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ORIGINAL PAPER

Examination of Clinical and Assessment Type Differences Between Toddlers with ASD from Multiplex and Simplex Families

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Abstract

Few studies have examined differences in autism spectrum disorder (ASD) phenotype between children from multiplex and simplex families at the time of diagnosis. The present study used an age- and gender-matched, community-based sample (n = 105) from the southwestern United States to examine differences in ASD symptom severity, cognitive development, and adaptive functioning. No significant differences between children from multiplex and simplex families were observed. Exploratory analysis revealed that parents underreported receptive and expressive language and fine motor skills compared to professional observation, especially among children from multiplex families. These findings suggest that diagnosticians may need to consider family structure when choosing and interpreting assessments of receptive language, expressive language, and fine motor skills.

Autism Spectrum Disorder (ASD) is identified in 1-in-44 children by the age of 8 years (Maenner et al., 2021). With such high prevalence rates, early identification of ASD is important for increasing the likelihood of positive long-term outcomes (Clark et al., 2018; Gabbay-Dizdar et al., 2021). As ASD is heterogenous (American Psychiatric Association, 2013; Lord et al., 2018; Masi et al., 2017), investigations of early signs of ASD must include individuals with a variety of presentations. To date, such studies have had challenges with sample size, racial and ethnic diversity, and gender representation (Berends et al., 2019; Dissanayake et al., 2019). Additionally, a significant proportion of research into early signs of ASD is biased toward children from multiplex families (i.e., families with more than one child with ASD) to the exclusion of children from simplex families (i.e., families with only one child with ASD) due to reliance on infant sibling designs (Szatmari et al., 2016). Additional research is needed to understand the full spectrum of early signs of ASD.

Results from studies that compared presentations of children from multiplex and simplex families contribute to our understanding of genetic contributions to ASD symptom expression (Arnett et al., 2019; Cuccaro et al., 2003; Nayar et al., 2018). Recently summarized by Dissanayake and colleagues (2019), previous studies in this area have yielded mixed results with some reporting between group differences in autism symptoms or cognitive abilities, and others reporting no differences between groups. Critically, Dissanayake and colleagues (2019) identified methodological limitations in earlier studies, including wide age ranges, a combination of children with and without siblings in simplex groups, and/or reliance on clinically referred samples. Two recent studies addressed some or all of these methodological limitations (Berends et al., 2019; Dissanayake et al., 2019). Neither study reported statistically significant between group differences in autism symptom severity using the Autism Diagnostic Observation Schedule (Lord et al., 2000, 2012), although there was some evidence of elevated developmental quotients (DQ) for children with ASD in multiplex families (Berends et al., 2019; Dissanayake et

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al., 2019). Parent report scales measuring adaptive functioning (i.e., age-appropriate skills necessary for independent living) were only included in one of the studies, which found no difference among groups (Berends et al., 2019).

Interestingly, only one published study has utilized a sample derived from the general population at the time of diagnosis (Dissanayake et al., 2019). Samples in other previous studies were derived from clinical or treatment settings, and therefore participants may have had a more severe symptom presentation than a sample drawn from the general population. Participants in these studies are already at heightened risk for ASD or are otherwise seeking support for ASD-related challenges, which could potentially result in the samples being skewed by cases with more severe ASD symptoms. Community-based samples identified from the general population may include participants with milder ASD symptoms who need less support, which may be more generalizable to the overall population.

Further, existing research has not examined whether and how discrepancies between direct professional observation and parent-report measures contribute to the mixed findings discussed above. Discrepancies between parent report and direct observation of ASD core symptoms are reported elsewhere (Chawarska et al., 2007; Taylor et al., 2015), but the nature and direction of discrepancies in specific skill areas remains unexplored. Both the Mullen Scales of Early Learning (Mullen, 1995) and the Vineland Adaptive Behavior Scales – Parent Interview (Sparrow et al., 2005) include receptive language, expressive language, and fine motor skills subscales, which allows for comparison of these key developmental domains. Both scales include age equivalency scores. Thus, it is possible to compare direct professional observation and parent report results.

Despite the progress that has been made in understanding differences between children from multiplex and simplex families, several limitations continue to persist. Many studies in this area have focused on school-aged children and adolescents (Berends et al., 2019; Cuccaro et al., 2003; Oerlemans et al., 2016; Taylor et al., 2015), but only one published study has focused on young children at the time of diagnosis (Dissanayake et al., 2019). This leaves significant gaps in our understanding of ASD presentation in young children from different familial groups.

The Current Study

The primary aim of this study was to examine differences in autism severity, cognitive development, and adaptive functioning between children from multiplex and simplex families in young children at the time of diagnosis. To account for possible environmental effects on development, only second diagnosed siblings were included in the multiplex sample. Children from simplex families were split into groups based on the presence of siblings at the time of evaluation. Consistent with findings from previous research that has compared children from multiplex and simplex families (Berends et al., 2019; Dissanayake et al., 2019), it was hypothesized that children from multiplex families would demonstrate higher cognitive DQs compared to children from simplex families, but autism severity and adaptive functioning scores would not differ significantly.

An exploratory aim was to examine inconsistencies between direct professional observation and parent report by family group (multiplex or simplex). Inconsistencies in parent-report and direct professional observation have been previously identified on measures of ASD core symptoms (Chawarska et al., 2007; Taylor et al., 2015), but have not, to our knowledge, been examined for specific skill areas (i.e., receptive language, expressive language, and fine motor skills) across family groups. To better understand potential differences between children from multiplex and simplex families, it is important to identify whether discrepancies between direct professional assessment and parent report follow similar patterns across groups. Further, many aspects of ASD diagnostic practices rely on parent report. It is therefore important to document the accuracy of parents' perception of their child's skills compared to direct professional observation of receptive and expressive language, and fine motor development.

