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# Comparing Spoken Language Treatments for Minimally Verbal Preschoolers with Autism Spectrum Disorders

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Abstract	Preschoolers with severe autism and minimal speech were assigned either a discrete trial or a naturalistic language treatment, and parents of all participants also received in parent responsiveness training. After 12 weeks, both groups showed comparable improvement in number of spoken words produced, on average. Approximately half the children in each group achieved benchmarks for the first stage of functional spoken language development, as defined by Tager-Flusberg et al. (J Speech Lang Hear Res, 52: 643–652, 2009). Analyses of moderators of treatment suggest that joint attention moderates response to both treatments, and children with better receptive language pre-treatment do better with the naturalistic method, while those with lower receptive language show better response to the discrete trial treatment. The implications of these findings are discussed.
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#### ORIGINAL PAPER

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6 7 Preschoolers with Autism Spectrum Disorders

**Comparing Spoken Language Treatments for Minimally Verbal** 

4 Rhea Paul · Daniel Campbell · Kimberly Gilbert ·

5 Ioanna Tsiouri

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8 Abstract Preschoolers with severe autism and minimal 9 speech were assigned either a discrete trial or a naturalistic 10 language treatment, and parents of all participants also 11 received in parent responsiveness training. After 12 weeks, 12 both groups showed comparable improvement in number 13 of spoken words produced, on average. Approximately half 14 the children in each group achieved benchmarks for the 15 first stage of functional spoken language development, as 16 defined by Tager-Flusberg et al. (J Speech Lang Hear Res, 17 52: 643-652, 2009). Analyses of moderators of treatment 18 suggest that joint attention moderates response to both 19 treatments, and children with better receptive language pre-20 treatment do better with the naturalistic method, while 21 those with lower receptive language show better response 22 to the discrete trial treatment. The implications of these 23 findings are discussed.

24

- Keywords Autism · Language · Treatment ·
   Intervention · Communication · Speech
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Children with autism spectrum disorder (ASD) are almost 27 universally delayed in the acquisition of spoken language. 28 Although rates of functional use of speech have increased 29 in this population during the last decade (Rogers 2006), the 30 acquisition of spoken language remains an especially 31 important attainment for children with ASD. Children who 32 do not acquire speech as a primary means of communica-33 tion by school age tend to have restricted outcomes in 34 terms of independence and integration (Howlin 2005). 35 Therefore it is important to make every attempt to induce 36 speech in preverbal children with ASD during the pre-37 school period in order to maximize opportunities for social 38 39 interactions with family and peers and participation in mainstream settings in school and later life. The motivation 40 behind this study is to investigate the most effective ways 41 to induce speech in minimally verb dren with ASD. 42

43 A variety of intervention approaches-from the most structured discrete trial instruction methods to more open-44 ended, child-centered methods-demonstrate some effi-45 cacy both for increasing communication and eliciting first 46 words from nonspeaking young children with ASD (See 47 National Research Council 2001; Paul 2008; Prelock et al. 48 49 2011; Rogers 2006 for review). One method that has a strong evidence base for eliciting first words from these 50 children is Discrete Trial Training (DTT; Lovaas 1987) 51 which makes use of the Skinnerian principles of operant 52 learning (Skinner 1957). (Reichow and Wolery 53 <u>109</u>) reviewed research using such methods for childr 54 55 ASD and found that, although few studies consistently met standards for establishing evidence-based practice, 5/6 56 57 studies that met minimum criteria showed significant improvement for children receiving DTT for expressive 58 language, based on effect size. Moreover, the four studies 59 comparing DTT to other methods for improving spoken 60 language all demonstrated greater gains in both expression 61

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62 and comprehension for the DTT intervention than the alternative treatment. Thus DTT approaches would appear 63 64 to have some degree of efficacy for facilitating early lan-65 guage development. Nonetheless, DTT approaches have 66 long been criticized (e.g., Delprato 2001; Smith 2001), 67 particularly in the area of communication skills (Fey 1986; Owens 2009; Prizant and Wetherby 2005), due primarily to 68 69 the fact that gains made often fail to generalize outside the 70 training setting, to be used for spontaneous, functional 71 communication, or to be maintained by the ordinary con-72 tingencies of daily life, when tangible reinforcement is 73 removed (Fey 1986; Stokes and Baer 1977).

Another approach to inducing first words in nonspeaking children with disabilities was developed in response to these shortcomings. Milieu Communication Training (MCT) has been supported in a range of studies of children with disabilities (e.g., (Hester et al. 1995; Yoder and Warren 2002) including children with ASD (e.g., Hancock and Kaiser 2002; Ross and Greer 2003; Christensen-Sandfort and Whinnery 2011; Yoder and Stone 2006). MCT aims to address some of the identified shortcomings of DTT by means of several strategies, including

- teaching within natural environments (Kaiser et al. 1992), since research has demonstrated increases in generalization (Hancock and Kaiser 2002), maintenance (Spradlin and Siegel 1982), and spontaneous use of language (Yoder and Warren 2002) in natural environments over isolated clinical settings;
- mand-modeling (Rogers-Warren and Warren 1980), or
  providing a model of desired communicative act and
  correcting child responses;
- 3. time delay, involving the adult's providing a stimulus
  and then waiting approximately 5–15 s, for a childinitiated response (Kaiser 2010);
- incidental teaching strategies (Hancock and Kaiser 96 4. 97 2002; Hart and Risley 1975) such as free play in which 98 the child controls the teaching episodes by signaling 99 interest in the environment, which the adult has 100 organized so that access to desired objects is contin-101 gent upon solicitation of adult assistance, which is followed by both praise and access to desired 102 outcomes. For example, an adult may "accidentally" 103 forget to give a child milk during snack, then would 104 105 give a prompt for the child to request it ("What do you 106 need?"), praising the child for correct responses, and 107 giving the child the milk (contingent access).

