 (26%)	127 (20%)		
Advanced degree	956 (30%)	790 (31%)	166 (27%)		
Unknown/Missing[b]	57 (2%)	45 (2%)	12 (2%)		
Maternal ethnicity				0.313	0.97
Not Hispanic	2440 (76%)	1957 (76%)	483 (77%)		
Hispanic	353 (11%)	275 (11%)	78 (12%)		
Unknown/Missing[b]	403 (13%)	339 (13%)	64 (10%)		
Maternal race				0.017	0.33
White	2349 (73%)	1915 (74%)	434 (69%)		
Black, African-American	601 (19%)	455 (18%)	146 (23%)		
American Indian	14 (0%)	12 (0%)	2 (0%)		
Asian, Pacific Islander	169 (5%)	139 (5%)	30 (5%)		
Other, Multiracial	39 (1%)	34 (1%)	5 (1%)		
Unknown/Missing[b]	24 (1%)	16 (1%)	8 (1%)		

***p* value comparisons across case status for categorical variables are based on Chi square test of homogeneity; *p* values for continuous variables are based on ANOVA

[a]Values expressed as N (%) or mean ± standard deviation

[b]*p* value calculation does not include not specified, missing, or unknown categories

groups. The majority of DD and POP children were ND (63% and 74% respectively).

Adjusted means of receptive and expressive language raw and AE scores in [Table 3] show that children with ASD tended to have lower mean scores compared to their peers in the DD and POP groups of the same dominance type. For example, expressive dominant children with ASD had an average ELAE score of 42.4 (95% CI 40.9, 43.9), whereas expressive dominant children in the DD and POP groups had average ELAE scores of 53.2 (95% CI 51.7, 54.8) and 58.2 (95% CI 56.5, 59.9), respectively. Similarly, receptive dominant children with ASD had an average RLAE score of 45.1 (95% CI 43.9, 46.4), and receptive dominant children in the DD and POP groups had average RLAE scores of 50.5 (95% CI 49.5, 51.5) and 54.1 (95% CI 52.7, 55.5), respectively. Specifically looking at discrepancies across the language dominance patterns, the largest differences between children with ASD and their peers with DD or in the POP group were seen in comparisons for the ED pattern. In particular, for expressive dominant children, the average ELAE score for children with DD was 10.8 (95% CI 7.6, 14.0) units higher than that of children with ASD; the average ELAE score for POP children was 15.8 (95% CI 12.3, 19.3) units higher than that of children with ASD.

Figure 2 displays the pattern of the expected ELAE and RLAE scores by case status and language dominance. For the group with ASD, the difference in ELAE scores for children with ED versus RD profiles was significantly smaller ($p < 0.01$) than the corresponding difference for the DD and POP groups; the difference for children with ASD was 7.6 (95% CI 5.0, 10.2), for children with DD was 13.6 (95% CI 10.8, 16.5), and for children in the POP group was 15.6 (95% CI 12.3, 18.9). A contrasting pattern was seen when comparing the RLAE scores of children with ED versus RD profiles in each of the case groups, where the difference for children with ASD was significantly larger (8.7) (95% CI 6.1, 11.2)