Methods

Participants

Participants were drawn from a larger study (Smith et al., 2022) that investigated the effects of the Get *SET* Early Model, which was designed to identify autism and developmental disorders in toddlers through universal developmental screening in the general population at 12-, 18-, and 24-month well-baby visits (Pierce et al., 2021). Depicted in Fig. 1, 648 children were evaluated for ASD between 2015 and 2018 as a part of this study, and 308 of these children were diagnosed with ASD. From these children, three groups were formed for the current analyses: (1) Multiplex: children with a sibling previously diagnosed with ASD; (2) Simplex1: children with no other siblings; and (3) Simplex2: children with siblings who were believed to be typically developing at the time of the evaluation. Each group included 35 children who were matched on age and gender across groups for a total sample of 105 participants (see Fig. 1; Table 1). Children's ages ranged from 12 to 37 months (M = 22.32 months of age, SD = 7.32). The sample

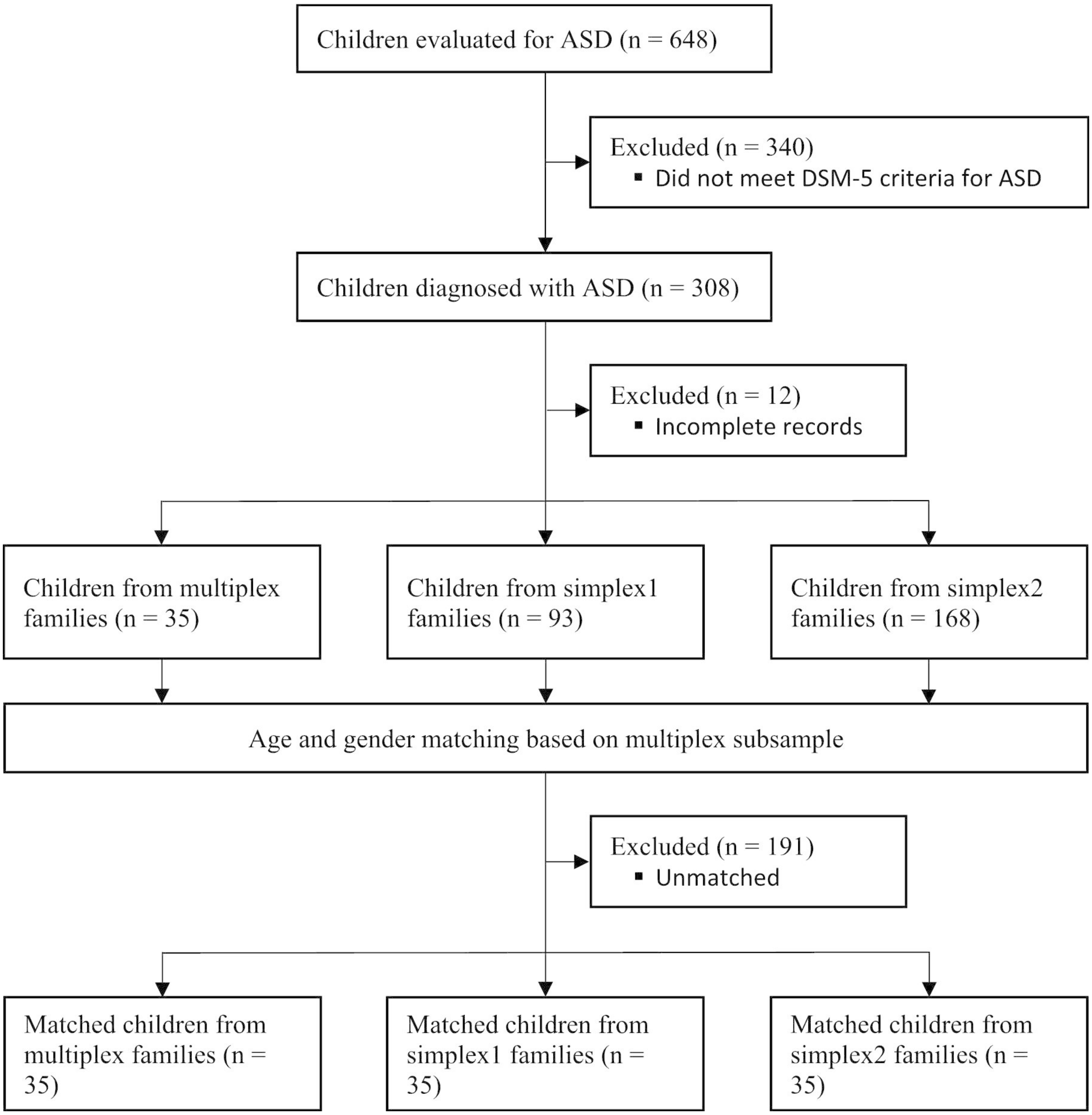

Fig. 1 Flowchart of study participants

was mostly reflective of the racial and ethnic makeup of the community based in the southwestern United States (U.S. Census Bureau, 2022), although non-Hispanic African American children were underrepresented ($n=2$, 1.90%). Children who were non-Hispanic Whites made up the largest share of the sample ($n=52$, 49.52%) with Hispanic children being the second largest group ($n=33$, 31.42%).

Procedures

As part of the larger study, which was approved by the Institutional Review Board of the University of California, San Diego, all participants were evaluated by a licensed clinical psychologist who performed a gold standard developmental evaluation for ASD. Assessments included the Autism Diagnostic Observation Schedule – 2nd Edition (ADOS-2; Lord et al., 2012), Mullen Scales of Early Learning – AGS Edition (MSEL; Mullen, 1995), and the Vineland Adaptive Behavior Scales – Parent Interview, Second Edition (VABS; Sparrow et al., 2005). Informed consent was obtained from at least one parent/guardian of each child.

Table 1 Sample demographics

	Multiplex		Simplex-1		Simplex-2		Significance
	n	%	n	%	n	%	*p*
Matched Total (n = 105)	35	-	35	-	35	-	
Race/Ethnicity							
White, not Hispanic	16	45.71	20	57.14	16	45.71	$p = 0.70$ǂ
Black, not Hispanic	0	0.00	1	2.86	1	2.86	
Asian, not Hispanic	4	11.43	1	2.86	1	2.86	
Hispanic	10	28.57	10	28.57	13	37.14	
Other, not Hispanic	5	14.29	3	8.57	4	11.43	
Missing	-	-	-	-	-	-	
Gender							
Male	28	80.00	28	80.00	28	80.00	
Female	7	20.00	7	20.00	7	20.00	
Income							
<$20,000	5	14.29	3	8.57	3	8.57	$p = 0.30$ǂ
$20,001 - $40,000	3	8.57	7	20.00	4	11.43	
$40,001 - $60,000	2	5.71	7	20.00	5	14.29	
$60,001 - $80,000	4	11.43	1	2.86	4	11.43	
$80,001 - $100,000	5	14.29	1	2.86	5	14.29	
>$100,000	6	17.14	10	28.57	10	28.57	
Missing	10	28.57	6	17.14	4	11.42	
	Mean	**Std. Dev.**	**Mean**	**Std. Dev.**	**Mean**	**Std. Dev.**	
Mean Age (in months)	22.34	7.68	22.06	6.73	22.57	7.71	$p = 0.98$†
Mean Number of Siblings	2.06	1.21	0	0	1.63	0.97	$p = 0.84$‡
Mean Number of Siblings without ASD	0.97	1.25	0	0	1.63	0.97	$p = 0.001$‡
Mean Number of Siblings with ASD	1.09	0.28	0	0	0	0	-

[?]Significance determined by chi square (?^{2}) or Fisher?s exact analysis. For race, White participants were compared to a collapsed category that included all non-White participants. For income, three categories were compared: participants reporting ? $60,000, participants reporting ? $60,000, and participants with missing data

[?]Significance determined by Kruskal-Wallis test

[?]Significance determined by Dunn?s test between the Multiplex Group and Simplex2 group after Bonferroni correction.