108 In their research on MCT with young children with 109 ASD, Yoder and colleagues (e.g., Yoder and Stone 2006) 110 have incorporated parent responsiveness training into the 111 MCT intervention package, and have argued for the 112 importance, in this context, of not only increasing child 113 communicative initiations, but of providing responsive feedback to these initiations in order to effect lasting, 114 functional change in communicative behavior. 115

One aim of the present study was to examine the useful-116 ness of a DTT treatment that we believed would be espe-117 cially efficacious for minimally verb children with ASD. A 118 primary reason that some children who were tried on DTT to 119 induce speech did not succeed was that they were unable to 120 produce any vocal imitation. Vocal imitation is the necessary 121 first step in a DTT approach, since only with some vocal 122 behavior to shape can the child's behavior be modified 123 toward speech. We believed that a DTT approach developed 124 by (Tsiouri 2002) had potential to address this problem. This 125 procedure, Rapid Motor Imitation Antecedent (RMIA) 126 training, required the child to produce a series of simple 127 motor imitations before being presented with opportunities 128 to imitate verbal "mands" (requests) or "tacts" (labels). 129 This instructional strategy utilized the child's motor imita-130 131 tion repertoire to facilitate the emergence of first instances of vocal imitation ("echoics"), which could then be shaped into 132 verbal imitation and eventually to independent word pro-133 duction. The unique contribution of RMIA is hypothesized 134 to reside in its capacity to induce vocal imitations through 135 behavioral momentum (Mace et al. 1990; Nevin et al. 1983). 136 Several researchers (e.g., Mace and Belfiore 1990; Mace 137 et al. 1990) have demonstrated that behavioral momentum 138 can be harnessed to elicit behaviors previously resistant to 139 140 treatment, and have shown that when children's compliance with easy instructions was highly reinforced, compliance 141 persisted when more difficult instructions, with which the 142 children were normally non-compliant, were chained after a 143 144 series of easy behaviors. Both Tsiouri and Greer (2003) and we (Paul 2009; Tsiouri et al. 2012) have been able to show, in 145 published case series, that this momentum can, in fact, lead 146 to production of first words in some minimally verb pre-147 148 schoolers with ASD.

149 Because so few DTT approaches have been subjected to experimental procedures such as controlled trials compar-150 ing them with alternative approaches, we also aimed to 151 provide such a contrast in this study. Because of the known 152 efficacy of MCT for increasing communication (and 153 sometimes, speech) in minimally verb young children, we 154 believed comparing RMIA to MCT would constitute a fair 155 test of the relative efficacy of the two approaches and, at 156 least, MCT would have positive benefits in increasing 157 communication in participants who received this treatment. 158 159 But, since most recent research on MCT had incorporated a parent responsiveness training component, as well as to 160 address some of the limitations of DTT in terms of gen-161 eralization, a parent responsiveness training component 162 was included in BOTH interventions. Thus, the study 163 contrasts two intervention packages, one consisting of 164 RMIA plus parent responsiveness training, and the other of 165 MCT with parent responsiveness training. 166

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167 We were also interested in the identification of pre-168 treatment characteristics of participants that could be 169 associated with positive responses to each of these treat-170 ment packages. Research suggests that the relative efficacy 171 of one treatment over another is likely to vary by pre-172 treatment child characteristics (National Research Council 173 2001; Yoder and Stone 2006). Thus, the identification of 174 pre-treatment variables associated with enhanced response 175 to one treatment or the other would aid in identification of 176 treatments most likely to work best for particular children.

Thus, the package of intervention developed for thisstudy included assignment of participants to one of twoclinician-delivered interventions:

- 1. a DTT program enhanced with a behavioral momentum component (RMIA), to help children acquire the vocal/verbal imitation skills necessary for speech acquisition, or
- 2. an MCT program of naturalistic, play-based intervention.

186 Regardless of which intervention the child received,
187 parents of all participants were provided with Parent
188 Responsivity Training, following Yoder and Warren (2002).
189 The aims of the study were:

to determine whether either RMIA or MCT were more effective overall, in conjunction with parent responsiveness training, in eliciting spontaneous functional speech from minimally verice eschoolers with ASD;

194 2. to examine pre-treatment subject characteristics as
195 moderators of response to treatment in order to identify
196 subject profiles that could predict better response to
197 one treatment package or the other.

#### 198 Methods

199 Participants

Participants were recruited through written and electronic 200 201 media advertisements. Flyers and brochures were distrib-202 uted to local special education departments and early 203 intervention providers. Additional participants were 204 recruited through the university's website. A speech-lan-205 guage pathologist screened all interested individuals. All 206 participants' families completed informed consent proce-207 dures approved by the Institutional Review Board for the 208 Protection of Human Subjects. Inclusion criteria were:

DSM-IV-TR (2000) diagnosis of Autistic Disorder or
 PDD-NOS as conferred by an experienced clinical team
 and confirmed by scores within the autism spectrum
 range on the Autism Diagnostic Observation Scale

*–Module 1* (Lord et al. 2000) administered by highly 213 trained clinicians; 214

- spontaneous expressive vocabulary by parent report of 215 fewer than 15 words as measured by the Communication 216 and Symbolic Behavior Scales-Caregiver Questionnaire 217 (Wetherby and Prizant 2003)-73 % of the participants 218 had fewer than 5 words reported-and fewer than 8 219 intelligible words produced during a 20-min clinician-220 221 child play observation Communication and Symbolic 222 Behavior Scales-Behavioral Observation (Wetherby and Prizant 2003)—91 % produced fewer than 5 words; 223
- expressive language age-equivalent of less than 18 months as measured by the Vineland Adaptive 225 Behavior Scales—II (VABS-II; Sparrow et al. 2005) 226 Expressive Language subdomain; 227
- non-verbal mental age of at least 12 months as measured by the *Mullen Scales of Early Learning* (Mullen 1995), Visual Reception subdomain; 230
- generalized motor imitation, which for the purposes of this study, was defined as the ability to accurately imitate a repertoire of motor actions using the (Meltzo) 233 1988) motor imitation procedure.

