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Mediating Parent Learning to Promote Social Communication for Toddlers with Autism: Effects from a Randomized Controlled Trial

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Abstract

A randomized controlled trial was conducted to evaluate effects of the Joint Attention Mediated Learning (JAML) intervention. Toddlers with autism spectrum disorders (ASD) aged 16–30 months (n = 144) were randomized to intervention and community control conditions. Parents, who participated in 32 weekly home-based sessions, followed a mediated learning process to target preverbal social communication outcomes (social visual synchrony, reciprocity, and responding and initiating forms of joint attention) throughout daily interactions. The analysis found post-intervention effects for all outcomes, with all except initiating joint attention sustaining 6 months post-intervention. Findings support the value of very early intervention targeting explicitly social functions of preverbal communication and of promoting active engagement in the learning process for both toddlers and parents.

Keywords Toddlers · Parent-mediated intervention · Social communication · Joint attention

Introduction

As early as 6 months of age, infants with later-identified autism begin to differentiate from their peers by showing a relative lack of interest in and aversion to social interactions, which will eventually more clearly characterize their differences (Jones and Klin 2013). Understanding the

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early emergence of this core feature of autism allows for the design of intervention programs that are developmentally fitting and naturally situated to strengthen early social competencies and promote their sustained use. When toddlers are identified with autism spectrum disorders (ASD)¹, the privileged parent-toddler relationship and natural interactional learning opportunities in the home setting create prime conditions for early social learning and make parents the natural mediators of such learning. The purpose of this study was to examine the efficacy of home-based parent-mediated early intervention that focuses on promoting early prelinquistic forms of social communication for toddlers with autism.

Social communication difficulty, the principal challenge in autism, appears preverbally in toddlers (Mundy 2016) and is therefore central to decision-making about early intervention content. Subtle signs that appear in early infancy are not reliably distinguishable on an individual basis but become more prominent in toddlers. A first sign of social engagement, visual social synchrony, occurs between typically developing infants and parents in earliest infancy and is associated with later relational competencies (Feldman 2007) while infants who will be later identified with autism tend to show a preference for non-social stimuli and passive insensitivity to partners' social gaze (Klin et al. 2009;



Hereafter referred to as "autism."

Moriuchi et al. 2017). A second and subsequent sign of difficulty is in reciprocal forms of interaction such as social imitation and turn taking with caregivers, abilities that typically develop during the first year and are associated with later language facility (Ingersoll 2008).

A third sign is difficulty with joint attention, which typically develops by the end of the first year as infants share attention about objects while showing social interest in caregiver perspectives. Joint attention, which correlates with later challenges in verbal language, social and cognitive skills, and autism severity (Bottema-Beutel 2016; Charman 2003; Freeman et al. 2015; Gillespie-Lynch et al. 2012; Mundy et al. 2007), is reliably identifiable as absent or compromised in toddlers with autism (Mundy 2016). Compared to typical peers, they are relatively competent in communicating for instrumental purposes by requesting and following directions at the preverbal level (Curcio 1978; Mundy 1995) but are uniquely challenged to initiate or respond to joint attention overtures for social purposes by showing objects to parents or sharing interest in what their parents show them (Adamson et al. 2001).

The research reviewed here illustrates how these three areas of challenge in early autism—social visual orientation, reciprocity, and joint attention—are developmentally ordered and foundational. These findings have implications for selecting early intervention content. Because of joint attention's association with later milestones, a logical focus of early intervention for toddlers with autism would be the promotion of these progressive stages of preverbal social communication.

Reviews of the emerging body of general intervention research for toddler-aged children with autism, including small-scale studies, have indicated generally promising effects for social communication outcomes (Bradshaw et al. 2015; Schertz et al. 2012). Bradshaw's reviewed studies focused on intervention for participants under age 24 months and, of the five group-comparison studies, all except one (Dawson et al. 2010), which is described following, were parent implemented. Main effects on child outcomes were reported for two of the remaining four parent-implemented interventions. In Schertz and colleagues' (2012) reviewed studies, six (several of which were also reviewed by Bradshaw and colleagues) used group designs. Of these, three studies that used parent-implemented interventions found between-group effects ranging from none to large and the three that using professionally implemented interventions reported small to medium effects.

The recently emerging randomized controlled trials (RCTs) of interventions for toddlers with autism have largely used parent-implemented intervention approaches, though they differed in their intervention focus. Several early RCTs focused broadly on general developmental or communication outcomes. Dawson et al. (2010), reporting on

the intensive 2-year professionally implemented (with parent carry-over) Early Start Denver Model, found effects on general developmental and parent-reported adaptive behavior measures. Although cognitive benefits were not maintained at 4-year follow-up, autism symptoms were less marked then in the treatment group (Estes et al. 2015). Of the five additional RCTs that tested effects of parent-implemented interventions on measures of general communication and other outcomes, none reported main effects on child outcomes. One implemented a 12-week low intensity intervention that promoted child-centered responsive parent-child interaction with embedded teaching opportunities (Rogers et al. 2012). Two implemented a short-term play-based intervention (Focused Playtime Intervention focusing on parents' responsive communication) to study effects on child language and joint attention and on observational measures of parent-child interaction in which parents were asked to interact with their children as they normally would (Kasari et al. 2014; Siller et al. 2013). A fourth study implemented a very low-intensity communication-focused intervention to assess communication outcomes using observational measures of parent-child interaction in which parents were asked to engage their children in toy play and book-reading (Carter et al. 2011). Finally, a very low-intensity intervention was implemented to promote compliance, social communication, and language, and assessed outcomes with a parent-report measure (Oosterling et al. 2010).