Measures

Autism Diagnostic Observation Schedule – 2nd Edition (ADOS-2)

The ADOS-2 (Lord et al., 2012) is a standardized assessment tool used to assess autism symptoms in children and adults. For this study, all participants were administered either the Toddler Module, Module 1, or Module 2 by a research reliable rater. Modules 1 and 2 of the ADOS-2 include a calibrated severity score to enable comparison between the modules (Lord et al., 2012). Previous research has documented how to calculate calibrated severity scores to enable comparison between the Toddler Module and other ADOS-2 modules (Esler et al., 2015; Janvier et al., 2021). Calibrated severity scores range from 1 to 10, with higher scores indicating higher concern (Toddler Module) or higher level of autism symptoms (Modules 1 and 2).

Mullen Scales of Early Learning (MSEL)

The MSEL (Mullen, 1995) is a standardized measure of verbal and nonverbal development among children up to 68 months of age, which assesses four domains: visual reception, fine motor skills, receptive language, and expressive language. Raw scores are generated and converted to age equivalency scores, which are used to calculate a verbal, nonverbal, and overall DQ (Stephens et al., 2018). The MSEL has a long history of use among children at risk of ASD, with its validity and reliability having been documented elsewhere (Akshoomoff, 2006; Swineford et al., 2015).

Vineland Adaptive Behavior Scales – Parent Interview, 2nd Edition (VABS)

The VABS (Sparrow et al., 2005) is a measure of adaptive behavior that can be used with individuals between birth and 90 years of age. The VABS yields an adaptive behavior composite score and several domain scores, including: communication skills, daily living skills, socialization skills, and motor skills. From raw scores in these domains, age equivalency scores can be calculated, including for receptive language, expressive language, and fine motor skills. Reliability and validity of the VABS has been reported as strong, with α coefficients ranging from 0.86 to 0.98 depending on the domain being assessed and the age of the participant.

Data Analysis

All analyses were conducted using R statistical software version 4.2.1. To address the primary aim, differences among the multiplex, simplex1, and simplex2 groups on ADOS-2 calibrated severity scores and the MSEL overall DQ were assessed with Kruskal-Wallis tests. The MSEL nonverbal and verbal DQs and VABS domains (communication, daily living skills, socialization, and motor skills) were assessed with MANOVAs using Wilks' lambda. Post-hoc tests were conducted as appropriate.

Using age equivalency scores, the effect of assessment method (direct professional observation vs. parent report) was examined with three (receptive language, expressive language, and fine motor skills) separate mixed ANOVAs (3 study group x 2 measure: MSEL, VABS) and pairwise comparisons.

Results

Aim 1: ASD Symptom Severity, Cognition, and Adaptive Functioning

There was no statistically significant group effect on ADOS calibrated severity scores (χ^2 (2) = 1.04, p = 0.59; see Fig. 2). Cohen's d indicated a small effect size for lower severity scores in the multiplex group compared to the simplex1 group (see Table 2).

There were no statistically significant group effects on overall DQ (χ^2 (2) = 3.80, p = 0.15), and nonverbal and verbal DQs (F(4, 196) = 1.14, λ = 0.96, p = 0.34; see Fig. 3) from the MSEL. For all DQs, Cohen's d indicated small effect sizes for higher scores in the multiplex group (see Table 2).

There was no statistically significant group effect on adaptive functioning (F(8, 198) = 0.94, λ = 0.80, p = 0.60; see Fig. 4) from the VABS but Cohen's d suggested some small and moderate effect sizes for the simplex groups having higher scores than the multiplex group (see Table 2).