Exclusionary criteria consisted of any uncorrected235vision or hearing disability. Table 1 provides a description236of participants at their entrance into the intervention program. One-way analysis of variance revealed no significant237differences between the two treatment groups on any of239these pretreatment variables.240

- Assessment Procedures
- **Pre-treatment Assessment**

Each participant completed two, 2-h evaluations to ensure 243 they met entrance criteria for the study and to collect 244 information on their pre-treatment level of functioning. 245 The following standardized measures were included: 246

- I. *Mullen Scales of Early Learning* (Mullen 1995) was used to establish nonverbal cognitive level; 248
- II.The Autism Diagnostic Observation Schedule—Module 1 (ADOS; Gotham et al. 2008) was used to confirm<br/>diagnosis of ASD;249250251
- III.Communication and Symbolic Behavior Scales—Devel-<br/>opmental Profile (CSBS; Wetherby and Prizant 2003)252<br/>253was used to assess frequency and types of spontaneous<br/>words used, frequency of joint attentional communica-<br/>tive acts, and frequency of symbolic play behaviors.254

Each participant completed a motor imitation assessment (Meltzoff 1988), which included imitation of actions with objects (e.g., shaking a rattle), gross motor imitation (e.g., stomping feet, tapping knees), fine motor imitation 260

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 Table 1
 Participant

 characteristics
 Image: Characteristic state

Rapid motor i	imitation antecend	dent (RMIA) Tx	Milieu comm	nunication traini	ng (MCT) Tx
Subject	Age	Gender	Subject	Age	Gender
007TS	5.90	Female	005TS	3.64	Male
015TS	4.08	Female	008TS	2.71	Male
020TS	5.47	Male	016TS	2.80	Female
033TS	3.66	Male	049TS	3.70	Male
036TS	6.15	Male	070TS	2.63	Male
043TS	4.76	Female	074TS	4.14	Male
046TS	3.44	Male	077TS	3.49	Male
060TS	3.53	Female	061TS	3.15	Male
076TS	4.76	Male	075TS	3.22	Male
081TS	2.40	Male	078TS	4.88	Male
			079TS	3.32	Male
			080TS	4.56	Male

261 (e.g., touching nose, touching mouth), and oral motor 262 imitation (e.g., opening mouth, smiling, puckering). Stan-263 dardized measures were administered by a speech-lan-264 guage pathologist and licensed clinical psychologist. In 265 addition to direct observation measures, parents completed 266 questionnaires including the Vineland Adaptive Behavior Scales-II (Sparrow et al. 2005), the MacArthur-Bates 267 268 Communicative Development Inventory (Fenson, et al. 269 2007), the Caregiver Questionnaire of the CSBS, and a 270 description of current and previous intervention. Parents 271 were also videorecorded while engaged in a 10 min play 272 session with their children with a standard set of toys 273 (following Yoder and Warren 2002), and the percentage of 274 parental acts responsive to the child's focus was computed.

#### 275 Follow-up and Maintenance Assessments

276 Within 2 weeks of the completion of the 36 treatment ses-277 sions, each child was re-assessed, using the same procedures 278 as for pre-treatment assessment, with the exception of the 279 Mullen, which was not re-administered at this time. Three to 280 6 months following the end of treatment the entire assess-281 ment battery, including the Mullen, was re-administered. 282 Assessors at Follow-up and Maintenance were blind to the 283 treatment assignment of the participants, and were different 284 from the clinicians delivering the intervention as well as 285 from the examiners at the Pre-treatment Assessment.

#### 286 Pre-treatment procedures

Based on the responses to the motor imitation probes
during Pre-treatment Assessment, participants who had
generalized motor imitation in their repertoire (as defined
by performance of 60 % correct or better on the motor
imitation probes) were randomly assigned to either MCT or
RMIA treatment.

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Participants who were unable to imitate 60 % of actions 293 during the Pre-Treatment Assessment were provided with 294 295 ten, 30-min training sessions on motor imitation in order to develop their generalized motor imitation repertoire. 296 A standard DTT format was used to teach the participants 297 to independently and accurately imitate motor actions, 298 through gradual prompt fading and reinforcement proce-299 300 dures, within a specific inter-response time (1 s). The goal for this training procedure was to teach the child to imitate 301 302 at least 6 different motor actions (three gross and three fine) in sequence within 6-8 s. Following this training, children 303 who achieved this criterion in motor imitation were ran-304 domized to one of the two treatments: however five chil-305 306 dren who did not achieve the criterion for motor imitation 307 were non-randomly assigned to the MCT group, resulting in a design for this study that is only quasi-experimental, 308 309 rather than a standard randomized controlled trial. Figure 1 310 summarizes the in-take procedure for this study.

#### Treatment Procedures

Participants assigned to one of the two treatments received 312 36 45-min sessions over the course of 12 weeks with cer-313 tified speech-language pathologist (SLP) specifically 314 trained in RMIA by the third author and in MCT by the 315 fourth (all clinicians were trained in both approaches, with 316 periodic retraining throughout the course of the study). 317 Fidelity of treatment was monitored by having the each 318 treatment's trainer (third and fourth authors) code, via 319 video recording, a randomly selected sample of 10 % of the 320 treatment sessions (clinicians were blind to which sessions 321 322 were being rated for fidelity). This procedure revealed an average of 96 % agreement between clinician and consul-323 tant as to the appropriateness of the clinician's response to 324 child behaviors within our established criteria for fidelity 325 with RMIA treatment; and 92 % for MCT. 326







#### 327 RMIA training procedures

Preferred items used during treatment were selected individually for each participant, using a variation of the
Multiple Stimulus Without Replacement Preference
Assessment procedure (DeLeon and Iwata 1996) conducted

before the onset of the study, as well as periodically 332 throughout the intervention to ensure reinforcers remained 333 powerful. The instructor obtained the participant's attention, then rapidly and randomly presented three large (hand 335 and foot movements) and three small (pointing to parts of 336 the face) motor actions with the antecedent, "Do this," 337

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338 allowing the participant 1 s to respond to each action. The 339 participant imitated actions one by one as they were pre-340 sented. If the participant failed to imitate more than one 341 action in the sequence within the 1 s. Time frame, the 342 sequence was begun again. Immediately after the com-343 pletion of the 6 motor actions, the instructor said the target 344 working displayed the target item (preferred items for required and non-preferred for labels). The child was 345 required to say the target word (or a predetermined 346 347 approximation of the word, which was gradually shaped 348 toward the target word through the course of the inter-349 vention) in order to receive the preferred item (for requests) 350 or to receive a choice of two preferred items different from 351 the target (for labels). Detailed descriptions of the RMIA 352 procedures can be found in Tsiouri and Paul (2012).