Two RCTs, which focused more exclusively and directly on parent-mediated promotion of social communication, found effects on measures of social communication. The Early Social Interaction project, which used practicing and problem solving strategies (Wetherby et al. 2014), reported findings on standardized social-communication measures. The Joint Attention Mediated Learning project used mediated learning processes in a pilot RCT and reported social-communication results from observational measures of parent—child interaction in which parents were instructed to play with their children in their typical manner (Schertz et al. 2013).

Because existing measures of early learning vary in their sensitivity to targeted early communication or social changes toddlers with autism, a number of the parent-mediated RCTs reviewed here developed observational measures to assess child changes manifest in parent-child interaction (Carter et al. 2011; Kasari et al. 2014; Oosterling et al. 2010; Schertz et al. 2013; Siller et al. 2013). Together these early RCT findings point to the possibility that focusing early intervention on the core social communication difficulty in autism may produce a different pattern of results than do more generally focused interventions.

Given the early stage of parent-mediated social communication-focused early intervention research and the mixed outcomes across the reported RCTs, additional controlled



research using varied approaches is needed. In particular, given the centrality of the social difficulty across the autism spectrum, research is needed to explore effects of socially focused early intervention on social communication outcomes. In Vygotskian sociocultural theory, the young child learns through a process of internalization from everyday interactions with familiar adults (Vygotsky, 1934/1986), emphasizing the role of social processes in learning and development (Odom 2016). Rogoff (1990) viewed these early learning interactions as participatory apprenticeships as children are guided in a social context to actively build on prior learning towards enhanced competency levels and more complex understandings of the world. For toddlers with autism, the importance of socially embedded developmental learning is evident in the associations of preverbal social communication with the later related outcomes referenced above.

If early learning is socially contextualized and social communication is the primary intervention target for toddlers with autism, a question is how best to build parent capacity to promote active social learning as an alternative to reliance on adult-directed responding and its associated generalizability concerns. The mediated learning approach is based on a theoretical framework that addresses this concern. This framework begins with Vygotskian theory by framing learning as a guided social endeavor (Feuerstein 1980) and has been used to help parents promote infant cognitive learning (Klein 2003). Building on these theoretical bases, the Joint Attention Mediated Learning (JAML) program was implemented in single case design and small RCT studies (Schertz and Odom 2007; Odom et al. 2011; Schertz et al. 2013). JAML is a home-based, parent-implemented early intervention program designed to promote social communication at the preverbal level. It leads toddlers with autism through acquisition of three developmentally sequenced social communicative competencies: focusing on faces, turn taking, and joint attention (both initiating and responding). To promote these outcomes, home visitors work with parents to employ five mediated learning principles in interactions with their children: focusing, organizing and planning, giving meaning, encouraging, and expanding.

Other considerations for intervention process relate to feasibility in typical practice. The Office of Special Education Programs (OSEP) (Workgroup on Principles and Practices in Natural Environments: Part C Settings 2008) views early intervention's primary mission as building upon and supporting families to enhance infant and toddler learning through everyday learning opportunities. For this study, feasibility is defined as the extent to which intervention is implementable in typical or authentic family settings, the reasonableness of training requirements, cost effectiveness, and flexibility for integration within diverse systems—all contributors to scalability. Social validity, or acceptability to

end users of an intervention's goals, activities, and outcomes (Wolf 1978), contributes to implementation fidelity with socially important outcomes and procedures implementable by typical practitioners in authentic settings (Horner et al. 2005). If intervention is to be translatable to the field of practice, these facets of usability require researchers to test its practical application under real-world conditions. As noted, the JAML intervention is intentionally based in the home and implemented in routine family interactions and activities.

Finally, an additional question in this early stage of toddler autism intervention research is whether intervention effects may differentially affect subsets of participants. In the study by Carter et al. (2011), toddlers with lower initial attention to objects showed a stronger response to intervention than toddlers with better initial object attention. Other researchers found that children with lower initial language levels showed greater benefits on language outcomes from intervention that focused on parents' responsive communication (Siller et al. 2013). Questions of conditional intervention effects have implications for how intervention might be differentiated for groups of children depending on age, developmental factors, parent responsivity, or autism severity.

The purpose of this study is to examine the implementation and efficacy of the JAML intervention for promoting early social communication for toddlers with autism. Our specific research questions are:

- 1. What are the effects of the JAML intervention on joint attention (initiating and responding) and earlier forms of social communication for toddlers with ASD in interaction with their parents?
- 2. Do treatment effects maintain after the intervention ends?
- 3. Which, if any, variables moderate the effects of the JAML intervention?
- 4. How do parents assess the social validity of the JAML intervention?

Methods

Participants

Parents and toddlers aged 16–30 months were recruited in metropolitan and rural areas in Indiana, Kansas/Missouri, and North Carolina. Recruitment efforts focused on Part C providers, physicians, diagnostic centers, speech/language clinics, a university research database, and autism publications targeted to families. Eligibility was established if children met each of the following inclusion criteria: scores above the designated cut-off levels on