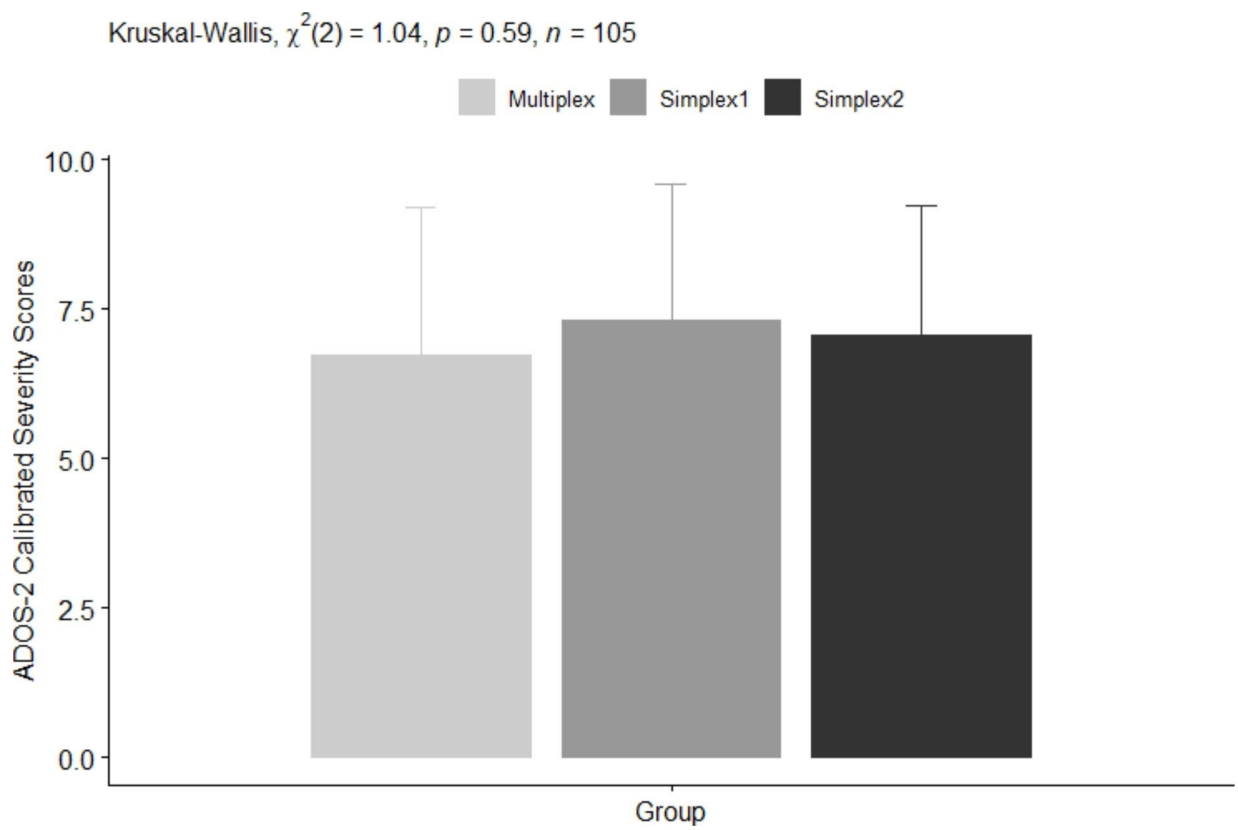

Fig. 2 Kruskal-Wallis Results comparing mean ADOS calibrated severity scores between age and gender matched multiplex children, simplex-1 children, and simplex-2 children

Table 2 Effect size of differences between multiplex and simplex groups in the ADOS, MSEL, and VABS

Evaluation by Grouping	Effect Size (Cohen's d)	Magnitude
ADOS-2 Calibrated Severity Scales		
Multiplex-Simplex1	-0.25	Small
Multiplex-Simplex2	-0.15	Negligible
Simplex1-Simplex2	0.12	Negligible
MSEL Nonverbal Developmental Quotient		
Multiplex-Simplex1	0.36	Small
Multiplex-Simplex2	0.39	Small
Simplex1-Simplex2	0.06	Negligible
MSEL Verbal Developmental Quotient		
Multiplex-Simplex1	0.37	Small
Multiplex-Simplex2	0.23	Small
Simplex1-Simplex2	-0.19	Negligible
MSEL Overall Developmental Quotient		
Multiplex-Simplex1	0.45	Small
Multiplex-Simplex2	0.43	Small
Simplex1-Simplex2	-0.05	Negligible
VABS Communication Skills		
Multiplex-Simplex1	-0.22	Small
Multiplex-Simplex2	0.031	Negligible
Simplex1-Simplex2	0.25	Small
VABS Daily Living Skills		
Multiplex-Simplex1	-0.32	Small
Multiplex-Simplex2	-0.21	Small
Simplex1-Simplex2	0.13	Negligible
VABS Socialization Skills		
Multiplex-Simplex1	-0.27	Small
Multiplex-Simplex2	-0.22	Small
Simplex1-Simplex2	0.01	Negligible
VABS Motor Skills		
Multiplex-Simplex1	-0.56	Moderate
Multiplex-Simplex2	-0.35	Small
Simplex1-Simplex2	0.17	Negligible

Aim 2: Exploratory Analyses

There was a significant main effect of measure (MSEL vs. VABS) for receptive language ($F(1, 102) = 25.39$, $p < 0.001$), expressive language ($F(1, 102) = 11.74$, $p < 0.001$), and fine motor skills ($F(1, 101) = 14.31$, $p < 0.001$). For all three skill sets, MSEL scores were higher than VABS scores. For receptive language and fine motor skill scores, significant measure by group (multiplex, simplex1, simplex2) interactions were observed ($F(2, 102) = 3.65$, $p = 0.03$ for receptive language, $F(2, 101) = 5.16$, $p = 0.01$ for fine motor skills; see Figs. 5 and 6, respectively). No significant measure by group differences were observed in expressive language scores ($F(2, 102) = 2.93$, $p = 0.06$; see Fig. 7). For both receptive language and fine motor skills, the multiplex group had significantly higher MSEL scores compared to VABS scores after post-hoc pairwise comparisons with Bonferroni adjustment (ps < 0.001). Similarly, the simplex 2 group also had significantly higher MSEL scores for receptive language ($p = 0.006$). No significant difference in either simplex group was reported for fine motor skills.

Discussion

Leveraging a sample of toddlers with ASD detected via universal screening in the general population (Smith et al., 2022), this study replicates previous reports of no differences in autism symptom severity or adaptive functioning between children from multiplex and simplex families. In contrast to previous research, however, we did not observe significant differences in cognitive abilities between children from different family groups. Exploratory analyses revealed a pattern of lower expressive language, receptive language, and fine motor scores on the VABS (parent report) compared to these same skill areas on the MSEL (professional direct observation). This pattern was most pronounced in children from multiplex families. Unlike previous studies of this topic, an age-and-gender matched sample was used to create balance between the multiplex and simplex groups. This sample was also younger and more ethnically diverse than samples previously studied. The community-based origin and relative ethnic diversity of this sample are strengths as the sample may be more representative of the general ASD population. Consequently, the findings have increased generalizability relative to previous research in this area.

While results from previous research demonstrating higher DQs in multiplex compared to simplex groups (Berends et al., 2019; Dissanayake et al., 2019) were not replicated, effect sizes suggest numerous small differences favoring the multiplex group. It is possible that these small differences, which were not statistically significant, are a precursor to larger differences that could emerge later in life. This would be consistent with cascade theories of development, which propose that effects from experiences are compounded over time (Cox et al., 2010; Masten & Cicchetti, 2010). Therefore, as a group of individuals with differing experiences get older, developmental differences become more pronounced. The average age of participants in the present sample is four months younger than previously studied. Thus, inconsistencies between current findings and previous studies may indicate a widening gap between children from multiplex and simplex families that is more difficult to detect during the early developmental period. Future longitudinal research should explore whether and how clinically significant differences in cognitive abilities emerge over time between children from different family types.

