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Spoken language outcomes in limited language preschoolers with autism and global developmental delay: RCT of early intervention approaches

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Abstract

Preschool autistic children with significant global developmental delays and very limited language skills are at high risk for remaining minimally verbal at entry into primary school. This study compared two early intervention models for improving social communication and spoken language outcomes in 164 children who received intervention in their community preschool program for six months, with a six-month follow-up. The primary outcome measure was a standardized language assessment, and secondary measures focused on social communication. Results indicated children on average made six months gain in language development in the active six months of intervention with no difference between intervention models. Children who initiated joint attention more frequently, or who had higher receptive language at baseline made more progress if assigned to receive JASPER, a naturalistic developmental behavioral intervention. Children who received Discrete Trial Training made greater spoken language progress from exit to follow-up. These findings suggest that progress can be made in autistic children who have very little spoken language and who receive targeted early interventions. Individual trajectories vary and depend in part on initial abilities in social communication and receptive language. Future research might consider methods to systematically personalize approaches to fit child characteristics and family preference.

Lay Abstract

This study compared two different early intervention approaches for teaching spoken language to minimally verbal, globally delayed autistic preschoolers. Children were given an hour of therapy daily for 6 months and then reassessed 6 months later. The majority of the 164 participants were from historically excluded populations (low income and minority), and therapy was delivered in school community settings by expert clinicians. Results indicated that the participants made

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significant progress regardless of intervention approach: six months gain in standardized language scores over six months, but slower progress during the period after therapy ended. Children who initiated joint attention more frequently, or who had higher language understanding at baseline made more progress if assigned to receive JASPER, a naturalistic developmental behavioral intervention. Children who received Discrete Trial Training made greater language progress during 6-month period after therapy ended. These findings suggest that progress can be made in children with ASD who have very little spoken language and who receive targeted early interventions.

Keywords

RCT; JASPER; DTT; developmental delay; minimally verbal; preschoolers; language

Significant progress has been made toward elucidating the core features of autism and identifying effective interventions (Smith & Iadarola, 2015). Along these lines, quite different views are held about the nature of autism and active ingredients in intervention. In one of these views, autism is considered to be a disorder of discrimination learning. From this perspective, a major active ingredient involves teaching discriminations between stimuli and providing systematic reinforcement (Smith, 2001). This leads to a highly structured, adult-led intervention carried out in a controlled setting to reduce distraction. The main instructional method is Discrete Trial Training (DTT), which is anchored in applied behavior analysis (ABA). Through task analysis, treatment targets and activities are simplified to maximize successful responses (Smith, 2001).

Another view emphasizes that autism is characterized by core difficulties in social communication and interaction. In contrast to DTT, interventions based on this view usually utilize a blend of intervention strategies from applied behavior analysis and developmental sciences, known as Naturalistic Developmental Behavioral Intervention (NDBI; Schreibman et al., 2015). One NDBI, Joint Attention, Symbolic Play, Engagement & Regulation-JASPER (Kasari et al., 2006; 2021) focuses on increasing social communication skills by emphasizing symbolic processes involving play and language, social processes involving joint attention and affect sharing, and communication initiations and reciprocity. Principles of ABA are used but in contrast to DTT, a much greater emphasis is placed on the use of natural reinforcers, looser control over discriminative stimuli, and continuous back-and-forth interactions rather than separate trials.

DTT-based intervention programs were the first (and, for many years, the only) interventions for preschoolers with autism to be tested in group studies (Lovaas, 1987; Smith et al., 2000). As such, they came to be regarded as having the strongest empirical support and are now often considered the standard of care (Unumb, 2013). Children with autism can make significant gains in cognitive and academic skills in DTT-based ABA intervention (Weitlauf et al., 2014), but approximately 20% make no developmental gain, and another 50% make only moderate gains, not reaching normative standards despite many hours per week of individual instruction over 2 years (Eldevik et al., 2010). In addition, although much research shows that DTT can improve language skills including receptive understanding and expressive labeling of objects (Wong et al., 2015), there is little evidence that DTT increases social communication (Howlin et al., 2009).

Both DTT-based interventions and NDBIs have theoretical and empirical value, but there has not yet been a controlled study to assess whether the intervention methods and treatment content are more efficacious for teaching social communication and spoken language in children with more significant delays. This comparison may be especially important for autistic preschoolers who have extremely limited language since many “pre-verbal” preschoolers (30–50%) do not go on to develop functional speech by age 5–6 years and are re-classified as “minimally verbal” (Tager-Flusberg & Kasari, 2013), never moving past 20 functional, non-echoed words. We refer to these children as having “limited language” because they meet similar criteria to ‘minimally verbal’ or ‘non-speaking’ school-aged children, but at their young age are still considered preverbal with the potential for avoiding a minimally verbal trajectory. These children also differ from many preverbal preschoolers in that they often test in the severe or profound ranges of intellectual abilities and may experience significant global developmental delays.