Table 1 Participant Characteristics at Pre-Assessment

Participant characteristics: M (SD)	Intervention $(n=73)$	Control group (n=71) 24.79 (3.74)			
Age (mo.)	24.55 (4.22)				
Ethnicity (%)					
White	77.46	69.86			
Black	20.55	18.31			
Hispanic non-white	16.44	9.86			
Other	14.08	20.55			
Gender (% male)	79.45	79.45			
Parent age (years)	30.73 (5.91)	32.23 (5.77)			
Parent income (%)					
Less than \$19,091	21.92	14.29			
Between \$19,091 and \$30,971	9.59	4.29			
Between \$30,971 and \$40,000	10.96	10.00			
Between \$40,000 and \$59,000	16.44	24.29			
Over \$60,000	41.10	47.14			
Parent education (%)					
No high school diploma	12.33	2.82			
High school diploma or GED	8.22	11.27			
Some college/no degree	30.14	38.03			
Associate degree	1.37	4.23			
Bachelor's degree	35.62	28.17			
Graduate degree	12.33	15.49			
ADOS-T severity score					
Social affect	16.36 (3.45)	16.99 (3.15)			
RRB	2.36 (1.77)	2.76 (1.72)			
CSBS	49.36 (10.15)	49.42 (9.86)			
MSEL					
Early learning composite	104.48 (35.16)	104.27 (38.14)			
Receptive language	25.27 (9.80)	25.36 (10.26)			
Expressive language	26.73 (9.16)	26.97 (9.82)			
VABS					
Communication	24.75 (7.11)	23.87 (9.16)			
Socialization	32.48 (6.67)	31.21 (5.63)			
RBS-R	0.58 (0.41)	0.66 (0.41)			
MBRS	40.84 (42.80)	38.55 (45.33)			

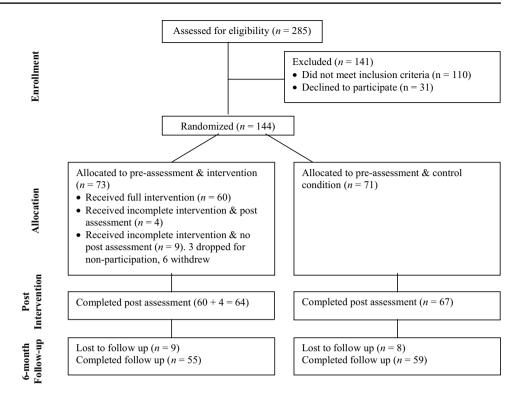
RRB repetitive and restrictive behavior, CSBS Communication and Symbolic Behavior Scales, MSEL Mullen Scales of Early Learning; VABS Vineland Adaptive Behavior Scale, RBS-R Repetitive Behavior Scale—Revised, SALT systematic analysis of language transcripts, MBRS Maternal Behavior Rating Scale—Revised

the Toddler Module of the Autism Diagnostic Observation Scale-II (ADOS-T; Lord et al. 2012), no more than three instances of responding to or initiating joint attention during a 10 min play session with parents in their home, and chronological age below 30 months at evaluation. Children who presented with a confounding condition were excluded from the study (e.g., failure to thrive, premature birth > 6 weeks, or other developmental disabilities such as Down syndrome). Participating families provided prior informed consent in accord with the human subjects protection procedures of each recruitment site's Institutional Review Board. Among toddlers screened, 144 met

inclusion criteria and were enrolled in the study. Parents received a stipend to compensate for time contributed to assessment activities. As participants were determined eligible, they were randomly assigned in a 1:1 allocation to either the JAML intervention or services-as-usual (which we label as "control") condition. Participant descriptions are summarized in Table 1. The mean age of child participants at enrollment was 24.55 and 24.79 for intervention and control groups, respectively, with similar scores between groups on the ADOS-T at entry. Parents were primarily (74%) Caucasian and most (83%) had some education beyond high school.



Fig. 1 Participant enrollment, randomization and retention



Design

This study employed a randomized controlled, intent to treat trial design. As noted, 144 children and their parents were randomized in a 1:1 allocation to either the JAML intervention or the control group. In intent-to-treat (ITT) models, analysis is based solely on group assignment and does not include corrections for changes in the sample (e.g., subject loss, non-compliance, etc.) that take place after participants are assigned to groups. Minor differences in attrition between groups are indicated in the consort diagram (Fig. 1), which illustrates the number assigned to treatment and control conditions and the number observed at each stage. Of the 73 assigned to treatment, 64 remained for post-assessment and 55 for 6-month follow-up assessment. For the 71 assigned to the control condition, 67 remained for post-assessment and 59 for follow-up.

Intervention Procedures

Three intervention coordinators (ICs), one at each site, coordinated the manualized parent-implemented JAML intervention in weekly 1-h home-based sessions for 32 weeks. Families designated a parent to participate in intervention and data collection activities. The IC's role was to facilitate parents' conceptual learning and confidence to mediate toddlers' active engagement using responsive interaction. A facilitative non-prescriptive approach was used to leverage parents' expertise and intimate knowledge of their children.

Parents took a lead role, with support, to translate targeted outcomes and mediated learning principles into parent—child interactional strategies that meshed with child and family interests and priorities. During the week following each 1-h home visit, the parent were expected to promote child engagement in planned play-based interaction for at least 30 min daily in addition to integrating targeted interaction strategies into daily home and family routines.

Both IC support for parent learning and parent facilitation of toddler social communicative engagement were guided by the five mediated learning principles that supported parent and toddler learning, respectively. The principles are defined for parents as "(a) focusing: orienting parents towards salient aspects of parent—child interaction that support the phase of intervention, (b) organizing and planning: promoting a sense of order, (c) giving meaning: helping parents discriminate between aspects of interaction that are more and less effective and relevant to the current intervention emphasis, (d) encouraging: facilitating parents' recognition of their impact on the child's learning, and (e) expanding: engaging parents to add breadth and depth to interaction opportunities" (reference omitted for blind review).