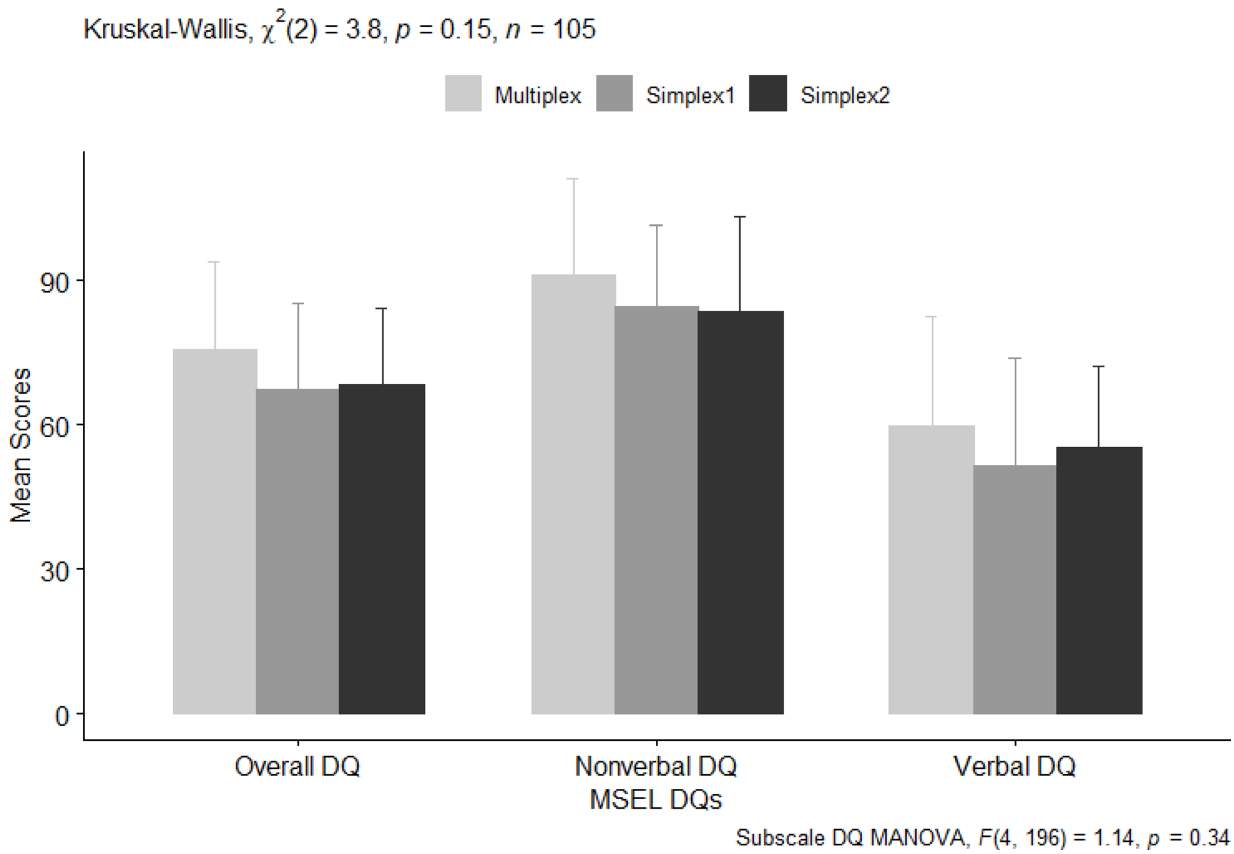

Fig. 3 Kruskal-Wallis and MANOVA Results comparing mean MSEL overall and subscale developmental quotients between age and gender matched multiplex children, simplex-1 children, and simplex-2 children

While interpreting findings from the primary aim, an interesting pattern of discrepancies was observed. Namely, the multiplex group appeared to have slightly better scores than the simplex groups on the MSEL but slightly lower scores on the VABS. Exploratory analyses were conducted to assess whether there was a relationship between family type and measure. For all comparable domains on the two measures (i.e., receptive language, expressive language, and fine motor skills), age equivalency scores from direct professional observation (MSEL) were significantly higher than scores derived from parent report (VABS). Interactions between assessment and family type were detected in the receptive language and fine motor skill subdomains such that discrepancies between MSEL and VABS scores were most pronounced in the multiplex group and least pronounced in the simplex1 group. These findings have several potential interpretations.

The MSEL and VABS include similar items within receptive language, expressive language, and fine motor skills subscales, and previous research suggests convergent validity between the two measures on the communication domain in children with and without ASD (Swineford et al., 2015). Although direct professional observation and parent report are often used together to provide a more comprehensive picture of children's functioning, they do not always agree, which can make clinical interpretation and diagnostic determinations more challenging. Further, both types of measures can be used independently to provide estimates of developmental functioning in these domains. Parent report measures may be relied upon when it is not possible to conduct direct observation of a child's abilities (e.g., child noncompliance; telehealth evaluation). For these reasons, it is important to understand how and why scores differ across measures.

It is possible that discrepancies in age equivalency scores between the MSEL and VABS reflect parents in multiplex and simplex2 families under-reporting their children's skills. Research has shown that parents of children with ASD face significant stress, which can be multiplied when there is more than one child with ASD in a family (Bonis, 2016). In the absence of siblings, parents of only children may be able to better focus attention to their child's functioning, which could allow them to better recall specific skills relative to parents of multiple children. Alternatively, it is possible that parent's thresholds for reporting skills is skewed by the presence of additional children compared to parents of one