#### 353 Milieu Communication Training Procedures

354 During each session, the clinician attempted to establish 355 play routines that were enjoyable and motivating to the 356 child, and engineer the environment to include multiple 357 motivating opportunities for the child to communicate, 358 such as placing desired items in closed containers the child 359 could not open without help, or requiring the indication of 360 a choice between two playthings before access to any play 361 objects was provided. When the child was highly motivated 362 to communicate, clinicians attempted to stimulate initiating joint attention through time delay, and mand-modeling the 363 364 use of recently learned communicative behaviors, focusing 365 on spoken, rather than general communicative responses, and shaping earlier occurring communicative behaviors 366 367 toward speech. More detailed description of the MCT can be found in Paul and Sutherland (2005), Warren and Yoder 368 369 (1998), and Yoder and Stone (2006).

#### 370 Parent Responsivity Training

371 To promote generalization of language learned in both 372 clinician-delivered interventions provided in this study, 373 procedures of Parent Responsiveness Training (Yoder and 374 Warren 2002) were followed. At least one parent of each of 375 the participants was required to attend 4, 2-h parent edu-376 cation classes. Parents completed the classes during the 377 time their child was enrolled in treatment. Instruction was 378 provided in the form of lecture, video, modeling and hands-379 on practice during class. Homework was assigned and then 380 discussed during the next class. Parents were also provided 381 with individual coaching. The purpose of the parent com-382 ponent was to guide parents to increase their use of 383 responsive strategies to help their children engage in pro-384 ductive, interactive play with objects and to facilitate their 385 children's communication and language development. It 386 should be noted that Parent Responsivity Training, 391

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although an integral part of the treatment package provided387in this study, cannot be considered an independent variable,388since parents in both treatment groups received this389training.390

#### Results

- Three sets of results are presented:
- Descriptive statistics for average performance on several variables measuring language and communication before and after intervention for each treatment, with tests of differences before and after treatment within each group, and after treatment between the two treatment groups;
   393 394 395 396
- Proportions of children within each treatment group who met Tager-Flusberg et al.'s criteria (2009) for the attainment of functional spoken language after treatment;
   399 400 401 402
- An assessment of the effect of pre-treatment moderator variables on expressive language outcomes across the two treatments.
   403

   404

#### Changes Pre-Post Treatment

407 Table 2 presents the scores on variables collected immediately post-treatment (12 weeks following the pre-408 treatment assessment) and at the maintenance point 409 (3-6 months following the post-treatment assessment). 410 One-way Analysis of Variance (SPSS 19) revealed no sig-411 nificant differences between the two treatment groups' 412 scores on any of the outcome variables at either the 413 post-treatment or maintenance time point. 414

Paired t tests were then used to look for differences in 415 our outcome variable of interest, spoken language output, 416 between assessment time points within each treatment 417 418 group. For the group that received RMIA, significantly more words were produced during the CSBS play session 419 (t = 2.9 [9], p < .02, Cohen's (1988) d = 1.7 [very large])420 421 and on the number of words said as reported by parents on the CDI (t = 2.3 [8], p < .05, Cohen's d = 1.0 [large]) at 422 the post-treatment assessment relative to pre-treatment. 423 There were no significant differences in these variables 424 from post-treatment to the maintenance time point, but 425 there was a significant difference between pre-treatment 426 and maintenance for both CSBS (t = 2.6 [9], p < .03, 427 Cohen's d = .93 [large]) and CDI (t = 2.4 [7], p < .05, 428 Cohen's d = 1.2 [very large]) word counts. The same 429 pattern of results was seen for the age-equivalent scores on 430 the Vineland Adaptive Behavior Scales Expressive Lan-431 guage scale (pre-tx—post-tx: t = 2.4[8], p < .04, Cohen's 432 d = .82 [large]; post-treatment—maintenance: NSD). 433

