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Early joint attention abilities measured by the ADOS-2 predict subsequent expressive language development in minimally verbal autistic children

Tanya Nitzan[1,2] | Moran Bachrach[1,2] | Michal Ilan[2,3] | Michal Faroy[2,3] | Danel Waissengreen[2,3] | Analya Michaelovsky[4] | Dikla Zagdon[2,3] | Yair Sadaka[2,5] | Omer Bar Yosef[2,6] | Ditza Zachor[2,7] | Einat Avni[2,7] | Idan Menashe[2,8] | Gal Meiri[2,3] | Judah Koller[9] | Ilan Dinstein[1,2]

[1]Psychology Department, Ben Gurion University of the Negev, Beer Sheva, Israel

[2]Azrieli National Centre for Autism and Neurodevelopment Research, Ben Gurion University of the Negev, Beer Sheva, Israel

[3]Pre-School Psychiatry Unit, Soroka University Medical Center, Beer Sheva, Israel

[4]Zusman Child Development Center, Soroka University Medical Center, Beer Sheva, Israel

[5]Child Development Center, Ministry of Health, Beer-Sheva, Israel

[6]Pediatric Neurology Units of Chaim Sheba Medical Center, Tel Hashomer, Israel

[7]The Autism Center/ALUT, Shamir (Assaf Harofeh) Medical Center, Tel Aviv University, Zerifin, Israel

[8]Public Health Department, Ben Gurion University of the Negev, Beer Sheva, Israel

[9]Seymour Fox School of Education, Hebrew University of Jerusalem, Jerusalem, Israel

Correspondence
Tanya Nitzan, Ben-Gurion University of the Negev, David Ben Gurion Blvd 1, Beer Sheva, Israel.
Email: tanya.nitzan@gmail.com

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Abstract

Background: Most preschool autistic children exhibit substantial language delays, yet only ~25% remain minimally verbal (MV) throughout life. Developing expressive language is crucial for improving outcomes. This study examined early predictors of later expressive language growth in MV preschool autistic children.

Methods: Data from 99 MV autistic children (mean age 27.7 months at diagnosis) who completed ADOS-2 assessments at diagnosis and 12–24 months later were analyzed. Children were stratified into three groups according to their language abilities at follow-up (MV, one-word, and phrases). Logistic regression was used to determine whether baseline ADOS-2 calibrated severity scores (CSS), non-verbal cognitive abilities, or Joint Attention (JA), derived from six ADOS-2 items, enabled prediction of expressive language abilities at follow-up.

Results: Children who successfully developed expressive language (one-word/phrases) had significantly lower baseline ADOS-2 social affect CSS and JA scores, but did not differ in their non-verbal cognitive abilities or ADOS-2 restricted and repetitive behaviors CSS. Gains in JA were significantly larger in children who developed expressive language.

Conclusion: MV preschoolers with better social abilities at diagnosis, specifically JA, were more likely to develop expressive language within 1–2 years. Joint attention scores derived from the ADOS-2 offer an easily accessible and widely available measure with important prognostic value for MV children.

KEYWORDS
autism, expressive-language, joint-attention, minimally verbal, predictors

Tanya Nitzan and Moran Bachrach contributed equally to this study.

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INTRODUCTION

Language delays are the most common initial concern that motivates parents of autistic children to seek clinical advice (Nitzan et al., 2023) and parents commonly rate language acquisition as a top priority for early intervention (Coonrod & Stone, 2004). Approximately 50%–60% of autistic children exhibit severe language delays and are minimally verbal (MV) at the age of three (Kissine et al., 2023). The definition of MV varies between studies, ranging from children who do not speak at all to those who use less than 50 words (Koegel et al., 2020). While some researchers reserve the term for school-aged children who continue to exhibit severely limited spoken language (Tager-flusberg & Kasari, 2013), in younger preschool samples children with no functional spoken language are often described as either "minimally verbal" or "preverbal." In the current study, we use the term MV to refer to preschool autistic children who demonstrated no functional spoken language, acknowledging that some of the younger children in this age range may alternatively be described as preverbal. We retain the term MV to remain consistent with prior longitudinal research in preschool autistic samples examining early language trajectories.

Despite intense intervention, only half of MV children will develop expressive language abilities by the age of seven, with the other half remaining MV throughout life, corresponding to ~25% of the entire autism population (Ellis Weismer & Kover, 2015; Fountain et al., 2012; Pickles et al., 2014; Rose et al., 2016; Tager-flusberg & Kasari, 2013). Of those who develop expressive language abilities between the ages of three to seven, various language trajectories are observed, ranging from gradual to unexpectedly rapid language gains at different ages (Anderson et al., 2007; Baghdadli et al., 2012; Ellis Weismer & Kover, 2015; Fountain et al., 2012; Georgiades et al., 2022; Pickles et al., 2014; Wodka et al., 2013). Most importantly, the emergence of expressive language is associated with the development of higher cognitive abilities, better adaptive functioning, and reduced severity of core autism symptoms, leading to better long-term life outcomes (Mouga et al., 2020) including wellbeing and mental health (Brignell et al., 2018; Howlin et al., 2013). There is, therefore, strong motivation to identify early predictors of expressive language development specifically for MV autistic children and develop new interventions for those who are unlikely to acquire expressive language using existing intervention protocols.