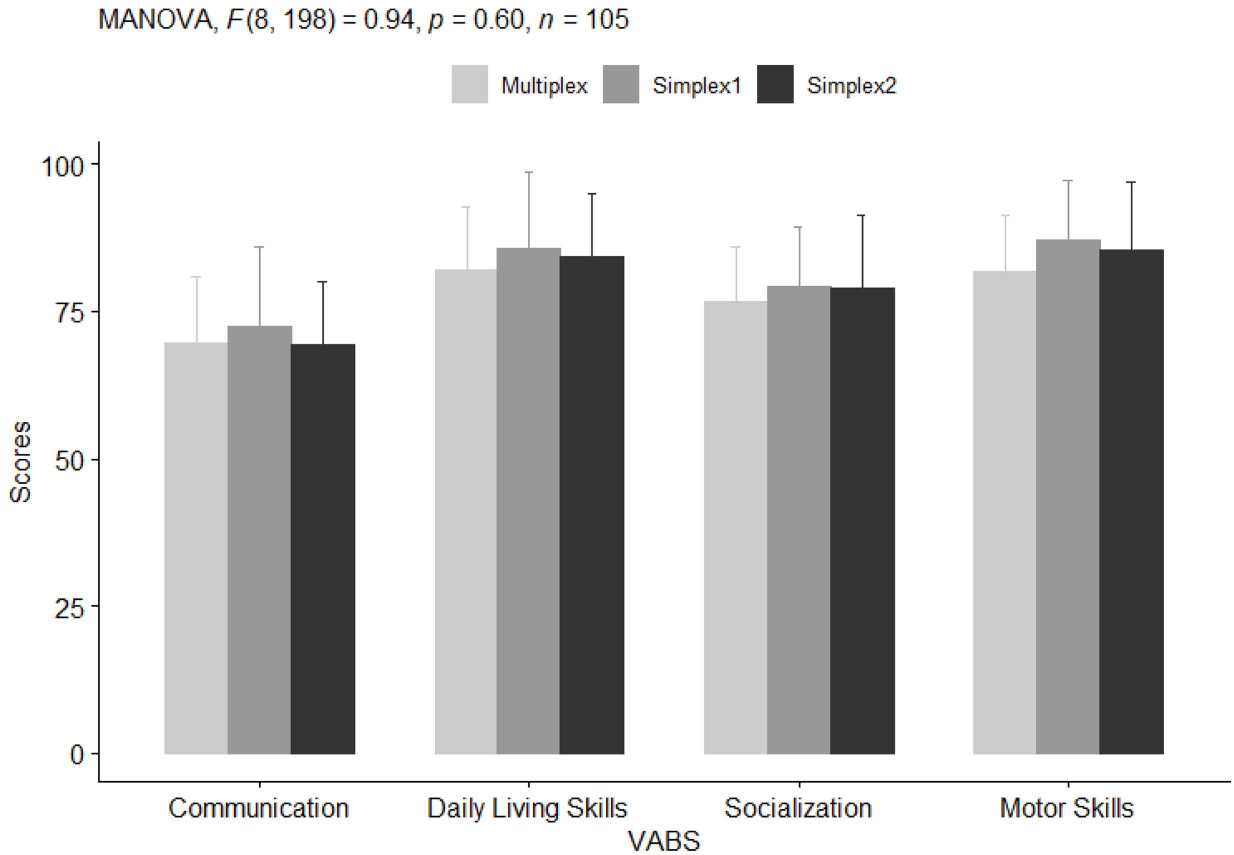

Fig. 4 MANOVA Results comparing mean VABS sub-scale scores between age and gender matched multiplex children, simplex-1 children, and simplex-2 children

child. If these findings are replicated, it may be prudent for clinicians to consider family group when interpreting parent report responses on all aspects of an ASD evaluation.

It is also possible that cognitive and adaptive skill development may be influenced by the presence of siblings, as research suggests that having a sibling leads to better adaptive functioning outcomes than not having a sibling (Ben-Itzchak et al., 2019; Rosen et al., 2022). However, these studies did not differentiate between multiplex and simplex grouping and the individuals assessed were older children, adolescents, or adults (Ben-Itzchak et al., 2019; Rosen et al., 2022). In the current study, there was not a significant difference in number of siblings between the multiplex and simplex2 group, but children in the simplex2 group had more siblings without an ASD diagnosis than the multiplex group. Adaptive functioning standardized domain scores did not differ across groups, and there was no clear pattern of an adaptive functioning advantage in specific skill areas on the VABS for simplex2 children (i.e., children with more neurotypical siblings) relative to children with fewer or no neurotypical siblings. We were unable to assess for potential associations between sibling birth order and adaptive skill development due to data limitations (i.e., birth order was not collected). To better understand the relative influence of genetics and environmental factors, future research with larger sample sizes should examine whether and how sibling presence and birth order is differentially associated with child cognitive and adaptive functioning in children from multiplex and simplex2 families.

Alternatively, these discrepancies could reflect a previously documented disadvantage in adaptive functioning relative to cognitive functioning in children with ASD (Perry et al., 2009). Notably, the gap between adaptive and cognitive functioning in ASD has mostly been examined in older children. It generally involves comparing composite IQ or DQ scores to adaptive functioning domains (i.e., communication, daily living skills, and socialization; Bradshaw et al., 2019; Fenton et al., 2003; Kanne et al., 2011; Perry et al., 2009) rather than directly comparing skill areas assessed by both measures as was the case in the current study. Recently, the first longitudinal study to examine discrepancies between adaptive functioning and cognitive functioning in infants and toddlers with ASD documented discrepancies for adaptive socialization and adaptive daily living skills, but not adaptive communication. Further, adaptive socialization and daily living skills scores declined from 12 to 36