To begin pinpointing active ingredients of early intervention that lead to improved acquisition of spoken language and social communication for autistic preschoolers with limited language and significant global developmental delays, this three-center randomized clinical trial (RCT) compared two 6-month intervention conditions, DTT and JASPER. We hypothesized that JASPER would lead to greater change than DTT in improving spoken language (primary aim) and core early childhood skills including (a) initiations of joint attention (IJA) and (b) level and diversity of play skills (secondary aims) at the end of intervention and at six-month follow-up. We also hypothesized that DTT would be more efficacious on standardized cognitive scores, a primary outcome of previous studies. Additionally, we explored potential moderators of the effect of interventions, specifically whether baseline IJA and receptive language skills would be associated with language progress with the hypothesis that more IJA and greater receptive language would result in faster spoken language progress. Child’s age at baseline was also explored as a moderator of intervention effects. An exploratory goal was to determine whether improvements in levels of spoken language would be differentially influenced by the interventions, with the ultimate goal to determine the percentage of children who could achieve phrase speech of 3–4 -word sentences by age 5–6 years (and therefore not be considered ‘minimally verbal’).

Methods

Participants

Children were recruited from three US metropolitan sites. Children were between the ages of 33 and 54 months, met criteria for autism confirmed by the Autism Diagnostic Observation Schedule (ADOS 2: Lord et al., 2012), demonstrated fewer than 30 spontaneous words across entry assessments (ADOS 2, MSEL, Reynell), received *at least 12 hours* of intervention per week (caregiver reported), and had a minimum 12-month age equivalent score on at least one of the following assessments: (a) Mullen Visual Reception Subscale, (b) Mullen Receptive Language subscale, or (c) Reynell Verbal Comprehension subscale. Children were excluded if they presented with other significant medical conditions (e.g., Down Syndrome), scored greater than 24-month age equivalent Reynell expressive language score, or were exposed to English <50% of the time.

Altogether, 194 children were screened, with 164 eligible children randomized to intervention (DTT n= 82; JASPER n=82; see Figure 1). Three children exited early, immediately after randomization, and four children exited before the end of intervention. The children's mean age was 44.5 months (*SD*=5.4 months), with significant global developmental delays (MSEL mental age approximately 20 months, developmental quotient of 45, and Vineland adaptive behavior composite (ABC) of 65) and Reynell expressive language age of 16–17 months, see Table 1. This diverse sample included families who self-identified as non-Hispanic Caucasian (40.85%), mixed raced (24.39%), Hispanic (20.12%), Asian (17.68%), African American (15.85%), and Pacific Islander (1.22%). In addition, 30% (n=48) of the children had no words, 53% (n=86) demonstrated single words, and 17% (n=27) had some 2-word combinations. No children spoke with phrase speech of 3–4 word sentences (Tager -Flusberg et al, 2009). Bivariate tests (Wilcoxon, χ^2, and Fisher's exact tests) showed no significant difference between groups in baseline child characteristics (see Table 1).

Ethical approval was obtained through Institutional Review Boards at all three sites. Caregivers gave written consent for their children's participation. The study was registered at ClinicalTrials.gov (identifier: NCT01018407).

Sample Size

Using a 2-sided, 2-sample t test with a type I error rate of 5%, and assuming an attrition rate of 10% at follow-up, the planned total sample size for this study was n=192 (96 in each group) to detect a moderate effect size of 0.5 in primary outcomes with at least 80% power. Due to difficulties in recruiting participants who met the inclusion criteria, it was not possible to meet the total planned sample of 192 children. However, attrition was lower than expected (<10%) with only four children leaving intervention prior to exit and five additional children who were unavailable for 6-month follow-up.

Randomization and Study Design

Conducted by an independent data-coordinating center, participants were randomized within each site and with equal allocation to intervention group. Adverse events were tracked with oversight by an independent Data Safety and Management Board. Two serious adverse events were noted, neither related to the study interventions (one child died in a swimming accident and one child was hospitalized following a home accident).

Intervention

All children were seen in their community preschool programs with research team members (research assistants, graduate students, post-doctoral fellows) as the interventionists. Children in both intervention groups received five hours per week of individual instruction for four months (one 60-minute session per day, five days per week) occurring at the child's community setting, typically a publicly- funded preschool where they received between 12 and 25 hours per week of interventions (most based in ABA). School-based intervention sessions were faded to three times a week in month five and twice a week in month six. During these final two months, caregiver coaching began in the home. Caregiver coaching in the allocated DTT or JASPER intervention was provided following manualized protocols for one hour per week for a total of eight sessions. These coaching sessions were designed to support caregivers' adoption of the intervention strategies and promote generalization of the children's skills to the home setting.