Table 2 Characteristics of study participants by case status

	Case status[a]				
	Overall (N = 2571)	ASD (N = 695)	DD (N = 987)	POP (N = 889)	*p* value**
Demographic characteristics					
Child CA (mo) at clinic visit	59.4 ± 7.1	59.3 ± 6.7	59.5 ± 7.2	59.4 ± 7.4	0.790
Male	1699 (66%)	570 (82%)	658 (67%)	471 (53%)	< 0.001
Maternal age (year)	31.8 ± 5.6	31.6 ± 5.6	31.7 ± 5.8	32.1 ± 5.4	0.19
Maternal education					< 0.001
8th grade or less	39 (2%)	11 (2%)	24 (2%)	4 (0%)	
9th–12th grade	113 (4%)	27 (4%)	56 (6%)	30 (3%)	
High school	395 (15%)	119 (17%)	175 (18%)	101 (11%)	
Some college, no degree	517 (20%)	159 (23%)	196 (20%)	162 (18%)	
Bachelor's degree	672 (26%)	174 (25%)	249 (25%)	249 (28%)	
Advanced degree	790 (31%)	192 (28%)	269 (27%)	329 (37%)	
Unknown[b]	45 (2%)	13 (2%)	18 (2%)	14 (2%)	
Maternal ethnicity					0.088
Not Hispanic	1957 (76%)	521 (75%)	740 (75%)	696 (78%)	
Hispanic	275 (11%)	76 (11%)	122 (12%)	77 (9%)	
Unknown	339 (13%)	98 (14%)	125 (13%)	116 (13%)	
Maternal race					< 0.001
White	1915 (75%)	474 (68%)	729 (74%)	712 (80%)	
Black, African-American	455 (18%)	148 (21%)	190 (19%)	117 (13%)	
American Indian	12 (0%)	7 (1%)	4 (0%)	1 (0%)	
Asian, Pacific Islander	139 (5%)	54 (8%)	44 (4%)	41 (5%)	
Other, Multiracial	34 (1%)	9 (1%)	14 (1%)	11 (1%)	
Unknown[b]	16 (1%)	3 (0%)	6 (1%)	7 (1%)	
Clinical characteristics					
ELAE	49.0 ± 16.6	35.0 ± 16.2	49.7 ± 14.6	59.2 ± 10.0	< 0.001
EL raw score	38.9 ± 10.4	30.1 ± 11.7	39.6 ± 8.7	44.9 ± 4.8	< 0.001
RLAE	51.2 ± 16.5	37.3 ± 17.9	52.4 ± 14.0	60.8 ± 9.0	< 0.001
RL raw score	39.7 ± 9.6	31.5 ± 11.7	40.7 ± 7.5	44.9 ± 4.1	< 0.001
Social affect severity	–	5.5 ± 1.4	–	–	
VRAE	53.7 ± 14.3	43.4 ± 16.9	54.9 ± 12.5	60.4 ± 8.0	< 0.001
VRAE:CA ratio	0.9 ± 0.2	0.7 ± 0.3	0.9 ± 0.2	1.0 ± 0.1	< 0.001
RLAE/ELAE ratio	1.1 ± 0.3	1.1 ± 0.5	1.1 ± 0.2	1.0 ± 0.1	<0.001

***p* value comparisons across case status for categorical variables are based on Chi square test of homogeneity; *p* values for continuous variables are based on ANOVA

[a]Values expressed as N (%) or mean ± standard deviation

[b]*p* value calculation does not include Unknown category

than the difference observed for those in the DD (5.5) (95% CI 2.8, 8.2) or POP (5.6) (95% CI 2.4, 8.7) groups, with *ps* of 0.01 and 0.02 comparing ASD to DD and ASD to POP, respectively.

The odds of a child with ASD showing a pattern of ED or RD, rather than ND, were much higher than for children in the DD or POP groups (Table 4). For example, the odds of a child with ASD showing a pattern of ED, rather than ND, was 3.6 times (95% CI 2.7, 4.8) the odds for a child in the POP group. Similarly, the odds of a child with ASD showing a pattern of RD, rather than ND, was 3.1 times (95% CI 2.4, 4.0) the odds for a child in the POP group. The odds of ED versus ND were similar for children in the DD and POP groups (1.3) (95% CI 0.9, 1.7), but children with DD were slightly more likely to be RD (versus ND) than POP children (1.9) (95% CI 1.5, 2.5).

We also analyzed odds ratios adjusted for sex, maternal age, and maternal education (Table 4). Differences between the unadjusted and the adjusted odds ratios were relatively small (with overlapping 95% confidence intervals), and the pattern of the associations between language dominance and case status remained the same.