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Table .	2 Descripti	ion of p	articipants a	ut pre-treatme	ant								
Tx group	Mean (and SD) Age (yrs.)	% male	Mean (and SD) <i>Mullen</i> VR AE <sup>1</sup>	Mean (and SD) <i>VABS</i> -II EL AE <sup>2</sup>	Mean (and SD) <i>VABS</i> -II RL AE <sup>3</sup>	Mean (and SD) <i>CSBS<sup>4</sup></i> : number of spoken words	Mean (and SD) reported words said on <i>CDI</i> <sup>5</sup>	Mean (and SD) number of joint attentional acts on <i>CSBS</i>	Mean (and SD) number of symbolic play acts on <i>CSBS</i>	Mean (and SD): % correctly imitated motor actions	% needing motor training	Mean (and SD) <i>ADOS</i> - SA <sup>6</sup>	Mean (and SD) PCI <sup>7</sup> %
RMIA MCT	4.3 (1.2) 3.5 (0.8)	63.6 90.9	22.6 (4.6) 22.2 (5.6)	$\begin{array}{c} 10.0 \ (4.8) \\ 9.4 \ (0.6) \end{array}$	14.8 (8.2) 12.6 (8.6)	1.7 (2.3) 1.4 (2.2)	4.6 (5.7) 3.8 (3.6)	1.0(1.3) 0.6(0.9)	3.1 (3.9) 4.8 (4.2)	44.9 (28.2) 31.8 (30.7)	63.6 63.6	14.0 (3.8) 14.6 (3.9)	38.3 (15.7) 37.4 (19.3)
1     Mull.       2     Vinel       2     Vinel       3     Vinel       4     Comu       5     Mac/       6     Autis       7     Paren       Cohen     Cohen	en Scales of and Adapti and Adapti and Adapti nunication Urthur-Bate m Diagnos, t-Child Int (1988)	f Early ive Behu ive Behu ive Behu and Sy is Comm tic Obse eraction	Learning (A avior Scales avior Scales mbolic Beha nunicative D ervation Sca (Yoder and	Aullen 1995) -II (Sparrow -II (Sparrow -II (Sparrow vvior Scale-D bevelopment 1 le-Module 1 le-Module 1	Visual Rece et al. 2005) ] et al. 2005) ] evelopmenta Inventory (Fe (Lord et al. 2 2) percentag	ption Age-equiva Expressive Langu Receptive Langu <i>I Profile</i> (Wetherl anson et al. 2007) 2000) Social-Affe e parents' respon	lent score (mon age Age-equival age Age-equival by and Prizant 2 by the Algorithm sive communica	ths) duent score (months) ent score (months) 2003) Spoken Word 1 score tion acts	) I (Type) Inventory (	during 20 min play	v session		

These data suggest that, on average, children who received434RMIA produced more words and used more language in435everyday situations after treatment than before, and these436gains were maintained for at least 3–6 months.437

Similar analyses were conducted for the children who 438 439 received MCT. In this group, an analogous pattern of change was seen for the number of words produced on the 440 CSBS (pre-tx- post tx: t = 2.5[11], p < .03 (Cohen 1988) 441 d = .73 [medium]; pre-tx—maintenance: t = 2.5 [7], 442 p < .04, Cohen's d = 1.2 [very large]; post-tx-mainte-443 nance: NSD) and CDI (pre-tx- post tx: t = 2.3[9], p < .05, 444 Cohen's d = .89 [large]; pre-tx—maintenance: t = 2.6445 [6], p < .04, Cohen's d = 1.3 [very large]; post-tx-main-446 tenance: NSD). However, for the adaptive use of language 447 on the Vineland Adaptive Behavior Scales-Expressive 448 Language Scale none of the differences over time reached 449 significance for the group receiving MCT. CSBS and CDI 450 data are displayed in Figs. 2 and 3. 451

Proportion of Children Achieving Verbal Language452Milestones453

Tager-Flusberg et al. (2009) set out criteria for determining 454 whether children with ASD undergoing an intervention for 455 expressive language can be considered to have made pro-456 gress from one broad stage of language development to the 457 458 next. All children in the current study would be considered to be in the pre- or minimally verbal stage prior to inter-459 vention, producing infrequent communicative acts, using 460 very few words, and no word combinations. All but one 461 scored below 15 months on the Expressive Language Age 462 Equivalent score of the Vineland Adaptive Behavior Scales 463



Fig. 2 Mean (and standard error) Number of words spoken by parent report on *CDI* (Fenson et al. 2007) at three time points





**Fig. 3** Mean (and standard error) Number of words produced during *CSBS* Behavior Sample (Wetherby and Prizant 2003) at three time points

464 (one child scored at 18 months) before treatment. In order
465 to be considered to have progressed to the next stage, that
466 of first words, Tager-Flusberg et al. suggest the following
467 criteria:

- 468 Language age equivalent ≥ 15 months, or five different word types and 20 word tokens in spontaneous speech
- 471 Production of consonant-vowel syllables, or production
  472 of 4 different consonants in spontaneous speech
- 473 Expression of at least two different communication
  474 functions (e.g., request, comment, social interaction)
  475 with words in spontaneous speech.

476 We examined outcomes in the current study to deter-477 mine how many participants in each group met these cri-478 teria. Five of the ten children who received RMIA met the 479 benchmarks; al Vineland Expressive Language Ageequivalents above 15 months (the child who started at 18 480 481 months achieved 30 months on this measure after 482 12 weeks of treatment), parent report of more than 35 483 words on the CDI; more than 7 different word types pro-484 duced during a CSBS play session (all but one of the five 485 were above 35 tokens), as well as expression of at least two 486 different communicative intentions with words, and four 487 different consonants used in CV syllables by the post-488 treatment assessment. All 5 retained or exceeded these 489 levels at the maintenance assessment. For the group 490 receiving MCT, 5 of the twelve children reached the

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Percent of Participants Achieving First Stage of Language
Acquisition Post-Tx



Fig. 4 Percent of participants in each treatment group who met Tager-Flusberg et al.'s (2009) criteria for first stage of language development at post-treatment

benchmarks of Vineland Expressive Language Age-491 equivalents above 15 months, parent report of more than 492 20 words on the CDI; more than 5 different word types 493 produced during a CSBS play session (all but one of the 494 five were above 50 tokens), as well as expression of two 495 different communicative intentions in words, and four 496 different consonants used in CV syllables by the post-497 treatment assessment. Again, all 5 retained or exceeded 498 these levels at the maintenance assessment. Figure 4 pre-499 sents the percentage of participants in each group who 500 achieved these milestones to acquire a basic form of 501 functional spoken language after intervention. 502

#### Moderator Variable Analysis

504 To investigate possible moderating effects on response to treatment, we performed multivariate linear models in 505 which CDI post-treatment was regressed on treatment 506 group (represented as a dummy variable, with 0 = RMIA507 and 1 = MCT), a moderator covariate, and an interaction 508 of treatment and moderator, using Preacher's calculator 509 (Preacher et al. 2006; http://www.quantpsy.org/interact/ 510 mlr2.htm). The results of these analyses appear in Table 3. 511 512 Examining these data suggests, first, that for all the pur-513 ported moderators except age, the model provided a good 514 fit to the data, as seen in the significant F statistics for these models. We then pursued further moderator analyses for 515 the variables for which a good fit was attained. 516