Previous studies have reported mixed findings regarding early child characteristics that may predict later expressive language development in MV autistic children. Examined predictors have included the overall severity of autism symptoms (Ellis Weismer & Kover, 2015), social abilities (Ellis Weismer & Kover, 2015; Latrèche et al., 2024; Thurm et al., 2015; Yoder et al., 2015; Zachor et al., 2007), cognitive abilities (Anderson et al., 2007; Ellis Weismer & Kover, 2015; Kilili-Lesta et al., 2024; Mouga et al., 2020; Norrelgen et al., 2015; Thurm et al., 2015), and JA abilities (Anderson et al., 2007; Kasari et al., 2023; Kilili-Lesta et al., 2024; McDaniel et al., 2017; Sandbank et al., 2017; Yoder et al., 2015). Note that we are focusing this introduction on longitudinal studies that specifically examined language development in MV autistic children (i.e., those with severe language delays), rather than studies that included autistic children with mild language delays.

Key Points

What's known?

- Developing expressive language is critical for minimally verbal (MV) autistic preschoolers, motivating research about early factors that predict expressive language development.

What's new?

- In this longitudinal study of 99 MV autistic preschoolers, joint attention (JA) and social affect (SA) at diagnosis predicted expressive language outcomes 1–2 years later.
- JA and SA were measured with the ADOS-2, which is used widely in research and clinical settings, offering easily accessible measures with prognostic value.

What's relevant?

- ADOS-2 scores can guide early identification of MV autistic children who are likely to successfully develop expressive language with existing interventions versus those who need greater support or new interventions.

Several studies have examined ADOS-2 or older ADOS scores as potential predictors of expressive language development in MV preschool autistic children and reported mixed findings. One study reported a significant relationship between early total ADOS Calibrated Severity Scores (CSS) and later expressive language scores (Ellis Weismer & Kover, 2015). However, others did not, whether using total ADOS-2 CSS (Thurm et al., 2015), ADOS-2 social affect (SA) CSS (Latrèche et al., 2024; Thurm et al., 2015), or ADOS social communication scores (Yoder et al., 2015). ADOS-2 restricted and repetitive behaviors (RRB) CSS also did not predict later expressive language development in MV preschool autistic children (Thurm et al., 2015).

One important aspect of early social communication is JA, where two individuals allocate their attention to the same object, action, or target in a synchronized manner, enabling them to communicate effectively (Mundy, 2018; Mundy et al., 2016). The ability of a child to respond to JA bids from adults seems to be particularly important for learning the meaning of specific words (Bottema-Beutel, 2016) and by extension, for developing language abilities. Different studies have quantified JA abilities in different ways. While some used manual coding of videotaped interactions during the Early Social Communication Scales (ESCS) (Kasari et al., 2023; McDaniel et al., 2017; Saul & Norbury, 2020) or the Communication and Symbolic Behavior Scales Developmental Profile Behavior Sample (CSBS) (McDaniel et al., 2017; Sandbank et al., 2017; Saul & Norbury, 2020; Yoder et al., 2015), others used scores from specific items of the ADOS (Anderson et al., 2007) or parent questionnaires (Kilili-Lesta et al., 2024). Most studies reported that different measures of early JA behaviors including child initiated JA (IJA) and responding to JA (RJA) are associated with better expressive language outcomes at later ages (Anderson et al., 2007; Kasari et al., 2023; Kilili-Lesta et al., 2024; McDaniel et al., 2017; Sandbank et al., 2017; Yoder et al., 2015), while a few studies did not (McDaniel et al., 2017; Saul & Norbury, 2020).

Similarly, most studies have reported that higher cognitive abilities at early ages are associated with greater likelihood of subsequent language acquisition (Anderson et al., 2007; Kilili-Lesta et al., 2024; Mouga et al., 2020; Norrelgen et al., 2015; Thurm et al., 2015), potentially outperforming autism severity measures as a predictor of language development (Ellis Weismer & Kover, 2015; Thurm et al., 2015). However, other studies did not find early cognitive ability to predict later expressive language development (Latrèche et al., 2024; Yoder et al., 2015).

The goal of the current study was to revisit this topic with a relatively large longitudinal sample of preschool autistic children who were defined as MV at diagnosis using strict criteria (i.e., children who do not speak at all). We prospectively followed these children over a one-to-two-year period as they participated in repeated standardized testing with ADOS-2 assessments, allowing us to compare the initial characteristics of those who successfully acquired basic language abilities with those who remained MV.

METHODS

Participants and procedures

Our sample of convenience included 99 children (82 boys), 18–50 months old at diagnosis, who were diagnosed with autism between 2016 and 2022, and did not have any known genetic disorders (Table 1). All children completed ADOS-2 (Lord et al., 2012) assessments at two time points, one at diagnosis (T1) and another 12–24 months later (T2). Children were included in the study only if they were classified as MV based on their score on the A1 item of the ADOS-2 at T1 (i.e., all children produced no spontaneous spoken words or only echoed speech at T1). Data was extracted from the National Autism Database of Israel (Dinstein et al., 2020), managed by the Azrieli National Center for Autism and Neurodevelopment Research (ANCAN). ANCAN is a collaboration between Ben-Gurion University of the Negev (BGU) and 8 clinical sites throughout Israel where children with autism are diagnosed and followed up prospectively. The study was approved by the Soroka University Medical Center Helsinki Committee (SOR-222-14). Since this was a retrospective study of anonymized data, there was no need to obtain signed consent from the participating families (exempt status).

Measures

ADOS-2

At T1, all children were non-verbal and, therefore, completed either the Toddler module or module 1 of the ADOS-2 (Lord et al., 2012). At T2, children completed the Toddler Module, Module 1, or Module 2, according to their age and verbal abilities. All ADOS-2 assessments were administered by experienced clinicians with research reliability. ADOS-2 CSS allow comparison of autism severity across children of different ages and language capabilities and across time points (Esler et al., 2015; Gotham et al., 2009) and were also computed separately for SA and restricted and repetitive behaviors (RRBs) (Esler et al., 2015; Hus et al., 2014).