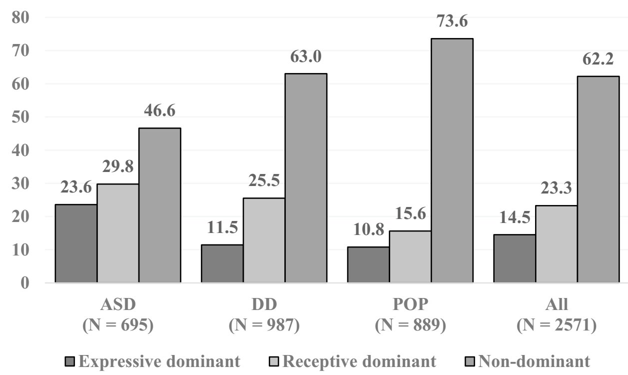

Fig. 1 Frequency distribution of language dominance by case status

Social Affect Severity Among Children with ASD

We assessed whether ASD social affect (SA) severity interacted with child CA at clinic visit, child sex, VRAE:CA ratio, maternal age, or maternal education to influence the ELAE and RLAE scores (EL/RL difference). Table 5 presents the model selection statistics and factors associated with the EL/RL differences among children with ASD. While SA severity, child age, and VRAE:CA ratio were independently associated with EL/RL difference, we found no evidence of interaction among these factors. Additionally, the covariates: maternal education, maternal age, and child sex were retained in the model although they were not significantly associated with EL/RL difference. Estimates of the associations between the factors retained in the final model and the EL/RL difference for children with ASD are displayed in Table 6 along with model diagnostic statistics. The expected EL/RL AE difference for females was 1.16 months (95% CI − 0.20, 2.52) greater than that for males, adjusting for the other variables in the model. Additionally, holding all other variables constant, increases in SA severity increased the expected EL/RL difference; specifically, a one-unit increase in SA severity increased the expected EL/RL difference by 0.53 months (95% CI 0.18, 0.89). Conversely, a one month increase in child CA reduced the expected EL/RL AE difference by 0.17 months (95% CI − 0.24, − 0.09), and a 0.10 increase in the VRAE:CA ratio reduced the expected EL/RL difference by 0.69 months (95% CI − 0.88, − 0.51).

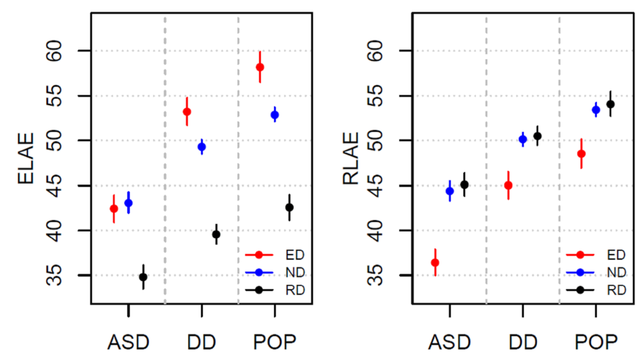

Fig. 2 Adjusted means (95% confidence intervals) of expressive (left) and receptive (right) language age equivalent scores by case status and language dominance. Values are adjusted by sex, final case status, child age (mo) at clinic visit, language dominance status, VRAE:CA ratio, maternal education, maternal age (yr), final case status by language dominance interaction, and sex by final case status interaction

Table 3 Adjusted means of receptive and expressive language raw scores and age equivalent scores by case status and language dominance (N = 2523)

	ASD	DD	POP
Expressive dominant	N = 159	N = 110	N = 92
Receptive raw score	30.13 (29.30, 30.97)	37.08 (36.22, 37.93)	39.12 (38.20, 40.04)
Receptive AE	36.45 (35.00, 37.90)	45.03 (43.54, 46.51)	48.54 (46.94, 50.14)
Expressive raw score	35.02 (34.09, 35.95)	41.51 (40.55, 42.47)	43.53 (42.50, 44.56)
Expressive AE	42.42 (40.91, 43.93)	53.23 (51.68, 54.78)	58.21 (56.54, 59.87)
Receptive dominant	N = 205	N = 248	N = 136
Receptive raw score	36.24 (35.52, 36.96)	39.52 (38.93, 40.11)	41.05 (40.26, 41.83)
Receptive AE	45.11 (43.85, 46.37)	50.51 (49.49, 51.54)	54.09 (52.73, 55.45)
Expressive raw score	29.95 (29.14, 30.75)	33.72 (33.06, 34.38)	36.03 (35.16, 36.91)
Expressive AE	34.82 (33.51, 36.12)	39.59 (38.52, 40.66)	42.57 (41.15, 43.98)
Non-dominant	N = 316	N = 610	N = 647
Receptive raw score	36.33 (35.71, 36.95)	39.31 (38.88, 39.74)	40.48 (40.04, 40.92)
Receptive AE	44.39 (43.31, 45.47)	50.12 (49.38, 50.87)	53.46 (52.70, 54.22)
Expressive raw score	35.67 (34.98, 36.37)	39.17 (38.69, 39.65)	40.49 (40.00, 40.98)
Expressive AE	43.11 (41.99, 44.24)	49.28 (48.51, 50.06)	52.90 (52.11, 53.70)