The ICs provided manualized intervention guidance materials to parents in weekly units. The materials contained descriptions and illustrative examples of sequenced social communication content and mediated learning processes. The developmentally and sequentially ordered content consisted of three phases, each with its own sub-units: (1) visually synchronous engagement (focusing on faces),



(2) reciprocal interaction (turn taking), and (3) joint attention (responding to the parent's joint attention overtures and initiating joint attention). Responding to and initiating joint attention were combined in this intervention phase because researchers found in earlier studies (references held for blinded review) that it was more efficient and natural to promote them together rather than as separate intervention goals. In Phase 1 children were encouraged to look freely at the parent in face-to-face interaction. Phase 2 consisted of back-and-forth simple reciprocal interactions in which toddlers, of their own volition, waited for the parent's turn as an indication of reciprocity. In Phase 3, toddlers shared attention with parents in relation to toys or objects. Joint attention (whether initiating or responding) was defined as directing gaze between the object and the parent's face with an indication of social interest, such as smiling.

To monitor child progress, intervention sessions began with parents reviewing weekly activity logs, which were based on the previous week's plans. Parents then engaged their child in a play session emphasizing the targeted social communication outcome. ICs recorded the play session and then the parent and IC watched the video together. The IC then guided the parent's reflection on the child's engagement relative to the targeted outcome and to the parent's use of mediated learning principles. The IC encouraged parents to identify what went well and what was challenging in the recorded interaction. The IC observed for additional positive examples, referred to previous weekly guidance materials and video examples when parents identified challenges, and provided questioning probes to elicit parents' new ideas. The purpose of the guided video reflection was to build on parents' strengths, promoting their initiative and positive views of their own competence rather than relying solely on professional expertise for translating conceptual learning into intervention strategies.

Following the video reflection the IC introduced new material related to the current targeted outcome and related mediated learning principles. For this, the IC drew from the compendium of print materials that were sequenced by targeted outcome and mediated learning principle. Each set of print materials was supplemented by verbal explanations and video clips exemplifying other toddlers with autism demonstrating the current targeted outcome and other parents applying targeted mediated learning principles to promote the current outcome with their toddlers. The purpose of providing examples from other parents' ideas rather than professional modeling was to demonstrate parents' competence in creating activities based on their intimate knowledge of their toddlers' interests and that a wide variety of parent-created activities are appropriate. Examples of shared parent ideas for promoting joint attention using the mediated learning principle giving meaning follows.

- 1. Use a small, lidded tin box to hide a surprise toy or snack. Pull the box close to your face to invite the child's look to your face; then hold it out to her, letting her find the hidden surprise. Then, *give meaning* by making an excited sound to draw her look back to you.
- 2. Use a remote controlled toy, such as a car, and make it go. *Give meaning* to the shared experience by stopping the car and gasping out loud. Wait for your child to look from the toy to your face. When he does, make the car go again, then stop and gasp again. Repeat.
- 3. Play dress-up with silly hats, loud ties, or scarves with interesting textures. When your child looks between the clothing and your face, *give meaning* by smiling and giggling.

In the final segment of the home visit, a joint process of activity planning for the coming week was conducted. ICs and parents identified activities in which mediated learning principles could be applied to promote the targeted phase outcome. ICs provided scaffolding to facilitate active parent involvement in this process and recorded planned activities that parents were free to adapt during the week. Parents used this form to record time spent in planned and routines-based parent—child interaction during the week and to record incidental comments for review at the next intervention session.

Control Group Procedures

During the intervention period, control group participants participated in services of their choice in their communities, which were reported in monthly interviews. Following the post assessment activities described below and prior to follow-up assessment, families in the control group received the entire set of manualized intervention materials. The materials were described and presented for their independent use in one to three visits (depending on their preference). Families in both intervention and control conditions were free to participate in other community services.

Assessment Procedures and Measures

All families in both groups participated in identical assessment events at four time points: (1) initial eligibility screening, (2) pre-intervention, (3) post-intervention, and (4) follow up. All assessment activities were conducted in family homes by research assistants who were trained on assessment procedures prior to enrolling families in the study. Assessment instructions were identical for both treatment and control conditions at all time points. Instructions were given by assessors who were naïve to treatment assignment.



Eligibility Assessment

Eligibility determination was based on parent interview and three measures for establishing criteria for inclusion. These measures were the Modified Checklist for Autism in Toddlers (Robins et al. 2012), the Toddler Module of the Second Edition of the Autism Diagnostic Observation Schedule (ADOS-T; Lord et al. 2012) and the Precursors of Joint Attention Measure (Schertz 2013).

During the first two home visits, a designation of high risk for ASD was established via administration of the M-CHAT, a validated autism screening tool for toddlers (Wiggins et al. 2010). For children who showed high risk on the M-CHAT, trained assessors administered the ADOS-T (Lord et al. 2012). ADOS-T scores for all participants met the strict cut-off for an autism diagnosis. Finally, the Precursors of Joint Attention Measure (PJAM; Schertz 2013) parent—child interaction observational protocol was administered in which parents were instructed to interact with their children as is typical for them. Ten-minute free-play interactions were video recorded. Videos were then coded by trained observers using the PJAM coding system, as described below, to establish inclusion criteria of three or fewer occurrences of initiating joint attention.