Non-verbal cognitive abilities

A total of 69 children (69.7% of the sample) completed cognitive testing at T1 with the Bayley Scales of Infant and Toddler Development, Third Edition (Vanegas, 2021), which was administered by a licensed psychologist. We used the Bayley cognitive composite scores in all analyses as this scale measures the non-verbal cognitive abilities of the children. This measure has a normed population mean of 100 and a standard deviation of 15. Missing data for the remaining children were imputed (see below).

Verbal abilities

Verbal ability was evaluated using the A1 item of the ADOS-2 assessment, which evaluates the spoken language abilities of the child. In module 1 and the toddler module, this item scores verbal abilities from no spoken language to using pairs of words, in module 2 from single words to combinations of three or more words, and in module 3 from combinations of 2–3 words to complex sentences. We used the coding system developed by Visser et al. (2017) (Visser et al., 2017) to create a common eight-point spoken language scale across modules with the following values:

1. Children use sentences in a largely correct fashion (complex utterances with >2 clauses).
2. Children exhibit relatively complex speech (occasional utterances with >2 clauses) with recurrent grammatical errors.
3. Children exhibit non-echoed speech with utterances of >3 words.
4. Children mainly use individual 2- to 3-word phrases, with or without minimal grammar.
5. Children mainly use individual words with occasional simple phrases.
6. Children only use individual words (minimum of 5 different words).
7. Children only use echoed speech (<5 words).
8. No language production at all.

While Mazurek et al. (2019) proposed another language scale that incorporated A1 item scores across multiple ADOS-2 modules including the Toddler Module, our analysis revealed that their scale was perfectly negatively correlated with the Visser et al. scale ($r = -1$). Hence, the results presented here using the Visser scale would be equivalent had we used the Mazurek scale.

We classified the sample into three verbal ability groups at follow-up (T2): MV (scores 6–7), One-word (scores 4–5), and Phrases (scores 2–3). None of the children in our sample displayed fluent speech (i.e., scores 0–1) at T2.

Joint Attention

We quantified Joint Attention (JA) at T1 and T2 by summing the scores of six items, similarly to those identified by previous factor analysis studies of the ADOS (Gotham et al., 2007, 2008). Items

TABLE 1 Characteristics of the children (all MV at T1), separated into three subgroups according to their verbal abilities at T2.

	MV (n = 49)	One-word (n = 33)	Phrases (n = 17)	Overall (n = 99)
Measures	**Mean (SD)**			
Age at diagnosis (M)	28.6 (7.31)	27.0 (7.22)	26.6 (5.80)	27.7 (7.03)
Age at follow-up (M)	42.9 (7.25)	40.8 (7.67)	44.2 (6.70)	42.4 (7.34)
Sex, % boys	81.6	84.8	82.4	82.8
Paternal age at diagnosis (Y)	39.4 (6.99)	35.4 (7.65)	36.3 (6.00)	37.5 (7.25)
Paternal education (Y)	12.8 (2.47)	13.2 (2.40)	12.7 (1.83)	12.9 (2.33)
Maternal age at diagnosis (Y)	35.9 (5.38)	32.4 (5.78)	33.3 (4.75)	34.3 (5.60)
Maternal education (Y)	13.1 (2.19)	13.1 (1.97)	14.2 (2.30)	13.3 (2.16)
Verbal ability on ADOS-2 at T2	6.47 (0.50)	4.58 (0.50)	2.71 (0.470)	5.19 (1.50)
ADOS-2 SA-CSS at T1	8.94 (1.48)	8.00 (1.89)	7.47 (2.24)	8.37 (1.84)
ADOS-2 RRB-CSS at T1	7.53 (1.57)	7.24 (1.35)	6.94 (1.52)	7.33 (1.49)
ADOS-2 CSS at T1	8.67 (1.30)	7.85 (1.73)	7.12 (1.93)	8.13 (1.66)
Non-verbal cognitive abilities at T1	70.9 (12.5)	74.7 (10.5)	78.1 (14.6)	73.4 (12.2)
JA score at T1	10.8 (1.42)	9.70 (2.08)	8.71 (2.73)	10.1 (2.06)
ADOS-2 SA-CSS at T2	7.39 (1.77)	6.27 (2.14)	5.35 (2.60)	6.67 (2.18)
ADOS-2 RRB-CSS at T2	8.22 (1.21)	8.09 (1.49)	8.12 (1.54)	8.16 (1.35)
ADOS-2 CSS at T2	7.84 (1.68)	7.03 (2.04)	6.06 (2.44)	7.26 (2.03)
JA score at T2	9.96 (2.02)	6.64 (2.99)	3.53 (2.55)	7.75 (3.45)
Duration in special education (M)	7.12 (5.55)	8.03 (5.82)	9.35 (7.64)	7.81 (6.03)

Abbreviations: ADOS-2, autism diagnostic observation schedule, second edition; CSS: calibrated severity scores; JA, joint attention; M, months; RRB, restricted and repetitive behaviors; SA, social affect; Y, Years.

included Pointing (item A7 in the Toddlers module and Module 1, item A6 in Module 2), Gesturing (item A8 in the Toddlers module and Module 1 and item A7 in Module 2), Showing (item B12 in the Toddlers module, B9 in Module1, and B5 in Module 2), Initiating JA (item B13 in the Toddlers module, B10 in Module1, and B6 in Module 2), Unusual Eye contact (item B1 in all modules), and Reaction to JA (item B14 in the Toddlers module, B11 in Module1, and B7 in Module 2). In line with the ADOS-2 algorithm, scores of 3 were changed to 2, and scores of 8 were changed to 0, yielding a total JA scale of 0–12 with higher scores indicating poorer JA abilities (i.e., larger difficulties). While the original studies (Gotham et al., 2007, 2008; Oosterling et al., 2010) included different subsets of items for different ADOS-2 modules, here we used a consistnat set of six items across modules for simplicity.