When these variables-Mullen VR, EL and RL age-517 equivalent scores, Vineland EL and RL age-equivalent 518 519 scores, % correct Motor Imitation, % parent responsiveness in Parent-Child Interaction, CSBS Joint Attention and 520 Play-were used as moderators of treatment, only CSBS 521 Joint Attention (JA) scores showed a main effect (p < 01). 522 This effect suggests that children with higher JA pre-523 treatment scores pre-treatment did better than those with 524 lower scores, regardless of which treatment was 525 administered. 526

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Treatment group	Mean (ar Mullen V	nd SD) /R AE <sup>1</sup>	Mean (an VABS-II I	d SD) EL AE <sup>2</sup>	Mean (an VABS-II I	d SD) RL AE <sup>3</sup>	Mean (and SD) <i>CSBS</i> <sup>4</sup> : number of spoken words		Mean (and SD) <i>CDI</i> <sup>5</sup> number of spoken words by parent report		
	Post-Tx	Maint.	Post-Tx	Maint.	Post-Tx	Maint.	Post-Tx	Maint.	Post-Tx	Maint.	
RMIA	*	30.0 (13.2)	1.2 (0.6)	1.6 (1.1)	1.3 (0.8)	1.7 (1.2)	5.1 (5.2)	6.6 (6.7)	88.6 (106.8)	119.3 (136.3)	
MCT	*	30.5 (6.7)	1.1 (0.4)	3.0 (3.0)	1.4 (0.6)	3.4 (4.7)	5.0 (6.4)	8.4 (8.2)	75.1 (89.3)	121.1 (123.4)	

Table 3 Outcome variables at post-treatment (Post-Tx) and maintenance (Maint.) time points

<sup>1</sup> Mullen Scales of Early Learning (Mullen 1995) Visual Reception Age-equivalent score (months)

<sup>2</sup> Vineland Adaptive Behavior Scales-II (Sparrow et al. 2005) Expressive Language Age-equivalent score (m

<sup>3</sup> Vineland Adaptive Behavior Scales-II (Sparrow et al. 2005) Receptive Language Age-equivalent score (mol

<sup>4</sup> Communication and Symbolic Behavior Scale-Developmental Profile (Wetherby and Prizant 2003) Spoken Word (Type) Inventory during 20 min. play session

<sup>5</sup> MacArthur-Bates Communicative Development Inventory (Fenson et al. 2007)

Table 3 also shows that the interaction terms were not

\* Mullen data were not collected at Post-Treatment

527

528 statistically significant for Mullen VR and EL, Vineland 529 EL, % correct Motor Imitation, % parent responsiveness in 530 Parent-Child Interaction, CSBS JA and Play, indicating 531 that levels of these pre-treatment moderator variables did 532 not affect the children's tendency to respond differentially 533 to one of our two treatments or the other. For Mullen Receptive Language and Vineland Receptive Language 534 535 age-equivalent scores, there were significant interaction 536 effects in these analyses, at p = 0.047 and p = 0.016, 537 respectively. To explore the interactions between these 538 moderator variables and treatment response, we ran a 539 Region of Significance (RoS) analysis (Bauer and Curran 540 2005, Preacher et al. 2006) to determine the range of values 541 of the moderator for which the relationship between our 542 CDI outcome and treatment was statistically significant. 543 The CDI-treatment relationship for any given value of the 544 moderator variable is described by the "simple intercept" 545 and "simple slope," which are functions of the moderator 546 that describe the effect of treatment on CDI for any par-547 ticular value of the moderator; the region of significance is 548 then defined to be the set of moderator values where the 549 simple slope is significantly different from zero. These 550 results are reported in Table 4.

551 The Region of Significance for Mullen Receptive Lan-552 guage was outside the interval from 0 to 18 months age-553 equivalent score, with a simple slope of -101.35 at the 554 lower endpoint of this interval and 218.37 at the upper 555 endpoint. Higher values of Mullen RL pre-treatment are 556 thus correlated with a stronger response to treatment under MCT than under RMIA (because the simple slope is 557 558 positive for these values), and lower values of Mullen RL 559 are correlated with greater improvement under RMIA 560 (because the simple slope is negative). For Vineland 561 Receptive Language age-equivalent scores, the RoS was 562 also outside the interval 0-18 months, with a simple slope of -173.19 at the lower endpoint and 77.91 at the upper 563 endpoint. These values are consistent with the results for 564 receptive language as measured by the Mullen and can be 565 interpreted analogously. Moreover, together these results 566 suggest that the cut-off score for deciding which treatment 567 to employ is a receptive language age-equivalent of about 568 18 months; with those scoring lower more likely to suc-569 ceed with RMIA while those scoring above an 18 month 570 571 level likely to do better with MCT. Graphs of the RoS analyses for these moderator variables appear in Fig. 5. 572

#### Discussion

573

Results of this study suggest that, on average, minimally574verbal preschoolers with ASD benefit from a relatively575brief treatment package including parent responsivity576training in conjunction with either577

- 1. discrete trial treatment enhanced with a behavioral<br/>momentum component in the form of rapid motor578579imitation antecedent training (RMIA), or580
- milieu communication training (MCT) focused specifically on eliciting speech.
   582

583 Gains made in these speech interventions were found to be maintained once the treatment concluded. Approxi-584 mately half the children in each treatment group progressed 585 from a classification of minimally verbal to the first stage 586 of spoken language development as defined by Tager-587 Flusberg et al. (2009). This finding suggests that at least 588 half of minimally verbal preschoolers like these with ASD 589 do seem to have the capacity to acquire spoken language as 590 591 a functional form of communication with focused intervention, strengthening the suggestion that this kind of 592 593 intensive speech treatment should be provided during the preschool period (Tables 5 and 6). 594