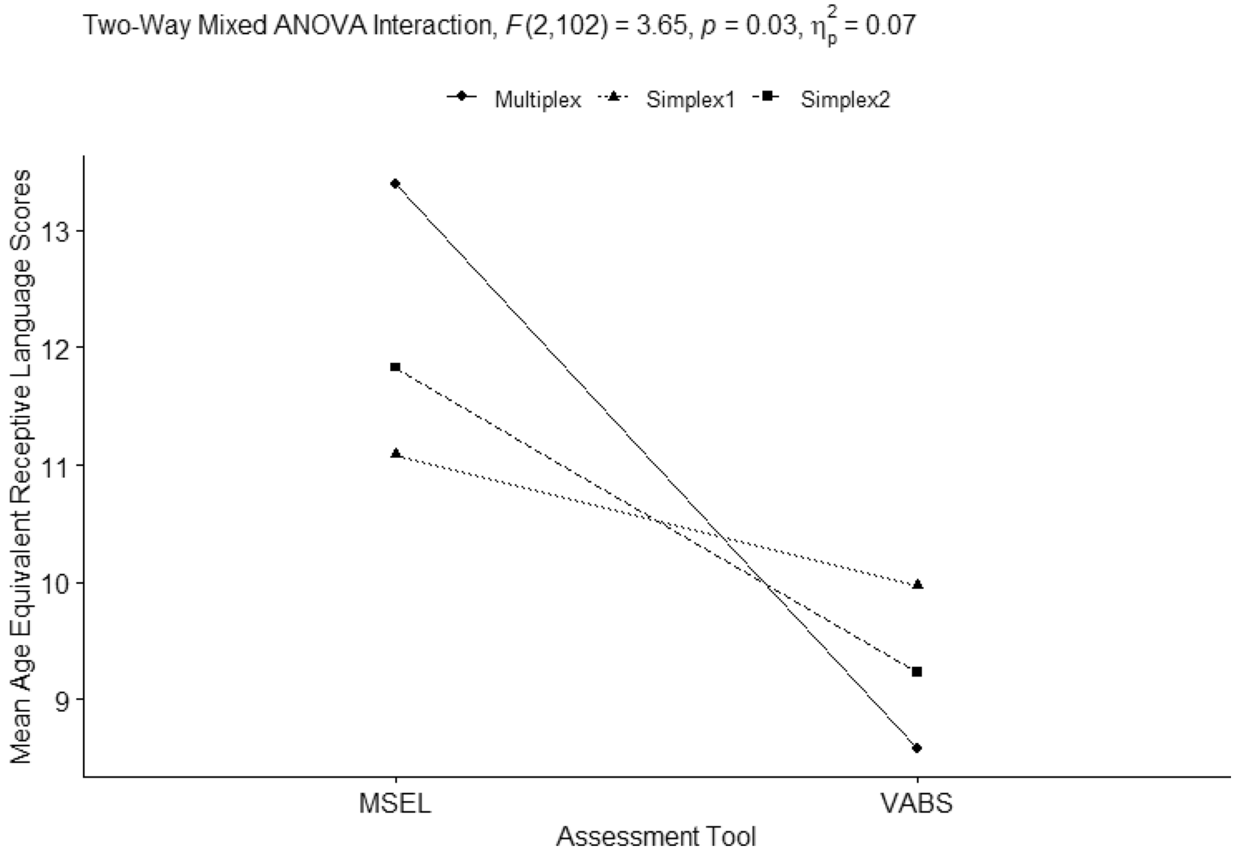

Fig. 5 Two-way mixed ANOVA results assessing if multiplex-simplex grouping influenced the relationship between receptive language results on the MSEL and VABS

months, whereas adaptive communication skills remained stable. These findings suggest that the relative disadvantage in adaptive functioning may not extend to communication skills in very young children with ASD (Bradshaw et al., 2019). Additional research is needed to better understand if discrepancies observed in the current study reflect a difference in assessment type (parent report vs. direct observation) or differences between adaptive functioning and cognitive skills.

Last, differences in MSEL and VABS scores may reflect differences in how specific skills are assessed and weighted within each of the assessments. While the receptive language, expressive language, and fine motor skill domains should theoretically be comparable between the two assessments, which are both standardized and norm-referenced, there is not perfect correspondence between specific items on each of the assessments, which could lead to a divergence of results. Future research should examine correspondence between specific VABS and Mullen items that assess the same skills to better understand differences between parent-report and direct observations of skills.

If these exploratory findings are replicated, they suggest that clinicians should consider family group (i.e., multiplex vs. simplex) when choosing assessments and interpreting assessment results. Only a few studies have examined the differences between direct professional observation and parent report (Noterdaeme et al., 2002; Saudino et al., 1998; Sturrock et al., 2020; Taylor et al., 2015; Virkud et al., 2009), and these studies were primarily focused on ASD behaviors. To our knowledge, no published research has examined the role family group may play in differences between direct professional observation and parent report, or in differences between adaptive and cognitive functioning. By splitting the current sample into multiplex and simplex groups, differential patterns of scores were uncovered. Future research could explore the potential effect these discrepancies may have on the diagnostic process for children at-risk for ASD and whether adjustments are warranted.

Limitations

A notable limitation of this study was the underpowered nature of the analyses (power = 0.61) for detecting moderate effect size differences. A larger sample size that is adequately powered to detect small and moderate between-group differences would enhance understanding of cognition and

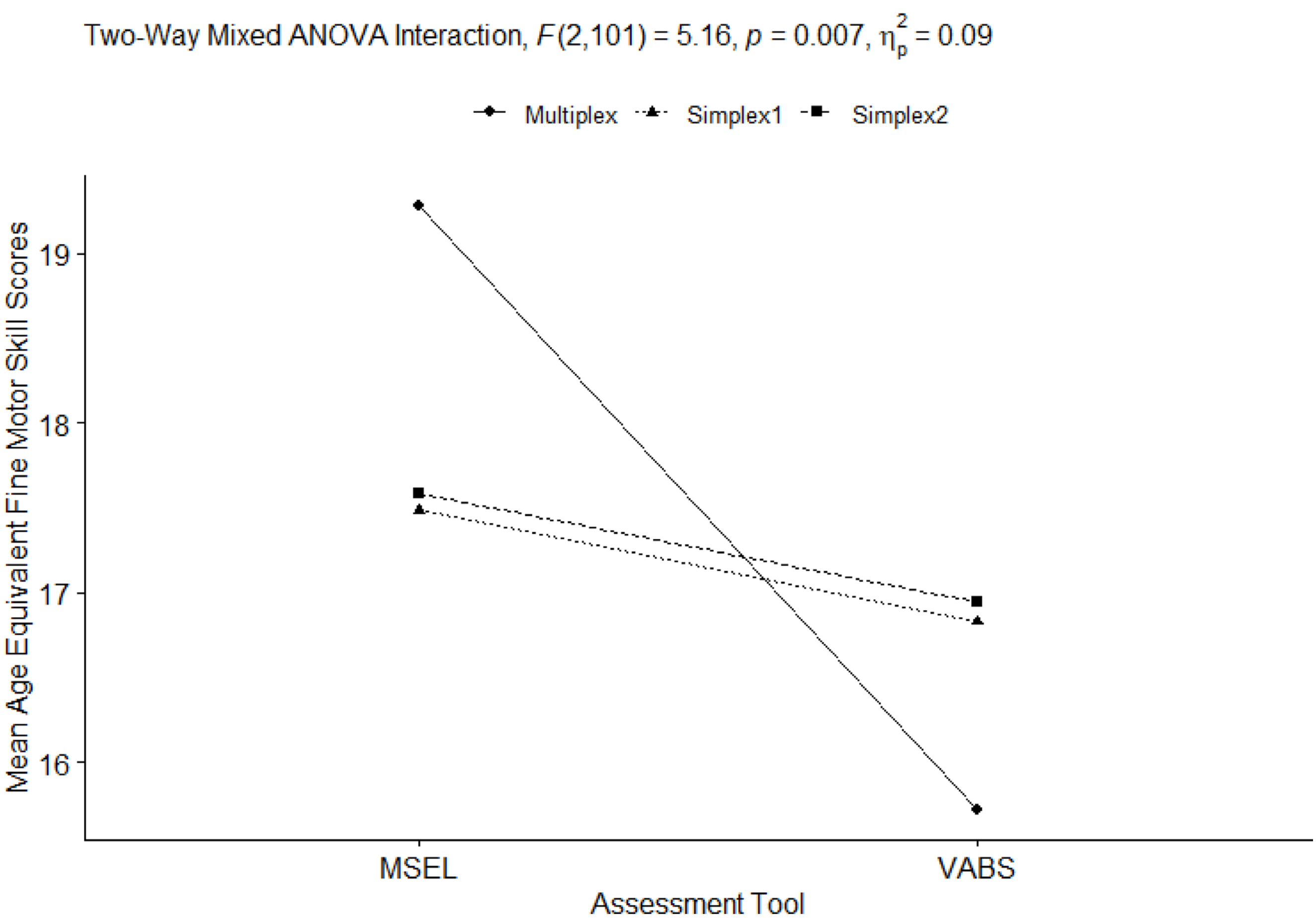

Fig. 6 Two-way mixed ANOVA results assessing if multiplex-simplex grouping influenced the relationship between fine motor skill results on the MSEL and VABS

adaptive functioning differences in toddlers at the time of diagnosis.