All values represent adjusted mean (lower confidence limit, upper confidence limit). Values are adjusted by sex, final case status, child age (mo) at clinic visit, language dominance status, VRAE:CA ratio, maternal education, maternal age (year), final case status by language dominance interaction, and sex by final case status interaction

Table 4 Unadjusted and adjusted odds ratio estimates and 95% confidence intervals for language dominance by case status (N = 2524)

	Language dominance comparisons		
	ED vs ND	RD vs ND	ED vs RD
ASD vs POP	3.56 (2.67, 4.76)	3.09 (2.39, 3.98)	1.15 (0.82, 1.61)
	3.52 (2.60, 4.77)[a]	2.91 (2.23, 3.79)[a]	1.21 (0.85, 1.72)[a]
ASD vs DD	2.81 (2.13, 3.71)	1.60 (1.27, 2.01)	1.76 (1.30, 2.39)
	2.92 (2.20, 3.88)[a]	1.60 (1.27, 2.02)[a]	1.82 (1.34, 2.48)[a]
DD vs POP	1.27 (0.94, 1.71)	1.93 (1.53, 2.45)	0.66 (0.46, 0.93)
	1.21 (0.89, 1.63)[a]	1.82 (1.43, 2.31)[a]	0.66 (0.47, 0.95)[a]

ED expressive dominant, *RD* receptive dominant, *ND* non-dominant, *ASD* autism spectrum disorder, *DD* developmental disability, *POP* population

[a]Odds ratios adjusted by sex, maternal age, and maternal education

Figure 3 displays the overall trends in the effect of increases in ASD SA severity on the expected EL/RL difference for varying child CAs and VRAE:CA ratios, stratified by sex (maternal age of 32 years and advanced degree held constant). ELAE exceeded RLAE in younger children and children with a lower VRAE:CA ratio. For example, a 36-month old female with a VRAE:CA ratio of 1, and SA severity of 6 had an expected EL/RL difference of 0.58 (95% CI − 1.89, 3.05) while at 60 months the expected EL/RL difference for the same child was − 3.39 (95% CI − 4.96, − 1.82). In contrast, children at or above the median age (60.5 months) and the median VRAE:CA ratio (0.76) had a higher expected RLAE score than ELAE score at all levels of SA severity. Thus, children who were younger, who were more impaired in nonverbal cognitive skills relative to their CA, and who had higher levels of social affect severity were more likely to have expressive language scores that exceeded their receptive language scores.

Discussion

In this study, we undertook analyses of expressive and receptive language measures in young children with ASD, other DD, and in a population sample with the goal of better understanding previous discrepant findings regarding whether receptive language skills are generally lower than expressive language skills in young children with ASD, and the extent to which this pattern may be more common among children with ASD than children in other groups. With the exception of Seol et al. (2014) and the current study, most studies related to discrepancies between receptive and expressive language scores in children with ASD have analyzed differences in scores at the group level. The current work characterized the expressive and receptive language ability within an individual child, i.e., focused on individual differences, which is a key consideration for interpreting our findings.

Our outcomes provide insights into the discrepant findings across previous studies related to the occurrence of an ED profile among children with ASD versus children without ASD. A commonality across the three groups in our study was that the ED profile was observed in the smallest proportion of children. Thus, the ED profile was not characteristic of our sample of children with ASD as a whole, aligning with some previous studies that did not find children with ASD to show an "expressive advantage" at a group level (Jarrold et al. 1997; Kjelgaard and Tager-Flusberg 2001; Loucas et al. 2008). Nevertheless, children with ASD in our sample were significantly more likely to show an ED profile than those in the DD and POP groups, which may explain why other previous studies reported higher mean

Table 5 Linear regression model selection statistics for factors associated with the EL/RL differences among children with ASD