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Rapid motor imitation ante	cedent (RMI	A) Tx
CSBS <sup>1</sup>		
Pre-Post Tx <sup>+</sup>	1.7	Very large
Pre Tx-Maintenance <sup>+</sup>	.93	Large
Post T-Maintenance	.08	NS
$CDI^2$		
Pre-Post Tx <sup>+</sup>	1.0	Large
Pre Tx-Maintenance <sup>+</sup>	1.2	Very large
Post Tx-Maintenance	.25	Small
VABS <sup>3</sup> expressive language	ge	
Pre-Post Tx <sup>+</sup>	.82	Large
Pre Tx-Maintenance	.82	Large
Post Tx-Maintenance	.33	Small
Milieu communication trair	ning(MCT)	Гх

Table 4 Effect sizes of pair-wise comparisons across time points

Cohen's d\*

Effect Size descriptora

within treatment groups

Comparison

CSBS <sup>1</sup>		
Pre-Post Tx <sup>+</sup>	.73	Medium
Pre Tx-Maintenance <sup>+</sup>	1.2	Very large
Post Tx-Maintenance	.20	Small
$CDI^2$		
Pre-Post Tx <sup>+</sup>	.89	Large
Pre Tx-Maintenance <sup>+</sup>	1.3	Very large
Post Tx-Maintenance <sup>+</sup>	.42	Small
VABS <sup>3</sup> expressive language	e	
Pre-Post Tx <sup>+</sup>	.64	Medium
Pre Tx-Maintenance <sup>+</sup>	.91	Large
Post Tx-Maintenance	.65	Medium

<sup>a</sup> Cohen (1988)

<sup>+</sup> Statistically significant difference (p < .05)

<sup>1</sup> Communication and Symbolic behavior Scales-Developmental Profile (Wetherby and Prizant 2003)

<sup>2</sup> MacArthur-Bates Communicative Development Inventory (Fenson et al. 2007)

<sup>3</sup> Vineland Adaptive Behavior Scales-II (Sparrow et al. 2005)

595 Our moderator analyses show, first, that children with 596 better joint attention pre-treatment do better with either 597 treatment than do those with very low joint attention. This finding supports others in the literature suggesting that 598 599 language learning is mediated by joint attention (e.g., 600 Mundy et al. 1990; Paul et al. 2008; Watt et al. 2006). In 601 the context of the present study it suggests, further, that for 602 minimally verbal children, those with some joint attention 603 tend to respond to a treatment focused on eliciting spoken 604 communication, particularly when it is combined with 605 parent responsivity training. The frequency of joint atten-606 tional (JA) acts necessary to provide this mediation 607 appears, in our data, to be quite low; 55 % of the partici-608 pants in this sample showed NO JA during the CSBS. a semi-structured play session would seem to be a good candidate for some focused intervention to elicit spoken communication. 616

The 45 % who did produced between 1 and 4 JA acts

during the 20 min CSBS behavior sample. Even this low

level of JA initiation seems to enhance response to treat-

Our analyses also suggest that the level of pre-treatment 617 receptive language skill, as measured either directly on the 618 619 Mullen or by parent report on the Vineland, moderates response to these two treatments differentially. Children 620 who start out with relatives receptive skills do better 621 in with MCT treatment; unose who start out with relative 622 lower receptive skills do better with RMIA. MCT treatment 623 may work better for these relatively good comprehenders 624 because they are more able to deduce linguistic information 625 from relatively natural play-based interactions in which 626 words and referents are saliently matched. The children 627 with low receptive skills may do better in RMIA because 628 the less natural, more intensely structured DTT interactions 629 require less deductive ability and profine fewer stimuli to 630 distract the child from the word rebut relations being 631 presented. The Region of Significance finding suggests that 632 a receptive language level of about 18 months is necessary 633 to derive most benefit from MCT; children with less 634 receptive language, at least those who can be taught to 635 produce motor imitation, may do better with RMIA. 636

#### **Clinical Implications**

As we have seen, both these treatments had positive 638 effects, on average, for the severely impaired minimally 639 verbal preschoolers in this study, and we were able to 640 identify two elements to assist clinicians in matching 641 children to treatments. That is, the study suggests that 642 minimally verbal preschoolers with ASD with nonverbal 643 levels above 12 months who show some, even very limited, 644 expression of joint attention pretreatment are more likely to 645 respond to a speech-focused treatment than those who do 646 not. We would argue that any child meeting these criteria 647 should receive an intensive speech-focused treatment, in 648 addition to any transitional AAC program implemented, in 649 order to maximize the child's opportunity to acquire spo-650 ken language during this critical preschool period. Children 651 with virtually no joint attentional behaviors are less likely 652 to respond to speech-focused treatment and may derive 653 more benefit from an approach focused more intensely on 654 AAC. Second, for children for whom speech-focused 655 treatment appears indicated, those with receptive language 656 scores above 18 months may do better with MCT, while 657 those with lower levels of receptive language who are able 658 to master motor imitation can be tried with RMIA. Our 659

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Fig. 5 Plots of the interaction effect of moderator variable and treatment on CDI posttreatment. The *plot* on the *left* shows the effect when the moderator variable is Mullen Receptive Language; the plot on the right is for Vineland Receptive AE score. For both plots, the solid line compares CDI post-treatment between the two treatments (MCT = 0, RMIA = 1) at the *lower end* of the moderator variable's range, and the dashed line depicts the same for the upper end of the moderator's range



#### Table 5 Results of regression analyses

**Author Proof** 

Moderator variable	R <sup>2</sup>	F statistic for model fit	Intercept	Main effect of treatment	Main effect of moderator	Tx-Moderator interaction
Age	.414	2.821	49.573	38.041	14.468	-32.288
Mullen VR	.510	4.158*	44.096	26.822	6.777	1.290
Mullen RL	.544	4.764**	38.812	29.976	-3.810	11.842*
Mullen EL	.668	8.035***	59.814*	-2.704	9.422	3.363
Vineland receptive AE	.621	6.154**	43.756	34.028	-26.634	132.535*
Vineland expressive AE	.478	3.429*	46.795	16.543	12.035	129.264
CSBS joint attention	.800	16.03***	51.150**	23.756	48.340*	20.763
CSBS play	.652	7.500***	34.600	57.257	8.000	8.751
Motor skills	.567	5.228**	49.775*	22.305	1.441	0.255
PCI resp.	.525	3.868*	45.681	44.608	-1.560	3.838

parent report of expressive vocabulary size on the MacArthur-Bates Communicative Development Inventory (Fenson et al. 2007)