For both MANOVA analyses, the assumption of homogeneity of variance was violated. Additionally, the assumption of univariate normality was violated for VABS data and three mixed ANOVA analyses. MANOVA and mixed ANOVA can still be used when these violations are present, especially when group sizes are equal, although it is recommended to take a more conservative approach when interpreting results (Blanca Mena et al., 2017; Tabachnick et al., 2007).

Although the sample was identified through universal developmental screening in the general population, families volunteered to participate in the study and may differ substantively from families who did not agree to participate. Further, while Hispanic children were well represented in this study, children from other minority groups were not. In particular, non-Hispanic African American children were underrepresented in this sample (n = 2, 1.9%) compared to their share of the general population (U.S. Census Bureau, 2022). To improve generalizability, future research should oversample racially and ethnically diverse groups.

Last, like many other previous studies in this area, the presence or absence of siblings with ASD was determined based on parent report and siblings were not directly assessed for ASD. It is possible that some children in the simplex2 group had siblings with ASD that were undiagnosed and/or some children in the multiplex group had siblings diagnosed with ASD who were misdiagnosed. This may have resulted in participants being misclassified into multiplex or simplex groups. Although clinical phenotyping of siblings would increase the rigor of future studies and validity of findings, assessing all siblings in a sample of this size would be both time and resource intensive.

Conclusion

This study replicates and extends the findings of previous research about children with ASD from multiplex and simplex families in an age- and gender- matched sample with increased ethnic diversity relative to previous samples. Our results challenge the notion that statistically significant cognitive differences between children from multiplex and

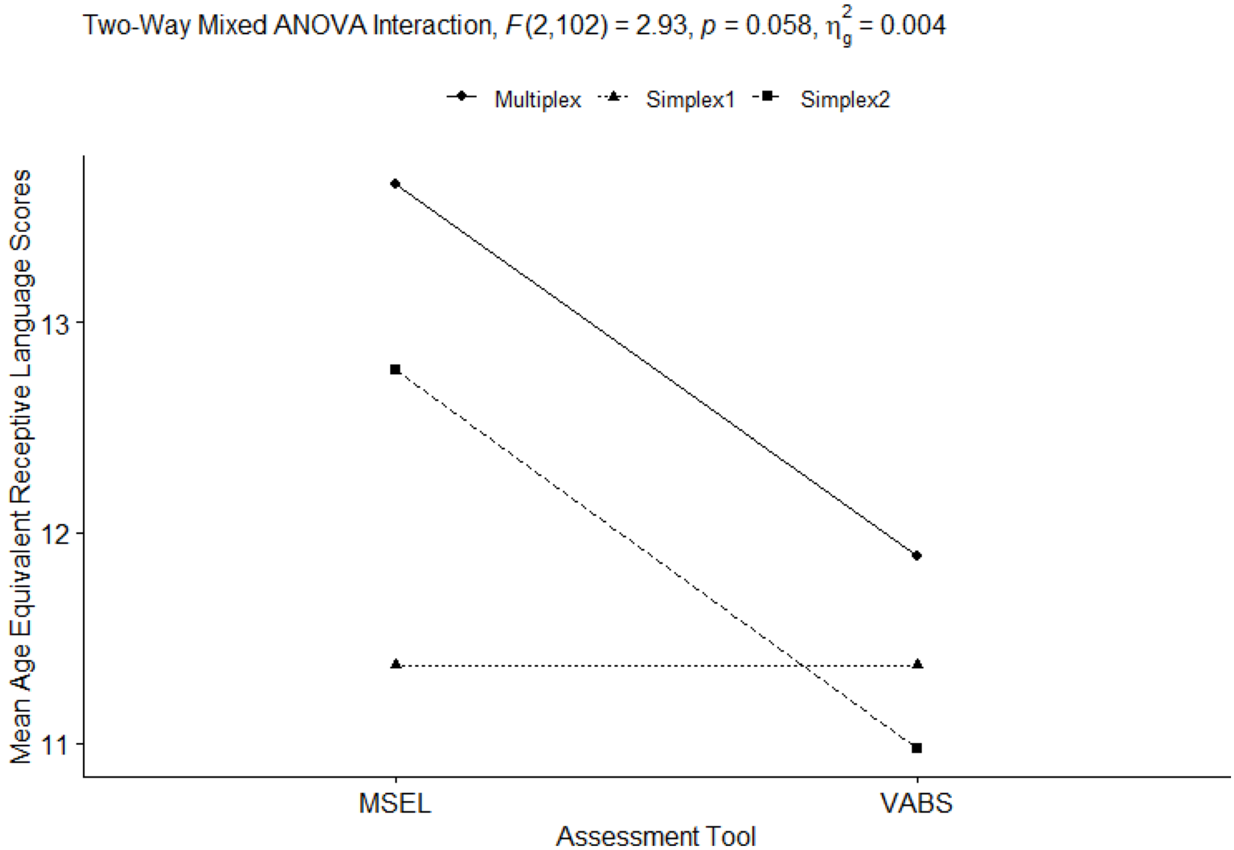

Fig. 7 Two-way mixed ANOVA results assessing if multiplex-simplex grouping influenced the relationship between expressive language results on the MSEL and VABS

simplex families can be detected at a young age. Interestingly, we found that across multiple skill areas (receptive language, expressive language, and fine motor skills) parents of children diagnosed with ASD underreported their child's skills compared to results from direct observation. Parents of children in multiplex and simplex2 groups significantly underreported receptive language skills relative to direct professional observation, whereas parents in the simplex1 group did not. Parents of children in multiplex families also underreported their child's skills in the fine motor skills domain relative to direct professional observation. Future research should attempt to replicate these findings with larger sample sizes and better understand whether parents are underreporting skills or whether VABS and MSEL subscales for receptive language, expressive language, and motor skills are measuring different constructs.

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Conflict of interest No conflict of interest.

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