The PJAM is a continuous, partial- interval observational coding system (Yoder and Symons 2010) that assesses the occurrence of four targeted outcomes (focusing on faces, FF; turn-taking, TT; responding to joint attention, RJA; initiating joint attention, IJA). The operational definitions were as follows: for FF the child looked once or more at any part of the parent's face during the interval; for TT the child performed one of at least two related actions in concert with a parent action within no more than two consecutive intervals; for RJA, after the parent attempted to draw the child's attention to an object the child alternated looks between the parent's face and the object for the apparent purpose of sharing social interest; for IJA the child alternated looks between the parent's face and an object for the apparent purpose of drawing the parent's attention to the object (i.e., "showing") while indicating social interest. All observational coding took place at a central observational coding laboratory at the Indiana site. Coders, who were blind to group assignment, recorded whether any of the noted behaviors occurred in each 10-s interval. Before data coding, observers were trained to an 85% agreement criterion on each code. The inter-observer agreement formula was agreement on a behavior occurring in an interval divided by agreement plus disagreement and multiplied by 100. During the study, two coders independently coded 25% of the videos. Cohen's Kappa was then calculated for observational sample. The mean kappas (and ranges) were 0.82 (0.27-1.0) for FF, 0.96 (0-1.0) for TT; 0.71 (0–1.0) for RJA; 0.74 (0–1.0) for IJA.

Pre-Intervention Assessment

All pre-intervention assessment activities were conducted during three visits, each of which included the 10-min video recording of parent—child interaction for later coding with the PJAM (see the description of observation and coding procedures in the previous section). The first parent—child interaction video sample was viewed to complete the Maternal Behavior Rating Scale—Revised (MBRS-R; Mahoney et al. 1998). The MBRS is a 12 item, 5-point Likert-type scale designed to assess four maternal interactive styles. Inter-rater and test—retest reliability as well as alpha coefficients are adequate (Mahoney et al. 1998). An alpha of 0.86 was obtained in the current study.

During the pretest visits the assessor administered the Communication and Symbolic Behavior Scales (CSBS; Wetherby and Prizant 2002) and the Mullen Scales of Early Learning (MSEL; Mullen 1995). Parents completed the Self-Report forms of the Vineland Adaptive Behavior Scales (VABS; Sparrow et al. 2005) and the Repetitive Behavior Scale—Revised (RBS-R; Bodfish et al. 2000).

Assessment of Intervention Implementation and Quantity

Two measures of implementation fidelity were collected in this study. First, data on each IC's fidelity to the JAML's protocol for implementing the intervention with parents was collected weekly. All implementation sessions were recorded and 25% were randomly selected, reviewed, and rated by an independent assessor. Items on this instrument focused on completion of required activities. Second, immediately following each intervention session, ICs completed the 12-item Parent Implementation Fidelity Checklist. Items on this checklist, which were assessed from family logs of daily activities and videos of parent-child interaction recorded during intervention sessions, focused on time spent in daily interaction, integration into daily routines, and use of mediated learning principles. For both the IC and parent fidelity checklists, each item was rated on a 3-point scale, ranging from "not observed," "observed partially," to "observed fully." High levels of implementation fidelity were obtained for ICs (M = 2.96; SD = 0.03; R = 2.85-3.0) and for parents (M=2.74; SD=0.17; R=2.06-2.95).

To assess inter-rater agreement for both IC and parent fidelity checklists, 25 percent of IC and parent fidelity checklists were randomly selected for review by the project manager and assigned to a reliability coder who were not privy to any information about the intervention, participant condition, or assessment time point. Primary coders were naïve to sessions selected for reliability assignment. Based on review of session audio-recordings, the reliability rater independently completed both an IC and parent fidelity checklist. Agreement was computed as the number of checklist items



on which raters agreed divided by the number of items \times 100. High interobserver agreement was found for IC implementation fidelity ratings (M=99%, R=50-100%) and for parent implementation fidelity ratings 93% (range=53-100%).

To assess the quantity of intervention, a structured Monthly Services Record interview was conducted by phone with parents to record the number of hours for which children received services outside of the JAML intervention.

Post-Intervention Assessment

Within 2 weeks following intervention completion, all study families participated in separate post-intervention assessment activities. To assess social validity of the intervention, an 18-item parent report questionnaire was completed to provide information about perceived acceptability of the intervention from the parents' perspectives. To assess intervention outcomes, the PJAM parent—child interaction assessment was re-administered and videos were viewed and coded by trained assessors who were naïve to time point and treatment conditions.

Follow-Up Assessment

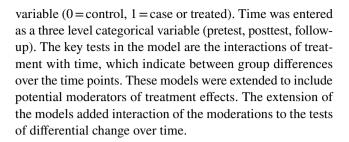
To assess whether post-intervention outcomes were maintained at 6-month follow-up, the PJAM parent-child interaction assessment was re-administered and parent-child interaction videos were coded by trained assessors naïve to condition assignment and time-point.

Results

Data Analysis

The primary research questions regarding treatment effects were addressed via an intent-to-treat analysis. This model focused on change over time and whether change was moderated by treatment; state was the only covariate.

The models were run as hierarchical linear models (HLM) using SAS "proc glimmix" in SAS/STAT v9.3 (SAS Institute 2014). The repeated (pretest, posttest, follow-up) nature of the data induced non-independence within observation. HLM manages this non-independence through the estimation of random effects (e.g., Burchinal et al. 2006), in this case random intercepts. These individual intercepts provide estimates of the variability in the average score within individuals over time. Our primary outcomes, the PJAM variables, had non-normal distributions. These variables were zero inflated, necessitating Poisson regression. The glimmix models were fit with maximum-likelihood estimate and Gauss—Hermite quadrature for the approximation of the likelihood. Treatment was included in the model as a dummy