Discrete Trial Teaching (DTT: Smith, 2001)—DTT emphasizes didactic, adult-led instruction and relies on teaching discriminations between stimuli, responses to stimuli, and providing systematic reinforcement for correct responses (Smith, 2001). The goals of DTT are to teach specific skills, to accelerate overall development and increase school readiness (Smith, 2001). Interventionists followed a curriculum map specifically focused on early social, communication and language targets (Smith, 2001) with supervision provided by DTT supervisors at site 2. Targets included imitation, play, matching, requesting, comprehension, learning readiness, and labeling. Children in DTT completed an average of 75.51% of all sessions (*SD*=13.68%)

Joint Attention, Symbolic Play, Engagement, & Regulation (JASPER: Kasari et al., 2021)—JASPER is a developmentally anchored behavioral intervention that uses the child's current play level to choose appropriate toys and materials to create a context for learning. Developmental principles and strategies are organized into seven subscales including environmental arrangement, balancing imitation and modeling, establishing play routines, responding to and expanding children's nonverbal and spoken social communication, programming for joint attention and requesting, expanding play routines, and supporting children's regulation and engagement (Kasari et al., 2021). JASPER supervision was provided by certified JASPER supervisors from site 1. Children in JASPER completed an average of 78.38% of all sessions (*SD*=10.97%)

Training and Implementation Support for Interventionists—Interventionists (graduate students and staff research assistants) were trained to fidelity prior to working with study participants. Weekly supervision was provided through cross site clinical calls (one each of JASPER and DTT) directed by the PI/Study Coordinators. Implementation fidelity was measured using scales specific to each intervention (Kasari et al., 2021; Smith, et al, 2000). Additional individualized support and booster training was provided to any interventionist who fell below 80% implementation fidelity. Twenty percent of the sessions across sites, participants, and study months were reviewed for implementation fidelity. Average fidelity for DTT was 92.6% (SD=9.32%) and for JASPER was 83.7% (SD=11.6%).

Measures and Outcomes

Primary Outcome: Expressive Language—Reynell Developmental Language Scales (Reynell: 1997) was selected as the primary measure of expressive language (standardized for children aged 1–7 years). The expressive language subscale was administered at entry, exit, and 6-month follow-up. Although used in a previous study of JASPER (Kasari et al, 2008), this sample demonstrated significant floor effects with over half scoring (55%−60%) below the floor of 12 months comprehension, and 14 months expressive language at baseline. Due to these floor effects, analyses relied on raw scores corresponding to the number of correct responses.

Mullen Scales of Early Learning (MSEL; Mullen, 1995), a standardized test of development includes subscales for fine motor, visual reception, receptive language, and expressive language. Children's age equivalent scores were applied to study eligibility (visual reception subscale age equivalent score of ≥12 months). MSEL mental age is defined as the average of all four subscales' age equivalency scores. The MSEL was administered at screening/entry, exit, and 6-month follow-up.

Natural Language Sample from Caregiver Child Interaction (CCX; adapted from Kasari et al., 2014), was used to capture children's verbal and nonverbal communication during the 10-minute video recording of caregiver child interactions. Caregivers were provided with a standard set of toys and asked to play with their child as they normally would. Interactions were transcribed, verified by separate raters, and child's utterances coded for spontaneity, function (joint attention, requesting, other), and form based on protocols previously published (Kasari et al., 2014) using conventions from the Systematic Analysis of Language Transcripts (SALT). Two variables were coded from the transcripts: spontaneous communicative utterances (SCU), sum of all spontaneous utterances and gestures to comment and to request, and number of different word roots (NDWR), all spontaneous unique words. Transcribers and coders were blinded to time point and intervention allocation. Reliability was checked at two levels, transcription and transcription coding. Each transcript was viewed at least twice – once for the actual transcription and coding and once more for verification of transcript and codes by another reliable transcriber. The average intra-class correlation (ICC) for transcript coding was among five coders (NDWR ICC=0.97; SCU ICC=0.99).

Secondary Outcomes

Receptive Language.: Reynell Verbal Comprehension subscale raw scores and MSEL Receptive Language age equivalency scores were used to assess children's receptive language at entry, exit, and follow-up.

Play Skills.: Children's unique play acts with toys were coded from a video recorded semi-structured assessment. During the Structured Play Assessment (SPA: Adapted from Ungerer & Sigman, 1981; Kasari et al, 2010), an independent evaluator presents the child with five sets of toys that are systematically designed to provide opportunities across functional and symbolic play levels. The video is coded by raters who are blinded to intervention allocation and time point. Coders capture the number of different unique play acts with toys (play types) within each of 16 play levels. Inter-rater reliability ranged from ICC=0.61 to ICC=0.94 (average 0.89).

Initiations of Joint Attention (IJA) and Behavior Regulation (IBR).: The type and frequency of children's initiations to socially share (joint attention) and to request (behavior regulation) were captured during the administration of the Early Social Communication Scales (ESCS: Mundy et al., 2003). The ESCS is a semi-structured video recorded assessment. Independent evaluators use a standard set of toys and standard number of opportunities to capture the frequency and form of children's spontaneous (non-prompted) IJA and IBR. Forms of IJA included gaze (coordinated joint looks and alternating gaze), gestures (point, show, give), and commenting spoken language. Forms of IBR included gaze, gestures (reach, give, point), and requesting spoken language. A total count score for each of IJA and IBR was obtained. Four independent coders blinded to time point and intervention allocation completed the coding. Inter-rater reliability was high: IJA (ICC=0.82), IBR (ICC=0.94).