As defined by the ADOS-2 guidelines, the selected items were assigned a score of 0 (i.e., no impairment) only if the child exhibited clear social-communicative intent, which is central to JA. For example, the 'pointing' item was assigned a score of 0 only if pointing was performed to a remote object and combined with eye contact to demonstrate shared interest. Pointing to simply request something received a higher score indicative of impairment. Hence, the selected items measured a composite of JA behaviors that are equivalent to those assessed by the Early Social Communication Scales (ESCS; Mundy et al., 2003) and the Communication and Symbolic Behavior Scales (CSBS; Wetherby et al., 2002), but without splitting JA into Responsive (RJA) and Initiated (IJA) components.

Additional variables

Parents completed follow-up questionnaires at T2 where they reported maternal and paternal age at time of their child's diagnosis, their years of education, and the duration of time that their child had attended special education up to T2. The number of respondents varied by variable, with paternal age reported for 87 children (87.9% of the sample), paternal education for 80 children (80.8 of the sample), maternal age for 94 children (94.9% of the sample), maternal education for 88 children (88.9% of the sample), and the Duration in special education for 93 children (93.9% of the sample). Missing data for the remaining children were imputed (see below).

Data analysis and statistics

All statistical analyses were conducted using RStudio (RStudio Inc., Boston, MA), version 4.4.3. All children included in the study were MV at diagnosis (T1) and classified into three groups according to their verbal ability score at follow-up (T2): MV (scored 6/7), One-word (scored 4/5), or Phrases (scored three or less). We imputed missing data, including cognitive scores and parent follow-up questionnaire responses, using the Random Forests technique with 20 iterations as implemented in the *mice* package in RStudio (van Buuren & Groothuis-Oudshoorn, 2011). To ensure that this imputation process did not introduce biases to the data we compared all study variables between children who completed cognitive testing versus

those who did not. This analysis demonstrated that there were no significant differences (all $p > 0.085$) across the two groups in any of the study variables (Table S1). Hence, missing cognitive data was not systematically biased toward children with specific characterisitcs.

We used the imputed dataset in all analyses. A series of one-way analyses of covariance (ANCOVAs) were conducted to determine whether initial cognitive, ADOS-2 SA-CSS, ADOS-2 RRB-CSS, or JA scores at T1 differed significantly across verbal ability groups as defined at T2. We controlled for multiple covariates including age of diagnosis, maternal education, parental age at diagnosis, duration of time in special education, and duration of time between ADOS-2 evaluations. The duration time between assessments ranged from 12 to 24 months and was modeled as a continuous covariate in all analyses. Post hoc pairwise comparisons were conducted using Tukey's Honestly Significant Difference (HSD) test to adjust for multiple comparisons following significant effects in the ANCOVA. Eta squared (η^2) was calculated for each ANCOVA to assess the effect size of group differences. Equivalent analyses were performed to compare longitudinal changes in ADOS-2 and JA scores across the three verbal ability groups.

We also performed several logistic regression analyses to predict verbal ability at T2 using multiple T1 measures as predictors. Model fit was assessed using McFadden's pseudo R^2 and the Akaike Information Criterion (AIC). In all analyses, statistical significance was set at $\alpha = 0.05$.

RESULTS

We first performed ANCOVA analyses to assess whether ADOS-2 SA CSS, ADOS-2 RRB CSS, non-verbal cognitive abilities, or JA scores at diagnosis (T1) differed across children who developed expressive language abilities versus those who remained MV at follow up (T2). These analyses (Figure 1) revealed significant differences in ADOS-2 SA CSS at T1 across the three verbal ability groups ($F(2, 90) = 5.66$, $p = 0.005$, $\eta^2 = 0.102$). Post hoc Tukey HSD comparisons showed that children in the MV group had significantly higher SA CSS than those in the Phrases group (mean difference = −1.47, 95% CI [−2.58, −0.35], $p = 0.006$) or One-word group (mean difference = −0.94, 95% CI [−1.83, −0.07], $p = 0.037$). There were no significant differences between the One-word and Phrases groups (mean difference = −0.53, 95% CI [−1.71, 0.65], $p = 0.537$) and there were no significant relationships with any of the covariates (maternal education, parental age, duration in special education, and duration between T1 and T2), except for age of diagnosis ($F(1, 90) = 14.98$, $p < 0.001$). This demonstrated that MV autistic children with lower ADOS-2 SA CSS at diagnosis were more likely to develop expressive language abilities at T2 even when accounting for their age of diagnosis.

JA scores at diagnosis (T1) also differed significantly across the three verbal ability groups ($F(2, 90) = 6.47$, $p = 0.002$, $\eta^2 = 0.15$). Post hoc Tukey HSD indicated that children in the MV group had significantly higher JA scores (i.e., poorer JA skills) than those in the Phrases group (mean difference = −2.07, 95% CI [−3.37, −0.77], $p = 0.0007$), and also significantly higher scores than the One-word group (mean difference = −1.08, 95% CI [−2.12, −0.04], $p = 0.040$). There were no significant differences between the One-word and Phrases groups (mean difference = −0.99, 95% CI [−2.37, 0.38], $p = 0.204$). All covariates were not significantly associated with language outcome. Hence, MV autistic children with better JA skills at T1 were more likely to develop expressive language abilities at T2.