PJAM

The PJAM data for each outcome variable are graphed in Fig. 2. The data are mean frequency of intervals in which the behavior occurred during a 10-min observation, averaged over the three observations at pretest and posttest and followup. For all measures, differences between the JAML and control group at pretest were minimal and nonsignificant. For FF, there was a significant treatment by time interac $tion^2$ (F = 10.74; p < . 001). Posthoc between group analyses revealed significant differences at posttest (F=5.51, p<.001, d = 1.20) and followup (F = 2.72, p = .007, d = 0.77). TT also had a significant time by treatment interaction (F = 13.57, p < .001). Posthoc between group analyses for TT revealed significant differences at posttest (F = 4.42, p < .001, d = 0.85) and follow-up (F = 4.33, p < .001, d = 0.78). Similarly, for RJA there was a significant time by treatment interaction (F = 6.66; p = .002). Posthoc between group analyses revealed significant differences at posttest (F=5.91, p<.001, d=2.80) and follow-up (F=4.85, p<.001, d=2.67). Last, for IJA, there was a non-significant time x treatment interaction (F = 2.25, p = .1080). Posthoc analyses suggested, however, a significant difference at posttest (F = 3.01, p = .003, d=0.90) but not a significant difference at follow-up despite a respectable effect size (F = 1.75, p = .082, d = 0.69).

Descriptive statistics for all measures are presented in Table 2.

Moderation

To examine whether severity of autism may have affected response to the treatment, we added the ADOS Social Affect scores as a moderator of treatment via a three-way interaction of ADOS Social Affect×treatment×time). This interaction was significant, F(1, 271)=5.77, p=.0169, only on RJA. Post hoc analyses revealed that ADOS scores did not impact change in the control group nor in the treated group from pretest to posttest. However, severity of autism, as indicated by the ADOS, did impact change in the treated group from post-test to follow-up such that higher levels of autism severity was associated with significantly greater increases



² For all tests that include time the degrees of freedom were (2, 221).

Fig. 2 Treatment effects over time on PJAM outcomes

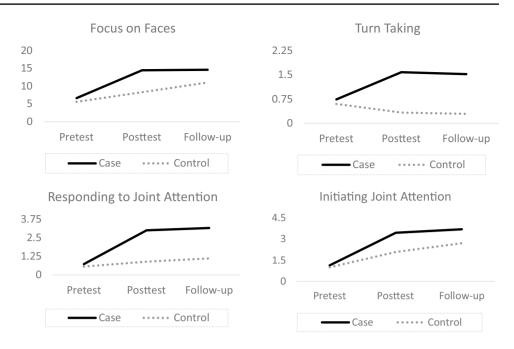


Fig. 3 Moderation of treatment on RJA by ADOS

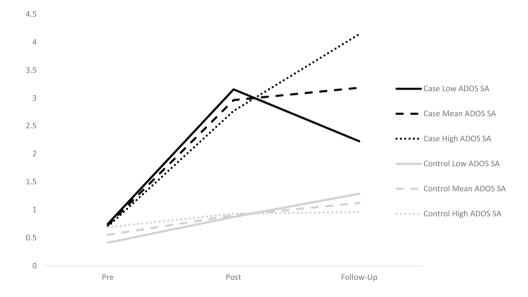


 Table 2
 Descriptive statistics

Variable	Control							Case										
	Pre			Post			FU			Pre			Post			FU		
	N	Mean	SD	N	Mean	SD	N	Mean	SD	N	Mean	SD	N	Mean	SD	N	Mean	SD
Video Coding-# FF	71	5.56	5.15	67	8.28	6.28	59	10.99	7.42	73	6.63	4.82	64	14.45	7.99	55	14.59	8.67
Video Coding-# TT	71	0.60	1.53	67	0.33	1.07	59	0.29	0.67	73	0.74	1.31	64	1.58	3.29	55	1.52	4.33
Video Coding-# RJA	71	0.56	0.75	67	0.89	1.16	59	1.11	1.32	73	0.73	1.14	64	3.00	2.72	55	3.15	5.04
Video Coding-# IJA	71	1.00	1.45	67	2.09	2.50	59	2.71	3.52	73	1.14	1.40	64	3.45	3.42	55	3.70	4.21
Instrument		N Mea				ın			SD			Min					Max	
Parent fidelity: total mean 67 2.74				ļ			0.17			2.06					2.95			
Intervention coordinator: total 67 mean			2.96				0.03			2.85					3.00			



Table 3 Social validity (acceptability of JAML to parents): mean percentage by item categories

Item categories	SA (%)	A (%)	N (%)	D	SD
Importance of the intervention (promoting nonverbal communication embedded in daily interactions)	93	6	1	0	0
The parent role (making own decisions for activities, daily notes, time required)	63	27	8	1%	0
Support received (activity suggestions, right amount of support, guided video reflection)	82	15	3	0	0
Child's progress (FF, TT, RJA, & IJA)	76	19	4	0	0
Parent's improved hopefulness about child's future and confidence with own ability	77	20	3	0	0

SA strongly agree, A agree, N neutral, D disagree, SD strongly disagree

in RJA from post-test to follow-up compared to those with lesser autism severity during this period. that is, the direction of change varies inversely with ADOS scores. Children with high ADOS scores change in a positive direction; as ADOS scores decrease, the direction of change shifts from positive to negative (Fig. 3).

Missing Data Analysis

Multiple imputation via the EM algorithm (see, for example, Enders 2010; Schafer and Graham 2002) was used to replace missing data. Twenty-five datasets with imputed data were estimated. We then replicated the analysis models on these data and combined the results using SAS "proc mi analyze" (SAS 9.4, SAS Institute 2014). Results for ITT models were confirmed.

Other Services Received

Excluding JAML sessions, reported weekly hours for intervention and control group participants, respectively, were 7.41 (SD=4.67) and 12.82 (SD=14.06) for Indiana, 17.88 (SD=9.06) and 21.35 (SD=11.51) for Kansas, and 2.98 (SD=1.25) and 6.25 (SD=6.49) for North Carolina. We tested amount of services as a moderator of child outcomes. Across all outcomes, the moderation effect (Treat \times Time \times Services) was non-significant, all p>.05.