Child-Initiated Joint Engagement.: Children's time spent in joint engagement with their caregiver was coded from the CCX, including **child-initiated joint engagement** (e.g., child initiates the interaction) and adult directed joint engagement (the adult recruits, instructs or otherwise directs the child's participation and compliance). Our goal was to support social engagement rather than child compliance; thus the outcome measure was child-initiated joint engagement. ICC coding reliability averaged 0.75 across 4 raters (range 0.67–0.96).

Determination of Phrase Speech at Follow-up.: One goal of this study was to determine progress towards phrase speech by age 5–6 years. Speech status (no words, single words, word combinations and phase speech of 3–4 word sentences) was determined by examination of word count on three different language measures, Reynell, MSEL, and ESCS. "No words" was the lowest level and "phrase speech" was the highest level. Agreement was required on at least two of the measures. If there were questions about total word count across the assessments (e.g., inconsistency) then other assessments were reviewed, including the SPA and Language sample from the CCX.

Statistical Analysis

Generalized linear mixed models (GLMM) with log link function were applied for count outcomes and identity link function were applied for Gaussian outcomes. For the analysis of speech status, an ordinal logistic mixed model was utilized. For each outcome, we first fit a model that estimated the overall site effect. The model included the main effect of intervention, time, site, and their two- and three-way interactions. A site difference was defined as significant differences in the intervention effect by site across time. If the site difference was found to be significant, post-hoc contrasts were applied to determine the effect of each intervention and intervention differences within each site and across time. If the site difference was not significant, all site interactions were dropped from the model. Similarly, moderator effects were examined as three-way interactions among intervention, time, and baseline moderators. Only significant moderator effects are presented in the results section and insignificant moderator effects (i.e., age) are not included for brevity.

Between-group differences in the intervention effect and maintenance effect were defined as significant differences in (a) the predicted means between entry and exit (i.e., intervention phase) and (b) between entry and the 6-month follow-up between groups, respectively. All models included child mental age equivalent at baseline with subject level random child level intercepts and unstructured covariance structure. Time is modeled such that the rate of improvement (slope) over the intervention phase (entry to exit) is allowed to differ from the rate of improvement in the follow-up phase (exit to the 6-month follow-up) by applying a piecewise linear model as children may change in their trajectories post treatment phase. Significance is determined by p-values less than 0.05. Effect sizes (ES) are reported as Cohen's f for F tests and Cramer's Phi for χ^2 tests where effect sizes of 0.1, 0.25, and 0.4 are generally regarded as small, moderate, and large (Cohen, 2016).

A hurdle model was applied to IJA. A hurdle model is a modified count model in which there are two processes, one generating the zeros and one generating the positive counts. The binary process models whether the count outcome has a zero or a positive value. If the count is positive (i.e., crossing the hurdle) then the conditional distribution of the positive counts is assumed to be zero-truncated Poisson. We modeled the binary and zero-truncated Poisson processes using the same set of predictors for ease of interpretation.

Results

Primary Outcome

Reynell: Expressive Language Subscale.—Contrary to hypotheses, children in both intervention groups improved significantly in expressive language raw scores from baseline to exit (F(1,308)=165, p<0.001, ES=0.73) and from baseline to follow-up (F(1,308)=237.5, p<0.001, ES=0.87). There was no difference in improvement by intervention group (χ^2(4)=0.9,p=0.754, ES=0.04).

Due to floor effects in Reynell scores (our primary outcome measure) we examined other measures of expressive language for sensitivity to intervention differences including the MSEL and Language Sample scored from caregiver-child interactions.

MSEL: Expressive Language Subscale.—Children in both intervention groups improved significantly in MSEL expressive language age equivalent scores measured from baseline to exit (F(1,309)=181.65, *p*<0.001, ES=0.77) and from baseline to follow-up (F(1,309)=344.90, *p*<0.001, ES=1.06).

The MSEL did yield a significant intervention by site difference in expressive language age equivalent scores baseline to exit (χ^2(24)=37.8, p=0.036, ES=0.10). Children receiving DTT at site 3 improved significantly more in expressive language (6.4 months improvement) than children in JASPER (3.3 months improvement) (F(1,155)=4.09, p=0.045, ES=0.16); no differences were noted between groups in sites 1 and 2. At follow-up, children across all sites improved from an average of 15.4 months in expressive language at baseline to 21.9 months at exit and to 24.7 months at follow-up.