	DF	Full		Reduced				Final	
		Model 1		Model 2		Model 3		Model 4	
		$R^2 = 0.1174$		$R^2 = 0.1174$		$R^2 = 0.1171$		$R^2 = 0.1114$	
		F value	*p* value	F value	*p* value	F value	*p* value	F value	*p* value
SA severity	1	0.32	0.572	6.80	0.009	7.06	0.008	8.58	0.004
Maternal education	5	0.81	0.545	0.81	0.545	0.81	0.543	0.54	0.743
Child age	1	0.83	0.363	15.69	< 0.01	15.51	< 0.01	17.30	< 0.01
Child sex	1	0.01	0.937	0.00	0.945	2.66	0.103	2.82	0.094
Maternal age	1	1.67	0.197	1.66	0.198	1.70	0.192	1.32	0.251
VRAE:CA ratio	1	52.05	< 0.01	52.12	< 0.01	52.33	< 0.01	52.71	< 0.01
SA severity × education	5	0.85	0.518	0.85	0.517	0.85	0.512	–	–
SA severity × sex	1	0.27	0.606	0.26	0.610	–	–	–	–
SA severity × child age	1	0.01	0.934	–	–	–	–	–	–

Table 6 Linear regression model estimates and diagnostic statistics for factors associated with the EL/RL differences among children with ASD

	Estimate	SE	t value	*p* value	Correlation (90% CI)[a]	Tolerance	VIF
SA Severity	0.53	0.182	2.93	0.004	0.010 (0.002, 0.028)	0.97	1.03
Maternal education[b]					– 0.003 (0.000, 0.006)		
8th grade or less	1.47	2.123	0.69	0.488		0.94	1.06
9th-12th grade	1.50	1.452	1.04	0.300		0.84	1.19
High school	1.05	0.853	1.23	0.219		0.65	1.55
Some college, no degree	0.42	0.747	0.56	0.575		0.68	1.48
Bachelor's degree	– 0.01	0.714	– 0.01	0.990		0.69	1.44
Child age	– 0.17	0.040	– 4.16	< 0.01	0.022 (0.008, 0.045)	0.97	1.03
Sex[c]					0.002 (0.000, 0.015)		
Female	1.16	0.691	1.68	0.094		0.96	1.04
VRAE:CA ratio	– 6.93	0.954	– 7.26	< 0.01	0.069 (0.042, 0.103)	0.94	1.07
Maternal age	0.06	0.051	1.15	0.251	0.000 (0.000, 0.011)	0.83	1.20

VIF variance inflation factor

[a]Denotes the semipartial correlation for each variable and conservative 90% confidence limits

[b]Advanced degree is the reference group

[c]Males are the reference group

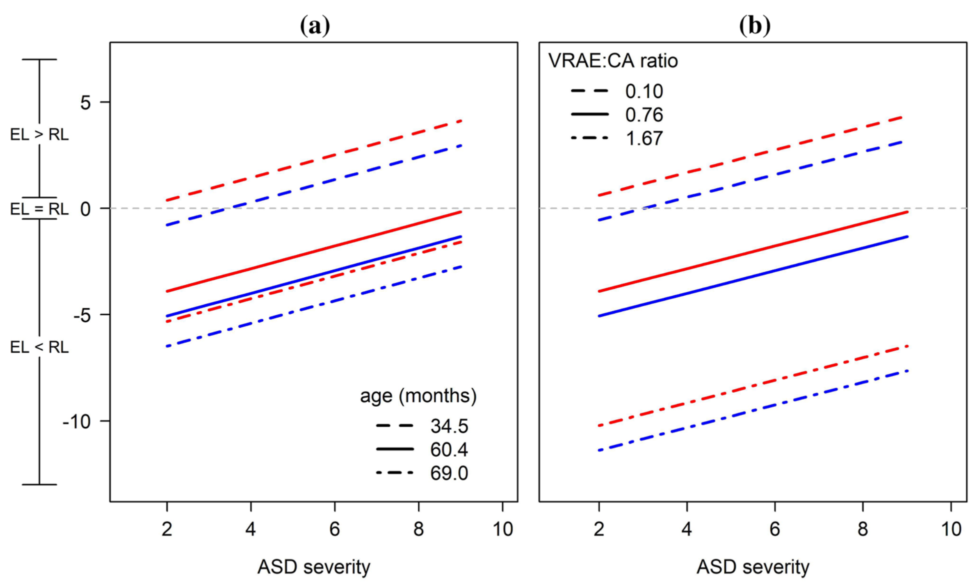

Fig. 3 EL/RL difference as a function of ASD SA severity for mothers of median age (32 years) and with an advanced degree for **a** changes in child age with median VRAE:CA ratio and **b** changes in VRAE:CA ratio with median child age, stratified by female (red lines) and male (blue lines) sex

expressive than receptive scores in their samples (Charman et al. 2003; Hudry et al. 2010; Kjelgaard and Tager-Flusberg 2001; Kover et al. 2013; Luyster et al. 2008; Maljaars et al. 2012; Volden et al. 2011; Ellis Weismer et al. 2010).