There were no significant differences in non-verbal cognitive abilities ($F(2, 90) = 0.66$, $p = 0.519$) or ADOS-2 RRB CSS ($F(2, 90) = 0.02$, $p = 0.983$) at diagnosis across the three verbal ability groups.

Next, we performed two logistic regression analyses (Table 2) to evaluate the ability of combinations of T1 measures to predict verbal ability at T2. The first logistic regression model included ADOS-2 SA CSS, ADOS-2 RRB CSS, cognitive score, age of diagnosis, maternal age at diagnosis, paternal age at diagnosis, maternal education, duration in special education, and duration of time between T1 and T2 (i.e., diagnosis and follow up). In this analysis only ADOS-2 SA CSS at T1 emerged as significant predictors of verbal ability at T2. In a second logistic regression analysis we replaced ADOS-2 SA CSS with JA scores and included all other predictors as described above. In this analysis, only JA scores emerged as significant predictors of verbal abilities at T2. Note that replacing SA CSS with JA scores yielded a model with similar predictive power as indicated by similar pseudo R^2 (0.172 vs. 0.176 respectively) and AIC values (133.62 vs. 133.03 respectively). Moreover, SA CSS and JA scores exhibited equivalent odds ratios (0.41) within their respective models demonstrating similar predictive power.

Longitudinal changes in core autism symptoms across language groups

Additional one-way ANCOVA tests demonstrated that longitudinal changes in ADOS-2 SA-CSS between T1 and T2 did not differ significantly across verbal ability groups ($F(2, 90) = 0.38$, $p = 0.686$) nor did longitudinal changes in ADOS-2 RRB-CSS ($F(2, 90) = 0.40$, $p = 0.674$). In contrast, longitudinal changes in JA scores did differ significantly across verbal ability groups ($F(2, 90) = 18.45$, $p < 0.001$, $\eta^2 = 0.28$). Post hoc Tukey HSD comparisons revealed that children in the MV group exhibited smaller gains (i.e., reductions in JA scores) than children in the One-word group (M difference = −2.24, 95% CI [−3.70, −0.79], $p = 0.001$) or the Phrases group (M difference = −4.36, 95% CI [−6.16, −2.55], $p < 0.001$). A significant difference was also found between the One-word and Phrases groups (M difference = −2.11, 95% CI [−4.03, −0.20], $p = 0.03$) (Figure 2). All covariates were not significantly associated with language outcome. Hence, MV autistic children who develop expressive language abilities also develop better JA abilities.

DISCUSSION

Our results demonstrate that early ADOS-2 SA CSS and JA scores are similarly predictive of successful expressive language development in young MV autistic children 1–2 years after diagnosis. This was evident in similar effect sizes for SA and JA (OR = 0.41) predictors in their respective logistic regression models (Table 2). Since JA scores were extracted from a subset of ADOS-2 SA items, this suggests that they contain the essential information needed to