Language Sample (SCU and NDWR) during Caregiver Child Interaction.—Children from both intervention groups improved in SCU and NDWR from baseline to exit (F(1,585)=46.40, p<0.001, ES=0.28; F(1,585)=67.54, p<0.001, ES=0.34 respectively) and from baseline to follow-up (F(1,585)=58.37, p<0.001, ES=0.32; F(1,585)=130.54, p<0.001, ES=0.47 respectively). There was no significant intervention difference in SCU from baseline to exit (F(1,585)=0.45, p=0.505, ES=0.03) and from baseline to follow-up (F(1,585)=3.23, p=0.073, ES=0.074). There was a moderate intervention effect on NDWR from baseline to exit (F(1,585)=3.98, p=0.05, ES=0.08) where children receiving DTT gained 1.5 more NDWR compared to children in JASPER.

Secondary Outcomes

Reynell: Verbal Comprehension Raw Scores.—Children in both groups significantly improved in raw scores from baseline to exit (F(1,308)=156.78, p<0.001, ES=0.71) and follow-up (F(1,308)=284.22, p<0.001, ES=0.96); the groups did not differ from each other from baseline to exit or follow-up (F(1,308)=1.11, p=0.293, ES=0.06; F(1,308)=1.69, p=0.195, ES=0.07).

MSEL: Receptive Language.—Children in both intervention groups improved significantly in receptive language age equivalents from baseline to exit (F(1,309)=155.05, p<0.001, ES=0.71) and from baseline to follow-up (F(1,309)=366.52, p<0.001, ES=1.09). There was no significant intervention difference by site (χ^2(24)=8.5, p=0.998, ES=0.05) and no overall significant intervention difference in receptive language (F(1,309)=1.06, p=0.304, ES=0.06) from baseline to exit. There was a significant intervention difference in receptive language age equivalents from baseline to follow-up F(1,309)=4.13, p=0.043, ES=0.11) where children in the DTT group (19.57 months at baseline to 31.38 months at follow-up) gained an average of 3.1 months more than children who received JASPER (19.93 months at baseline to 29.48 months at follow-up).

ESCS: IJA and IBR.—The overall treatment difference (averaged across sites) for IJA was significant in the truncated Poisson model (F(1,159)=4.44, p=0.037, ES=0.17), but not in the binary model (F(1,159)=0.07, p=0.793, ES=0.02). This showed that of those children who demonstrated at least one IJA skill at baseline (i.e., those who crossed the hurdle) and who received JASPER made significantly more improvements in IJA from baseline to exit than children who received DTT, averaged across sites. In addition, on average, children in both intervention groups increased in IBR from baseline to exit (F(1,149)=18.41, p<0.001, ES=0.35) but not differentially by intervention group (F(1,149)=3.49, p=0.064, ES=0.15).

SPA: Total Play Diversity and Play Level.—Children's total play diversity increased from baseline to exit (F(1,298)=24.83, ES=0.29) and from baseline to follow-up (F(1,298)=42.87, p<0.001, ES=0.38). There was no overall difference between interventions across sites (F(4,290)=1.15, p=0.335, ES=0.12) or difference in the rate of improvement in play diversity between intervention groups from baseline to exit (F(1,298)=1.45, p=0.230, ES=0.070) and from baseline to follow-up (F(1,298)=2.89, p=0.090, ES=0.098).

Children in both intervention groups improved in play level from baseline to follow-up (F(1,298)=32.78, p<0.001, ES=0.33). There was no intervention difference (F(1,154)=1.34, p=0.249, ES=0.09) or site difference (F(2,150)=0.51, p=0.602, ES=0.08) during the intervention phase (baseline to exit). However, children who received DTT made greater improvements in play level at follow-up compared to the children who received JASPER (F(1,298)=4.96, p=0.027, ES=0.13).

CCX: Child-Initiated Joint Engagement.—There was an overall improvement in child-initiated joint engagement for all children (F(1,576)=22.44, p<0.001, ES=0.20), with no significant intervention difference ($F(1,576)=0.01$, $p=0.910$, ES=0.004). MSEL receptive language moderated the effect of intervention on child-initiated joint engagement ($F(1,573)=5.15$, $p=0.024$, ES=0.09). Children in JASPER who began with higher receptive language improved more in child-initiated joint engagement from exit to follow-up compared to children in JASPER who had low receptive language. However, there was no difference in improvements in engagement for children in DTT by level of receptive language.

Speech Status.—Children who use phrase speech are no longer considered non-speaking or minimally verbal (Tager-Flusberg & Kasari, 2013). In this study, we determined the percentage of children who went on to use word combinations and those who went on to phrase speech by intervention exit or follow-up. No words was the lowest level and phrase speech was the highest.

Children in both intervention groups significantly improved in speech status from baseline to exit ($F(1,309)=44.69$, $p<0.001$, ES=0.38) and follow-up ($F(1,309)=54.89$, $p<0.001$, ES=0.42) (see Table 2). There were no significant differences by intervention group from baseline to exit or follow-up ($F(1,309)=0.15$, $p=0.699$, ES=0.02; $F(1,309)=0.22$, $p=0.640$, ES=0.03).