The relatively high occurrence of the RD profile we observed among children with ASD has received less attention in previous literature, and also may help to explain discrepant findings in some previous studies. That is, if means for receptive and expressive language scores are examined across a sample of children that includes both children with ED profiles and children with RD profiles, the opposite direction of differences in the two profiles will tend to cancel one another out, thereby obscuring the extent to which children with ASD show "uneven" language profiles of either type. For our sample, an uneven profile, either RD or ED, occurred in 53% of children with ASD, but in only 37% of children with DD and 26% of children in the POP group.

In comparing our three groups of children (ASD, DD and POP) based on children's language profiles (ED, RD or ND), we found that the children with ASD consistently showed lower expressive and receptive language scores than children in either of the other groups with the same language profile. This perhaps seems unremarkable given that children in the ASD group also had lower VRAE:CA ratios than children in the other two groups, suggesting they may have more impaired nonverbal intellectual functioning as well as more impaired language. However, we highlight the contribution that variable language dominance profiles can make when comparing children's language skills across groups. When comparisons were made across both groups and profiles, we found some comparisons for which children with ASD did not differ significantly from children in the DD or the POP groups with different language profiles. For example, ED children with ASD did not differ significantly from RD children in the DD or POP groups on their expressive language scores, and RD children with ASD did not differ significantly in receptive language scores from ED children in the DD or POP groups.

On the other hand, it is striking (see Fig. 2) that children in the ASD group with ED profiles have relatively more marked impairments in receptive language than do children with ED profiles in the other two groups. Potentially, expressive language is more impacted by factors that are not specific to ASD, such as general cognitive functioning, motor speech integrity, and quality of the language environment, whereas receptive language may be more specifically impacted by social-communication impairments associated with ASD, particularly for those children with more severe impairments in their ability to respond to joint attention bids. This would limit their ability to benefit from language learning opportunities even within a rich language environment.

Through this study, we contribute findings that may help to explain some of the inconsistencies in the existing literature. The sample size in this study is an advantage in that it provided adequate power for the multivariate linear regression model employed. The findings suggest a coherent explanation for expressive-receptive discrepancies among children with ASD. That is, younger age, lower nonverbal cognitive skills, and more severe social-affect symptoms of ASD contribute to an unusual language learning profile, and variability in any of the factors will tend to either strengthen or attenuate expressive-receptive language discrepancies. In his seminal report, Kanner (1943) described a tendency of children with ASD to be scripted learners of expressive language; that is, they often acquire a new word or phrase within a specific context and then repeat what they heard when triggered by a similarity in a future context, without having acquired the same level of understanding that typically underlies children's expressive language. Further, children with ASD at lower cognitive mental ages are more hyporesponsive to both social and nonsocial stimuli in their environment than children with other DD, (Baranek et al. 2013), decreasing their opportunities to develop a strong foundation of language comprehension, as well as limiting their progress in expressive language. Children who are younger, at lower cognitive levels, and with more severe autism symptoms in the Social Affect domain have more challenges with specific key skills such as response to joint attention (e.g., Schietecatte et al. 2012) and directing communication to others, which predict growth in both receptive and expressive language skills for young, initially nonverbal/low verbal children with ASD (Yoder et al. 2015). Our findings combined with those of other studies (e.g., Paul et al. 2008; Woynaroski et al. 2016) suggest that the ED profile in preschool children with ASD may be associated with a cluster of factors that are particularly disadvantageous to longitudinal progress in language learning. Additionally, these findings may help explain some of the discrepancies of outcomes in prior studies of EL and RL discrepancies. For example, Seol et al. (2014), who found a high occurrence of the ED profile in children with ASD, studied children with a mean age under three years. Consistent with our findings, however, they also found that the percentage of children with an ED profile diminished with age.