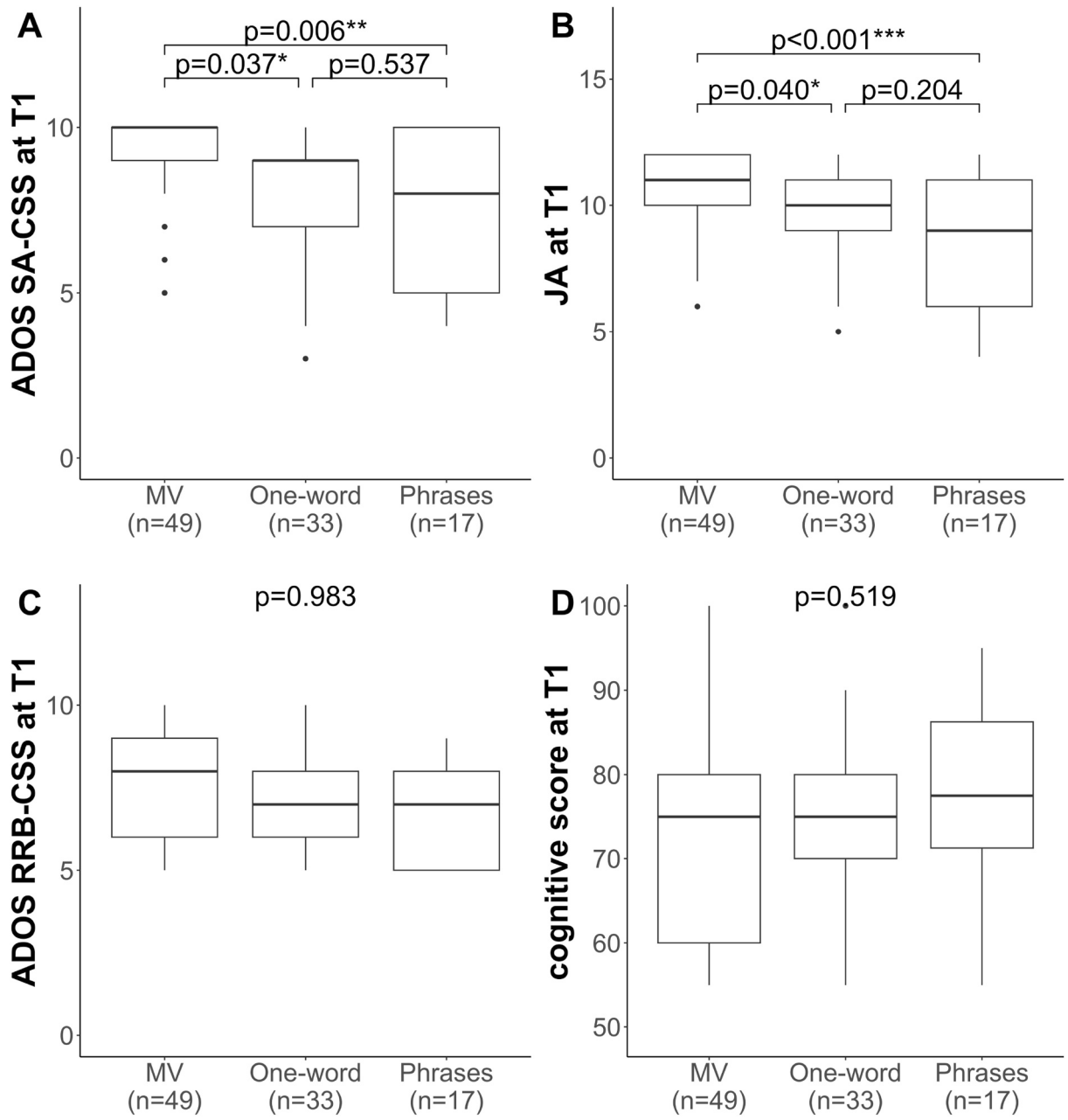


FIGURE 1 Box and whisker plots demonstrate T1 differences across children who remained minimally verbal versus those who developed expressive language abilities (One-word or Phrases) at T2. (A) ADOS-2 SA-CSS (B) Joint attention (C) ADOS-2 restricted and repetitive behaviors-CSS (D) Non-verbal cognitive scores. Statistical significance of differences across groups from ANCOVA analyses are presented in each panel. ADOS-2, autism diagnostic observation schedule, second edition; CSS, calibrated severity scores.

predict subsequent expressive language development. In addition, JA scores improved significantly with time in MV autistic children who developed expressive language while SA CSS did not. This suggests that there is a particularly strong relationship between JA as quantified by the selected ADOS-2 items and expressive language development.

JA scores and later expressive language development in MV children

Multiple previous studies have already reported that better JA skills at diagnosis were associated with greater development of expressive language skills in MV autistic children (Anderson et al., 2007; Kasari et al., 2023; Kilili-Lesta et al., 2024; McDaniel et al., 2017; Sandbank et al., 2017; Yoder et al., 2015). However, many of these studies used manual coding of videotaped interactions during the ESCS (Kasari et al., 2023; McDaniel et al., 2017; Saul & Norbury, 2020) or the Communication and Symbolic Behavior Scales Developmental Profile Behavior Sample (CSBS) (McDaniel et al., 2017; Sandbank et al., 2017; Saul & Norbury, 2020; Yoder et al., 2015), which are time-consuming and unscalable. An important contribution of the current study is in demonstrating that JA scores, extracted from six items of the ADOS-2 can predict expressive language development in MV autistic children.

The six ADOS-2 items included in our measure of JA were selected based on previous factor analysis studies suggesting that they contribute to a single factor (Gotham et al., 2007, 2008;

TABLE 2 Logistic regression analyses for predicting verbal ability at T2.

Model 1						Model 2					
Variable	**OR**	**SE**	**95% CI**	***z***	***p***	**Variable**	**OR**	**SE**	**95% CI**	***z***	***p***
Intercept	1.00	0.00	1.00, 1.00	2.18	0.029*	Intercept	1.00	0.00	[1.00, 1.00]	1.99	0.047*
ADOS-2 SA CSS	**0.41**	**0.31**	**[0.21, 0.71]**	**−2.95**	**0.003****	**JA**	**0.41**	**0.30**	**[0.22, 0.71]**	−2.98	**0.003****
ADOS-2 RRB CSS	0.98	0.25	[0.60, 1.62]	−0.07	0.946	ADOS-2 RRB CSS	1.05	0.26	[0.63, 1.75]	0.18	0.855
Non-verbal cognitive abilities	1.01	0.26	[0.60, 1.69]	0.05	0.960	Non-verbal cognitive abilities	1.08	0.26	[0.65, 1.79]	0.30	0.767
Age of diagnosis	0.62	0.28	[0.35, 1.04]	−1.76	0.079	Age of diagnosis	0.72	0.26	[0.43, 1.19]	−1.27	0.204
Maternal education	1.16	0.25	[0.70, 1.93]	0.60	0.551	Maternal education	1.27	0.25	[0.77, 2.12]	0.94	0.3498
Maternal age at diagnosis	0.66	0.32	[0.35, 1.23]	−1.30	0.194	Maternal age at diagnosis	0.65	0.31	[0.35, 1.20]	−1.37	0.170
Paternal age at diagnosis	0.75	0.30	[0.41, 1.34]	−0.97	0.332	Paternal age at diagnosis	0.76	0.30	[0.41, 1.36]	−0.92	0.358
Duration in special education	1.33	0.29	[0.76, 2.38]	0.99	0.321	Duration in special education	1.20	0.28	[0.70, 2.13]	0.66	0.508
Duration between T1 and T2	0.96	0.28	[0.55, 1.71]	−0.13	0.897	Duration between T1 and T2	0.98	0.29	[0.55, 1.73]	−0.08	0.939
Observations: 99						Observations: 99					
Pseudo R-squared (McFadden): 0.172						Pseudo R-squared (McFadden): 0.176					
AIC: 133.62						AIC: 133.03					

Note: Bold values indicate statistically significant predictors ($p < 0.05$).

*$p < 0.05$.