At baseline (combined across treatment groups), 32% of the children demonstrated no words, 54% had single words, 14% had word combinations (these were generally carrier phrases “I want” considered one word, plus noun) and no child demonstrated phrase speech of 3–4-word sentences. By exit, the number of children demonstrating phrase speech increased to 27.3%, while 17.4% showed word combinations, 40.4% single words only, and 14.9% remained with no words (see Table 3). Moreover, even for children who did not go on to phrase speech, improvements were made in both SCU and NDWR. For children who began the study with no words, SCU increased from 0.173 (entry), 2.714 (exit), to 4.068 (FU). Their NDWR increased from 0.23 entry, 2.22 (exit), to 4.045 (FU). For children who began with single words, SCU increased from 6.34 (entry), 11.012 (exit), to 14.89 (FU). Their NDWR increased from 5.79 entry, 11.025 (exit), to 17.08 (FU).

MSEL cognitive scores.—Contrary to hypotheses, children receiving DTT did not demonstrate higher scores on the nonverbal cognitive subscales of MSEL (visual reception and fine motor subscales combined: Non-verbal mental age) compared to children receiving JASPER, despite DTT direct targeting of skills on the MSEL (Non-verbal mental age - DTT: 24.9 at baseline to 32.5 24 at exit to 35.5 at follow-up; JASPER: 24.7 at baseline to 30.1 at exit to 34.at follow-up; $F(1,310)=2.81$, $p=0.095$).

Discussion

This is one of the first community-based, partial effectiveness (researcher implemented in a community site) studies comparing two disparate yet efficacious interventions (i.e., DTT and JASPER) for autistic children who have limited language and significant global developmental delay. As the presence of severe developmental delays has been widely considered a harbinger for less favorable outcomes, exploration of intervention efficacy specifically within this population is critical. Data on the primary research question (whether children experienced change in expressive language skills over the course of six months of DTT or JASPER) revealed that high-quality intervention resulted in language gains – *regardless of intervention type*. Namely, across both groups, children made an average of 6 months' progress on standardized language assessments. These improvements translated into meaningful clinical outcomes as well; nearly half (45%) of all participants were speaking in word combinations or phrase speech at the end of one year, thus moving away from a designation of minimally verbal (defined as fewer than 20 single words) by their entry to school. The subset of participants who spoke in only single words by the 1-year follow-up still demonstrated progress from baseline of no words or single words, with an average of 4 to 17 words gain. Improvements were greater from entry to the 6-month intervention timepoint (exit) than from the 6-month exit to follow-up. As such, the importance of active, ongoing evidence-based intervention is highlighted, especially for children who have minimal language at the onset.

DTT and JASPER showed similar efficacy for promoting spoken language in this population, and contrary to our hypothesized expectations. While no children spoke in phrase speech at the beginning, just under a quarter spoke in phrases/sentences by intervention end (6 months). At follow up a year post baseline, about a third spoke in phrases, and nearly half had at least word combinations. While both interventions were effective, there were some differential trends. DTT resulted in an average of 1.5 more unique words (NDWR) at exit compared to JASPER although this difference did not maintain at follow up. DTT at follow up yielded greater increases in play level, contrary to expectations. However, more in line with our hypotheses, two baseline factors moderated intervention effects on social communication outcomes. Social communication outcomes were greater for children receiving JASPER when they (a) began intervention with at least one instance of IJA during baseline assessments resulting in greater joint attention at exit and (b) had higher receptive language skills at baseline resulting in greater child-initiated joint engagement. A unique feature of JASPER is the inclusion of both IJA and joint engagement as specific intervention targets, which may have allowed children to capitalize upon foundational social-communication skills at baseline. These findings are in line with research on children with higher and more varied language skills at baseline (Shih et al., 2021).

While our hypothesis that children receiving DTT would make greater gains in cognitive scores (here measured by the combined fine motor and visual reception subscales of the MSEL) was not confirmed, receptive scores on the MSEL improved more for children receiving DTT than JASPER at the follow up. It may be that children who had DTT during the active treatment phase were better able to continue to access strategies taught in DTT such as direct instruction on object recognition; most children received DTT or direct instruction methods as another form of intervention outside of the study, while no child had access to JASPER after the active treatment phase. Further, DTT prioritizes teaching of pre-academic skills (e.g., matching, receptive object labels, object imitation), which can be either directly assessed or strongly aligned with success on standardized tests. In addition, the style of instruction in DTT which asks children to respond to specific instructions (e.g., what is it?) is similar to the instructions provided on standardized tests. Thus, standardized cognitive tests may be more proximal to the treatment approach used in DTT than in JASPER.

This study highlights the heterogeneity amongst even a subgroup of autistic children with significant global developmental delays. While more than 30% did transition to phrase speech (thereby avoiding classification as minimally verbal at school age), approximately 70% of this sample continued on a slow trajectory toward spoken language, and developmental progress. Strengths in nonverbal skills and younger age did not predict better or worse outcomes. While important to acknowledge from an efficacy standpoint, within-sample heterogeneity should not overshadow the unifying finding – that some autistic children with significant intellectual impairment can make progress with direct intervention, even when entering with no or very limited spoken language skills.