Language profiles that involve marked discrepancies between expressive and receptive language among young children with disabilities have implications for clinical practice. Focusing more specifically on young children with ASD, our data as well as the data of others indicates the high likelihood that these children will have an uneven language profile, either ED or RD, and these profiles are associated with especially severe delays, on average, in the more impacted language mode, i.e., either expressive or receptive language, for children with ASD. The clinical implications of each profile warrant consideration.

Expressive language deficits likely are quite salient to a child's interaction partners. For example, Warlaumont et al. (2014) presented evidence from children with and without ASD to support the role of a proposed social feedback loop in language development. They found that adults were more likely to respond when a child's vocalization was speech related, and a child's subsequent vocalization was more likely to be speech-related if their previous vocalization received an immediate adult response. Predictably, young children with ASD initiated fewer speech-related vocalizations than typically developing children; in addition, adult responses to the vocalizations of children with ASD were not as strongly contingent on whether the vocalizations were speech-related or not. These findings imply that a child's expressive language plays a role in eliciting responses from others in the environment that provide context-relevant, language-learning opportunities to the child. Potentially, a child who has weak expressive language relative to receptive language may fail to elicit language responses that optimally stimulate growth in his or her stronger modality of receptive language, unless the discrepancy is understood and considered in designing an individualized intervention.

Designing appropriate individualized intervention for children with ASD when comprehension skills require prioritizing may prove to be especially challenging. A recent study speaks indirectly to this point (Goodwin et al. 2015). The researchers found that even when children with ASD are closely matched to typically developing children on language developmental levels, the communicative transactions between adults and children may have different impacts on the comprehension development of children in the two groups. Although the measured characteristics of maternal input were very similar in the two groups, the predictors of child comprehension outcomes differed, suggesting that children with ASD and those who are typically developing process input differently. Particularly of concern is that a number of features of maternal input positively predicted increased comprehension across time in children with typical development, whereas with one exception, the features of input that were significantly associated with comprehension outcomes in children with ASD were negative predictors.

The limitations of the current study include the relatively limited chronological age range of the participants (30 to 68 months), with the mean age of 59 months being older than most previous studies reporting expressive scores exceeding receptive scores in children with ASD. Despite the limited age range, we found evidence that CA is one factor significantly associated with differences in expressive and receptive language scores.

Another potential limitation of this study is the varied distribution of race among the study groups. There are more White mothers in the POP group than in the ASD or DD groups. While this may reflect differential response by race, SEED had limited information available about the characteristics of the recruitment pools; thus, our ability to assess potential biases by race is limited. However, we do not have cause to believe that the results of the specific questions addressed in this analysis would differ by race.

Finally, we acknowledge the limitations of using AE scores as an index of comparison between receptive and expressive language scores on the MSEL. This issue is encountered frequently in studies of individuals with ASD, with many participants attaining the minimum standardized score on tests (e.g., MSEL), leading to floor effects that curtail variability in the sample (Ellis Weismer et al. 2010; Hudry et al. 2010; Luyster et al. 2008). Specific to the MSEL, the test manual does not provide a means to interpret scores that fall below the floor. Hudry et al. (2010) support the use of AE in such research, noting that in ASD samples, use of AE is often chosen over a more psychometrically-robust index such as a standard score because the data produced will be more interpretable and less subject to Type II errors.

This study and previous related investigations suggest many avenues for future research. For example, the variables associated with an ED profile suggest a particularly poor prognosis for later language outcomes; however, the current literature on expressive-receptive language discrepancies in children with ASD includes only a few longitudinal studies (e.g., Davidson and Ellis Weismer 2017; Woynaroski et al. 2016), and these have not focused explicitly on prediction of language outcomes based on language dominance profile nor on expressive-receptive discrepancies as a continuous variable. Relatedly, it is not clear whether expressive-language discrepancies would account for unique variance in language outcomes beyond the factors associated with a greater expressive advantage/receptive disadvantage (age, nonverbal cognitive level, social affect symptom severity). Additional previously unexamined factors may also contribute to the tendency of a child to demonstrate a specific profile; for example, attention deficits, motor delays and sensory defensiveness may all impact a child's performance on receptive language tasks such as those on the MSEL. Research to examine language profiles as potential moderators of treatment outcomes could provide empirical guidance to researchers and clinicians who seek to improve language outcomes for children with ASD.

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