**$p < 0.01$.

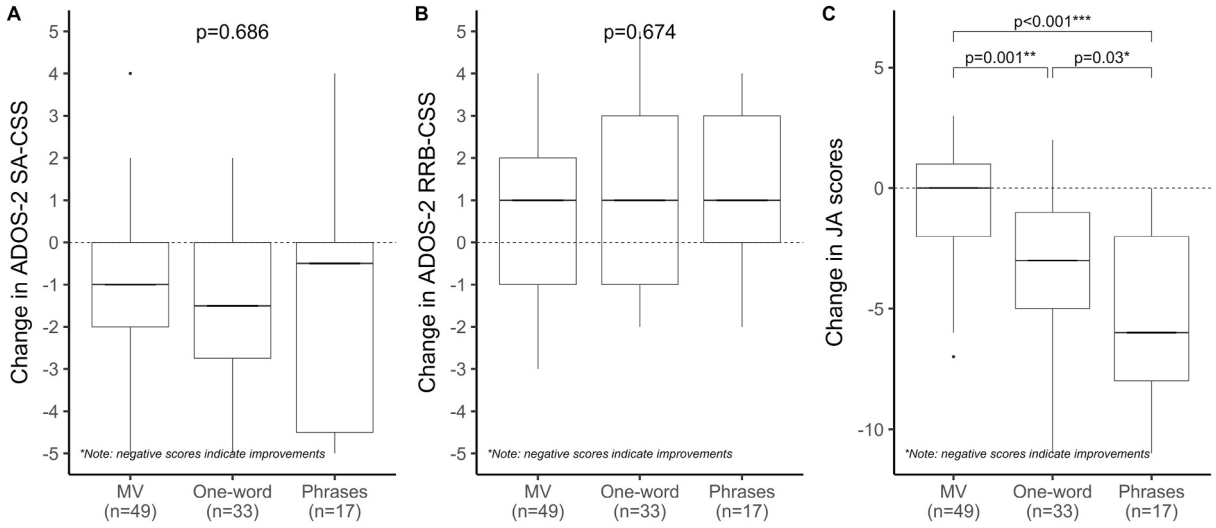


FIGURE 2 Box and whisker plots of longitudinal changes in ADOS-2 scores across verbal abilities groups. (A) Change in ADOS-2 social affect-CSS (T1–T2). (B) Change in ADOS-2 restricted and repetitive behaviors-CSS. (C) Change in joint attention scores. Solid horizontal lines represent the group median. Negative values indicate improvement (i.e., reduction in ADOS-2 scores) over time; no-change is represented by the dashed line. ADOS-2, autism diagnostic observation schedule, second edition; CSS, calibrated severity scores.

Oosterling et al., 2010). Indeed, these items measure different JA behaviors where the interaction between the child and clinician is scored according to its social-communicative inent. For example, when scoring the "pointing" item, the child must exhibit pointing to a distant object with coordinated eye contact to receive a score of 0 indicative of no impairment. Hence, the selected ADOS-2 items measured equivalent JA behaviors to those assessed by the ESCS and CSBS, but without splitting the final score to Responsive and Initiated JA. Our results suggest that a single summary measure of JA that can be easily extracted from the ADOS-2 of MV autistic children is indeed predictive of their later expressive language development.

Two previous studies using similar subsets of older ADOS items have already demonstrated that JA abilities from ADOS item scores were predictive of expressive language development in smaller mixed samples that included MV autistic children. Anderson et al. (2007) summed the scores of four ADOS items: gestures, pointing, showing, and spontaneous initiation of JA (children with some speech) or gestures, use of other's body to communicate, response to name, and

response to JA (nonverbal children). Thurm et al. (2007) used scores from the initiation of JA and response to JA items separately. In both cases JA scores predicted the development of expressive language in MV autistic children at follow-up. Our results extend previous findings by demonstrating that JA scores extracted from newer ADOS-2 items in relatively large sample of MV autistic children are predictive of their expressive language development.

Early ADOS-2 SA scores and later expressive language development in MV children

Despite the widespread use of the ADOS and ADOS-2 assessments in autism research, relatively few studies have examined their utility in predicting expressive language development of MV children and have yielded mixed results (Ellis Weismer & Kover, 2015; Latrèche et al., 2024; Thurm et al., 2015; Yoder et al., 2015). Importantly, two key studies that used the newer ADOS-2 test and extracted SA CSS reported no association with later expressive language development in 47 (Thurm et al., 2015) and 44 (Latrèche et al., 2024) children who were MV at the age of ~3.5 and 2.5 years-old, respectively. Our results did show a significant association between early ADOS-2 SA CSS and later expressive language abilities. This difference across studies may be due to several reasons.

First, one important difference between our study and both the Thurm et al. (2015) and Latrèche et al. (2024) studies was in how language groups were defined. Our MV criteria were the strictest, such that all children in the current study had no language or only echoed speech at diagnosis. In contrast, Thurm et al. (2015) included children who spoke some single words and occasional phrases in their MV group. Latrèche et al. (2024) used a parent-reported questionnaire to estimate language abilities, performed a clustering analysis, and defined MV as the cluster with expressive language scores that were 4 standard deviations below population standards (i.e., included children who spoke single words and occasional phrases according to parent report). Second, there were differences in the samples recruited for the different studies. Children in the Thurm et al. (2015) study were severely impacted individuals with cognitive scores of ~60, on average, while the Latrèche et al. (2024) and our study included children with cognitive scores of ~70, on average, which are more representative of the general autism population (Dinstein et al., 2020). We also did not include any children with known genetic disorders, a criterion that was not reported by the previous studies. Third, While the Thurm et al. (2015) study used ADOS assessments with children diagnosed according to DSM-4 criteria, our study and the Latrèche et al. (2024) study used ADOS-2 assessments with children diagnosed according to DSM-5 criteria. Finally, the sample size in our study was twice the size of the previous ones, thereby affording considerably larger statistical power.