Social Validity

Finally, we tested social validity via our measures of parent and interventionist fidelity. Social validity findings are presented in Table 3 with item categories summarized by topic. Responses largely indicated acceptability of all aspects of the JAML intervention.



This randomized controlled study of the JAML intervention found post-intervention effects for FF, TT, RJA, and IJA, with all but IJA sustaining treatment differences over a 6-month period after intervention. The effect sizes for these effects are quite large with all $d \ge 0.69$. The achievement of joint attention outcomes in parent-child interaction is important for its well-replicated associations with later language, cognitive, and social outcomes (Bottema-Beutel 2016; Charman 2003; Freeman et al. 2015; Gillespie-Lynch et al. 2012; Mundy et al. 2007). Although the intervention effects were generally not moderated by autism severity, there was an exception with RJA. There was some indication that more positive changes were occurring for the toddlers most affected by autism, as compared with other children in the study, a finding roughly paralleling that of Siller et al. (2013) in which children with lower language levels benefitted most from intervention that emphasized parent responsiveness. Measurement of fidelity indicated that the JAML intervention was consistently implemented as planned and social validity data indicated high levels of acceptability by parents.

Two key ingredients of the intervention may have led to these results: its targeted content and certain attributes of the intervention process. The targeted content concentrated the intervention focus on the primary area of concern in autism, social communication. Joint attention was supported by two simpler social communication outcomes, focusing on faces and turn taking, which precede joint attention in typical development and which each contain component aspects of joint attention. Given the now emerging evidence from research findings of Klin and colleagues (Jones and Klin 2013; Klin and Jones 2017; Moriuchi et al. 2017) as well as other research groups



(Chawarska et al. 2013; Elsabbagh et al. 2015) that early social gaze may be a very early diagnostic indicator of autism, JAML's impact on this variable may have future implications for intervention with children even younger than the toddler age group.

In addition, the intervention highlights a subtle but important distinction in how early social communication intervention is conceptualized. Specifically, joint attention incorporates visual synchrony with gaze shifts between the object and partner and reciprocal engagement by taking the partner's interest into account; the latter is also demonstrated in a simpler form in turn taking. Importantly, the focus throughout the intervention phases was on social rather than instrumental communicative functions. For example, in promoting joint attention, in which partners visually engage with one another in reference to objects, instrumental functions of following directions or requesting were explicitly excluded as intervention targets in favor of engaging around mutual interests. While these instrumental outcomes may be important for some children and parents, approaches to promoting instrumental functions may be different, as are their associations with later communication outcomes (i.e., the linkage is stronger for joint attention) (Adamson et al. 2001; Curcio 1978; Mundy 1995; Stone et al. 1997). Mundy (2016), in his comprehensive treatment of the joint attention literature, also notes the stronger connection of social than of requesting initiations with social relatedness. Naturalistic behavioral interventions may be well suited for promoting instrumental functions and we propose that a mediated learning approach may be best suited for promoting social functions, including joint attention.

Our definition of joint attention favors interaction with the partner over solitary orientation to objects around which partners are mutually focused or to which they are just in close proximity. This quality distinguishes joint attention from joint engagement, since, in the latter the child and parent attend to a common focus without actively attending to the partner (Adamson et al. 2009). We conceptualized the promotion of joint attention as an internalized social process in which the child shares attention with gaze shifts to the partner to register their partner's common interest. In this sense it is volitional and socially interactional. It also extends beyond addressing only one partner's interests by providing a reason to communicate beyond requesting or following the partner's direction. This internalization of a social reason to communicate may account for joint attention's association with later social and language outcomes.

A second key ingredient concerns the intervention process. JAML is predicated on family-centered and family capacity-building practices in which parents are supported as primary change agents, both recommended early intervention practices (Division for Early Childhood 2014). Integrating intervention throughout the totality of parent-toddler

interactions in natural settings differs in important ways from dosage-dependent traditional practices in which professionals intervene directly with children, and is supported by meta-analytic findings that dosage in early intervention did not predict better outcomes and that parent-implemented components contributed to communication gains (Hampton and Kaiser 2016).

Thus, effectiveness of parent-implemented intervention may depend less on an intensive professional time commitment than on the quality of professional support to promote active parent learning and participation. For JAML this was accomplished by systematic guidance related to its social communication content and mediating learning principles. In addition, role clarity assured that ICs provided conceptual guidance while parents, with assistance, translated learned concepts related to current outcomes and principles into natural parenting routines. An essential feature of mediated learning is that parents build from their own expertise and knowledge about their toddlers' interests and family priorities (Schertz and Horn 2017). Importantly, this approach to supporting parents' learning takes on a different form and function than do approaches labeled "parent training" in which parents are taught to implement predetermined intervention strategies and protocols. Thus, a critical ingredient may be implementation of systematic practices to support parents' knowledge, competence, confidence, and followthrough for guiding their toddlers' learning.

In sum, features of the JAML intervention that may account for its positive effects on social outcomes include its closely targeted social (vs. instrumental) partner-focused content and its explicit focus on family capacity-building in which parent learning is mediated rather than prescriptively trained. This latter feature may function as a proxy for "intensity" by offsetting earlier assumptions that high-intensity professionally delivered intervention is required for child outcomes to be realized (e.g., Lord and McGee 2001; Rogers et al. 2012). Also, as seen in differences in outcomes across the RCTs reviewed in the introduction, the use of socially focused measures may be essential to capture change in the this most important area of concern in autism.