Limitations to the study design leave us with remaining questions, particularly related to how to support children in the subgroup that did not acquire spoken language. These gaps should inform further empirical expansions that extend our knowledge through the use of novel methodology. For example, in clinical practice, children who are not making progress often receive additional hours of the same intervention. At present, we still have not identified the best way to address limited progress (e.g., increased dose of the same intervention, switching to a different intervention, or some other modification). While the current study attempted to neither measure response during intervention nor change the approach depending on the child’s response, this could be addressed in future work through inclusion of research designs that allow for adaptations based on a child’s response. For example, adaptive designs include assessment of a tailoring variable that gathers information about the participant’s response to intervention at a set point in time (Nahum-Shani & Almirall, 2019). Based on the child’s response, prespecified decision rules are employed to guide the next intervention option(s). Our current findings suggest promise for adaptive designs with this population to determine if better outcomes could be achieved for the large proportion of children still not fluent in spoken language. Further, interventions with minimally verbal older children suggest benefits to using alternative, augmentative communication supports within the context of behavioral interventions (e.g., Kasari et al, 2014). This study did not use AAC supports, and no child was using AAC or picture support systems prior to entering the study. These may be important to include in future studies with this age group of children.

While the lack of a control condition could be seen as a limitation, this study intentionally favored a comparative efficacy design over comparison with “business as usual” for two reasons. First, we reasoned that providing intervention to both groups, particularly in a school setting, would be preferable to families and teachers in the community. Second, this study prioritized young children with limited language and significant delays – a group that is often excluded from research – and we were therefore reluctant to withhold intervention to children who had such high need for support.

In addition to extending traditional research samples to children with minimal language, this study notably included a relatively large sample within autism research (n=164). Participants who are often under-represented in research based on income and race/ethnicity were wellrepresented, with the majority identifying as non-White and resource limited attending Title 1 public school programs (these are school programs where large percentages of the student body have low income and qualify for free and reduced meals). The provision of services within community school settings likely removed common barriers to participation in research. Such strategies are necessary to ensure adequate representation within samples.

In summary, the results of this study suggest that children who are limited in spoken language (and also have significant global delays) can make progress in developmental spoken language outcomes with evidence-based intervention (here, DTT or JASPER). Demonstrated gains included increases in language skills on standardized and observational assessment as well as overall speech status, with over 30% of the children transitioning to the use of phrase speech. Even so, their language levels and rate of language development fell below expected age levels given their nonverbal cognitive performance, highlighting the need for ongoing intervention. This rigorously executed study in a representative community sample of children demonstrates that multiple interventions may be effective and not all children respond similarly to a single intervention. To advance precision in matching children with the most effective programs, future studies must systematize and personalize interventions in ways that are tailored to child progress and considerate of child characteristics and family preferences.

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Data Availability Statement

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

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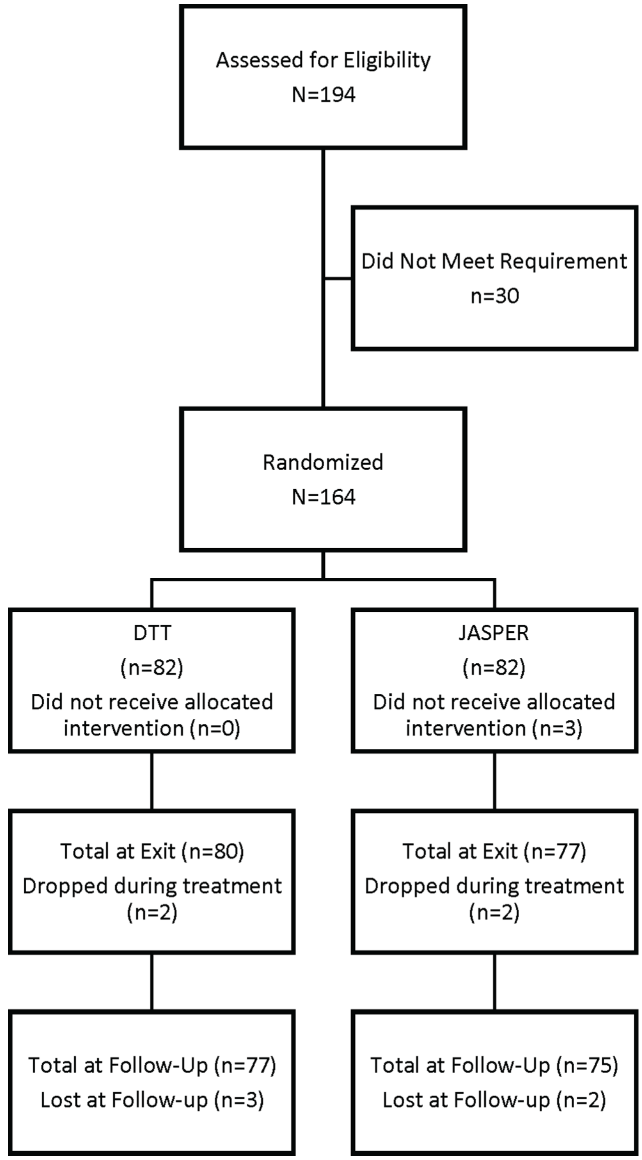