Hence, in contrast to the previous studies, our study compared strictly defined MV children who did develop expressive language with those who did not, while using a large sample of ADOS-2 assessments collected from community-recruited idiopathic autism cases and diagnosed according to DSM-5 criteria. We, therefore, believe that our findings are persuasive in demonstrating that early ADOS-2 SA CSS are associated with later expressive language outcomes in this specific subgroup of MV autistic children.

Other predictors of expressive language development in MV children

Previous studies have reported that early cognitive abilities are a significant predictor of later expressive language development in MV autistic children (Anderson et al., 2007; Kilili-Lesta et al., 2024; Latrèche et al., 2024; Mouga et al., 2020; Thurm et al., 2015) while others have not (Latrèche et al., 2024; Yoder et al., 2015). Our findings did not demonstrate such an association despite our relatively large sample, which had larger statistical power than most previous studies. There may be several reasons for these differences across studies. First, some of the studies relied on parent reports to estimate autism severity and/or language abilities (Anderson et al., 2007; Kilili-Lesta et al., 2024; Latrèche et al., 2024; Mouga et al., 2020), which may introduce differences in reporter bias. Second, the type of assessment used to estimate cognitive abilities differed across studies. We used the nonverbal subscale of the Bayley-III or MSEL at Time 1, excluding all language-dependent items, while others used total cognitive scores that include language-dependent items, which are inappropriate for assessing the cognitive abilities of MV children (Anderson et al., 2007; Ellis Weismer et al., 2010; Yoder et al., 2015). Nevertheless, Thurm et al. (2015), did report that a nonverbal cognitive score (MSEL NVDQ) predicted expressive language development in their sample. Hence, there remains some controversy regarding the potential ability of non-verbal cognitive scores to predict later expressive language development in MV autistic children.

In contrast, there seems to be a consensus that the severity of early RRB symptoms is not associated with later expressive language development in MV autistic children. This was clear in our results as well as in those of multiple previous longitudinal studies (Bal et al., 2020; Ray-Subramanian & Ellis Weismer, 2012; Thurm et al., 2015; Troyb et al., 2016). Consequently, these findings suggest that RRB behaviors do not interfere with the acquisition of expressive language in MV autistic children.

Limitations

We note several limitations in the current study. First, we did not have detailed information regarding the type, frequency, or intensity of the specific interventions received by each child. We relied on parent-reported duration in a special education setting as a proxy measure for the extent of intervention received by the child given that in the Israeli system, special education preschools typically provide more intensive and comprehensive intervention services than mainstream preschools. However, this is a gross estimate that does not necessarily quantify of the actual intervention received. Second, all our predictive measures were entirely based on clinical observations using the ADOS-2 and the Bayley III and MSEL cognitive assessments. Adding parental reported measures would have been informative in determining the relative predictive strength of parent reported measures versus clinical observation measures. Third, our study lacked a formal standardized language assessment that would have been important for assessing more refined expressive language changes over time.

CONCLUSIONS

Our findings demonstrate that preschool MV autistic children with better social abilities and particularly JA abilities are more likely to succeed in developing expressive language regardless of their initial non-verbal cognitive abilities or RRB symptom severity. Notably, our study offers a unique contribution by examining these processes within a large, representative, community-based sample (n = 99) followed longitudinally from the point of diagnosis. We show that JA scores derived from the ADOS-2 items specified by pointing, gesturing, showing, unusual eye contact, initiating JA, and reacting to JA, offer a simple, widely available measure that is predictive of later language development.

AUTHOR CONTRIBUTIONS

Tanya Nitzan: Conceptualization; data curation; methodology; formal analysis; visualization; writing—original draft; investigation. **Moran Bachrach**: Conceptualization; methodology; data curation; formal analysis; writing—original draft; investigation. **Michal Ilan**: Investigation; writing—review and editing. **Michal Faroy**: Investigation; writing—review and editing. **Danel Waissengreen**: Investigation; writing—review and editing. **Analya Michaelovsky**: Investigation; writing—review and editing. **Dikla Zagdon**: Investigation; writing—review and editing. **Yair Sadaka**: Investigation; writing—review and editing. **Omer Bar Yosef**: Investigation; writing—review and editing. **Ditza Zachor**: Investigation; writing—review and editing. **Einat Avni**: Investigation; Writing—review and editing. **Idan Menashe**: Writing—review and editing; funding acquisition; resources. **Gal Meiri**: Investigation; writing—review and editing; funding acquisition; resources; supervision. **Judah Koller**: Supervision; writing—review and editing. **Ilan Dinstein**: Conceptualization; methodology; supervision; funding acquisition; resources; writing—review and editing.

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CONFLICT OF INTEREST STATEMENT

The authors declare no conflicts of interest.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.

ETHICAL CONSIDERATIONS

The study received ethical approval from the Helsinki Committee at Soroka Medical Center (SOR-222-14; original approval date: December 22, 2014; current approval valid until January 23, 2026). Since this was a retrospective study of anonymized data, there was no need to obtain signed consent from the participating families (exempt status).

ORCID

Tanya Nitzan https://orcid.org/0000-0003-1263-3262
Idan Menashe https://orcid.org/0000-0003-1961-1461
Judah Koller https://orcid.org/0000-0001-9342-9751

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