Maintenance and moderation are two features of the study outcomes that deserve mention. As a group, children in the JAML intervention maintained, at six months post-treatment, gains made in the intervention, with only hardly noticeable declines for initiating joint attention. The treatment differences between groups narrowed for focusing on faces and initiating joint attention. Mainly this was because the control group children increased in both areas. Interestingly, the differences widened for turn taking and responding to joint attention, perhaps suggesting that the JAML intervention created a caregiving environment that is inherently more social, and which generates greater child responsiveness. Synchrony in communication and



responding to social overtures, as happens in turn taking and responding to joint attention are key elements of successful social communication. Findings that young verbal children with autism are more proficient in expressive than in receptive communication (Kim et al. 2014), unlike those with other disabilities, support the importance of targeting outcomes for preverbal social responding as well as initiating. Longer-term follow up may well link these key competencies to greater future communication facility. In addition, it should be pointed out that control group parents received the JAML materials after the posttest with description of and instructions about this intervention. It is not possible to rule out that motivated parents receiving this information could have influenced increases in the control group's performance in 6-month follow-up assessments.

The preferential response of toddlers with more severe expressions of autism on the RJA measure during the 6-month post-intervention period in our moderation test may have been an outcome of JAML's sequentially ordered approach to social communication intervention. The intervention protocol began by promoting a basic form of dyadic interaction, visual synchrony, and progressing to the more complex triadic forms of joint attention. This approach may have favored toddlers with a weaker social foundation who were more reliant than their less severely involved counterparts on the use of mediated learning principles to promote active engagement in social learning.

This study builds on our prior work (Schertz and Odom 2007; Odom et al. 2011; Schertz et al. 2013) and adds to other previously published RCTs of intervention that exclusively targeted social communication for toddlers with autism (Kasari et al. 2015; Landa et al. 2011; Wetherby et al. 2014). Given its low resource demands and high social validity, an intervention such as JAML appears feasible and appropriate for community implementation. The features that distinguish JAML, and thus advance the literature, are implementation in natural environments, a central focus on family capacity building for delivering the intervention, implementation across multiple sites, and monitoring of maintenance effects. As such, JAML adds to the emerging evidence-base for early intervention practitioners who work with very young children with ASD and their families.

Several limitations should guide future research. First, although we believe our sample is largely representative of the populations in our sites, the majority was white, of moderate income, and had some level of college education. Future research sites should target populations that are more culturally and racially diverse. Second, the minor differences in attrition between groups may have been influenced by the unequal time commitments required of experimental and control group participants, a circumstance that might

be controlled in future research by comparative intervention studies in which the intervention commitment is comparable.

Third, because parent-child interaction is the most proximal and authentic context for toddler social development, and best practice recommendations for very young children call for authentic assessment that includes observation of parent-child interaction for very young children (Bagnato 2005; Division for Early Childhood 2014), we employed direct observational assessment of parent-child interaction to address the central research question of whether toddler social behavior toward their parents changed as a function of the parent mediated JAML intervention. In JAML's transactional learning approach in which both partners' actions are viewed as influencing the other in social communication exchanges, parent learning is broadly mediated rather than prescriptively trained, and post-intervention assessment was meant to capture the interactional dynamic rather than child behaviors in isolation, recognizing that parents' actions are likely to influence those of their toddlers. However, a separate question for future research is how child competencies, learned within a closely supported parent-child relationship, may generalize to interactions with other familiar adults, a question that may produce different results. Fourth, related to parents' implementation, we used a rating scale to assess implementation fidelity of intervention participants. In the future, application of an observational coding system to assess parents' implementation of mediated learning principles would provide a more direct assessment of fidelity and could be applied to families in the control group as well.

Several additional needs for future research emerge from our findings. Although a more general measure of early parent self efficacy is available (Guimond et al. 2008), a reliable and valid of measure of parent self-efficacy specifically tested on parents of toddlers with autism is not. The JAML intervention uses multiple strategies to bolster parent self-efficacy and an autism-specific measure could assess possible effects of these strategies on parents' views of their own competence in specifically promoting child social communication learning in future studies. The field could also benefit from future study of JAML's sequencing of targeted foundational social competencies to determine whether the three-phase intervention sequence is optimal for promoting joint attention and to assess its long-term effects on later social communication outcomes. Finally, although our findings showed significant social communication differences between groups, assessment of the longer-term stability of these findings could help to determine their broader clinical significance.

In sum, this research showed evidence of improvement in preverbal social communication for toddlers with autism from parent-mediated intervention that builds on a series of foundational social communication competencies. The use of a mediated learning approach may have also contributed



to child gains as well as to maintenance of those gains through the 6-month follow-up period. Future studies are needed to extend this research by developing and examining a program to support early interventionists in the field to determine the transferability of intervention methods to community-based settings and to assess the level of support needed for practitioners to implement the program with fidelity.

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Author Contributions HS, SO, and KB participated in conceiving, designing, and implementing the study. HS coordinated development of the manuscript. KB substantially contributed to the methods description, SO oversaw the data reporting, and JS conducted the statistical analyses. HS, SO, and KB, who served as PI, Co-PI, and Co-PI, respectively, implemented site-based activities in coordination with the central site. HS provided direction for intervention activities and for the project as a whole, SO coordinated research design and analysis activities, and KB coordinated assessment processes. All authors read and approved the final manuscript.

Compliance with Ethical Standards

Conflict of interest The authors declare that they have no conflict of interest.

Ethical Approval All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Informed Consent Informed consent was obtained from all individual participants included in the study.

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