Figure 1.
CONSORT Flow Diagram

Table 1

Child Characteristics at Baseline

Child Characteristics: Mean (SD)	DTT (n=82)	JASPER (n=82)	*p*-value
Male: n (%)	70 (85%)	67 (82%)	0.674
Age (Months)	44.33 (5.45)	45.09 (5.23)	0.279
Ethnicity: n (%)			0.243
Hispanic	20 (24%)	13 (16%)	
Not Hispanic	62 (76%)	69 (84%)	
Race: n (%)			0.680
Asian	15 (18%)	14 (17%)	
Native Hawaiian/ Pacific Islander	2 (3%)	0 (0%)	
African American	11 (13%)	15 (18%)	
Caucasian	35 (43%)	32 (39%)	
Other/Mixed/Not Disclosed	19 (23%)	21 (26%)	
ADOS Calibrated Severity Score	7.32 (1.61)	7.40 (1.65)	0.633
Vineland Adaptive Behavior Score	65.99 (7.34)	65.41 (9.32)	0.478
MSEL (Age Equivalent)			
Visual Reception	24.77 (7.06)	24.49 (7.22)	0.774
Fine Motor	25.06 (5.83)	25.01 (6.80)	0.682
Receptive Language	16.15 (7.96)	16.38 (8.44)	0.941
Expressive Language	15.17 (7.08)	15.66 (6.85)	0.635
Overall Mental Age	20.29 (5.88)	20.38 (6.42)	0.933
Non-verbal Mental Age	24.91 (5.83)	24.75 (6.63)	0.741
Developmental Quotient	45.81 (12.16)	45.43 (13.84)	0.822
Non-verbal Developmental Quotient	56.44 (12.32)	55.36 (14.58)	0.533
Reynell			
Verbal Comprehension			
Raw Score	10.85 (8.97)	10.32 (8.82)	0.556
Standard Score	63.91 (0.8)	63.91 (0.91)	0.731
Expressive Language			
Raw Score	11 (6.05)	11.35 (7.09)	0.890
Standard Score	63.77 (0.76)	63.60 (0.72)	0.137
Mother's Education: n (%)			0.471
Less than 7th Grade	0 (0%)	1 (1.22%)	
Junior High	2 (2.44%)	0 (0%)	
High School or Less	15 (18.29%)	20 (24.39%)	
Some College or Special Training	23 (28.05%)	24 (29.27%)	
College Graduate	30 (36.59%)	22 (26.83%)	
Graduate/Professional Training	11 (13.41%)	12 (14.63%)	
Do Not Wish to Disclose	1 (1.22%)	3 (3.66%)	
Mother's Current Employment: n (%)			0.699
Not Employed	32 (39.02%)	40 (48.78%)	

Child Characteristics: Mean (SD)	DTT (n=82)	JASPER (n=82)	*p*-value
Self Employed-Part Time	6 (7.32%)	5 (6.10%)	
Self Employed-Full Time	0 (0%)	1 (1.21%)	
Employed	12 (14.63%)	8 (9.76%)	
Employed, Full Time	29 (35.37%)	25 (30.49%)	
Employed, Full Time + Second Job	1 (1.22%)	0 (0%)	
Do Not Wish to Disclose	2 (2.44%)	3 (3.66%)	
Total Outside Services	2.93 (1.72)	2.92 (1.80)	0.932

Table 2

Percentage of children who gained phrase speech

	DTT			JASPER		
Language Status: n (%)	**Baseline**	**Post Treatment**	**Follow-up**	**Baseline**	**Post Treatment**	**Follow-up**
No Words	27 (32.93%)	10 (12.2%)	8 (10.53%)	24 (30.38%)	14 (17.72%)	14 (18.42%)
Single Words	42 (51.22%)	33 (40.24%)	30 (39.47%)	45 (56.96%)	32 (40.51%)	26 (34.21%)
Word Combinations	13 (15.85%)	18 (21.95%)	16 (21.05%)	10 (12.66%)	10 (12.66%)	13 (17.11%)
Phrase Speech	0 (0%)	21 (25.61%)	22 (28.95%)	0 (0%)	23 (29.11%)	23 (30.26%)

Table 3:

Overall changes in speech status

Language Status at Baseline and Change by Exit: n (%)	Total
No Words at Baseline	**51**
Did not improve by exit	23 (45%)
Improved by exit	28 (55%)
Single Words at Baseline	**87**
Did not improve by exit	42 (48%)
Improved by exit	45 (52%)
Word Combinations at Baseline	**23**
Did not improve by exit	6 (26%)
Improved by exit	17 (74%)