\* Significance at the 0.05 level

\*\* Significance at the 0.01 level

\*\*\* Significance at the 0.001 level

For each moderator variable, a linear regression model was fit with treatment (0 = MCT, 1 = RMIA), moderator, and treatment-moderator interaction as predictors and  $CDI^1$  post-treatment as response. Moderator variables for which the interaction term's p value was less than 0.05 were investigated further using Region of Significance analysis

Table 6	Results of the	Region of	Significance	analysis for	moderators	whose inte	eraction with	th treatment	is statistically	significant
---------	----------------	-----------	--------------	--------------	------------	------------	---------------	--------------	------------------	-------------

Moderator	Region of significance		Simple intercept	Simple slope	Simple intercept	Simple slope
	Minimum	Maximum	at minimum	at minimum	at maximum	at maximum
Mullen RL	-158.10	5.60	81.07	-101.35	-21.81	218.38
Vineland receptive AE	5 months	17.6 months	74.12	-117.06	-3.65	269.94

For both measures, the region of significance is outside the interval

results suggest that 3-5 sessions per week for 3-6 months is 660 661 a sufficient time frame to determine whether a child is 662 responding to a treatment. It is always possible to try a second approach if an intensive trial of the first does not 663

work, but these guidelines can be helpful in chosing an 664 initial program for young children with ASD.

Apart from the guidance in choosing intervention 666 approaches provided by this study, we believe it has a 667

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671 communication were being assigned to augmentative and 672 alternative modes of communication (AAC) approaches 673 without a concentrated effort to elicit functional speech. 674 We do not debate the value of AAC for many children with 675 severe speech impairments, nor do we argue against the use 676 of AAC as a transitional modality for young children with 677 ASD or as a primary modality for older children who have 678 not acquired speech. But we do believe that the unique 679 learning challenges seen in ASD may constitute a special case when it comes to providing communicative opportu-680 681 nities for minimally verbal young children. That is, the 682 deficits that are unique to early communication in ASD, 683 including low level of social motivation inherent in the 684 autistic syndrome, reduced attention to child-directed 685 speech (Paul et al. 2007), immaturity of speech motor 686 development (Gernsbacher et al. 2008), reduced engage-687 ment in reciprocal babbling (Paul et al. 2011), an inability 688 to use gaze cues to discern the relations between a speak-689 er's words and their intended referents (Baron-Cohen et al. 690 1997) and generally poor imitation skills (Rogers et al. 691 2005) may lead, in some children, to lack of sufficient 692 attention to others' verbal output and motor speech patterns 693 along with fewer attempts to use these patterns for com-694 municative purposes. These conditions can result in a 695 child's both trying less often and therefore getting less 696 practice in articulating speech and tending to rely on less 697 precise vocalizations and gestures for the few attempts that 698 are made. If this view is correct, then intervention that 699 actively focused attention on speech production and 700 enabled the child to learn through intensive guided practice 701 to produce a few accurate word forms, combined with 702 parent training to provide distributed opportunities for the 703 child to observe the connections between words and their 704 referents in affectively engaging settings, may be enough to 705 "turn on" the speech learning process, which may help 706 explain why for the children in this study who responded to 707 treatment, they tended to go on to acquire words that were 708 not explicitly taught in the intervention (See Tsiouri et al. 709 2012 for details). We have referred to this process as a 710 "speech insight," which could, in the context of respon-711 sive parent interactions, lead not only to the use of newly 712 learned words in generalized settings, but to an expansion 713 of word use beyond those taught in the intervention, as the child begins to "tune in" to words in the environment, to 714 715 see their connections to pleasing objects and activities 716 through responsive parent interactions, and to use newly 717 gained vocal output skills to practice and refine more word 718 productions. 719 All this suggests that, for minimally verbal children with 720 ASD, it may not be speech motor difficulty that obstructs

second set of implications. This study was motivated in

part by a concern on our part that too many preschoolers

with ASD who did not spontaneously acquire spoken

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the acquisition of useful speech, as some have suggested 721 722 (e.g., Gernsbacher et al. 2008; Velleman et al. 2009). It may be, rather, the failure to seek out opportunities for 723 reciprocal interactions mediated by vocal and verbal 724 exchanges, to "tune in" to speech models, and to "tune 725 726 up" production through emulation of significant others and 727 extensive practice in myriad playful interactions. This "speech attunement" framework has been supported in 728 several studies of early speech development in young 729 730 children with ASD carried out in our laboratories (Schoen 731 et al. 2009, 2011; Shriberg et al. 2011), which suggest that when young children with ASD learn to speak, their speech 732 733 skills are commensurate with and driven by their language abilities and they show no evidence of apraxic or speech 734 motor disorders in their verbal productions. 735

736 We would suggest, then, that the results of the current study should encourage clinicians to provide intensive, 737 speech-focused intervention for minimally verbal pre-738 schoolers with ASD who show at least a modicum of joint 739 attention behavior. For those with low receptive language 740 and the capacity to learn motor imitation, RMIA may be a 741 good choice as an initial intervention approach. For those 742 with better receptive language but otherwise limited spo-743 ken output, MCT may be the more appropriate option. For 744 those children without joint attention behaviors, a focus on 745 AAC modalities should be accompanied by attempts to 746 747 elicit the initiation of joint attention. When such behaviors 748 do begin to emerge, it would make sense to attempt speechfocused treatment at that point in time. 749

We believe our results also argue for including training 750 in parent responsiveness as an accompaniment (not a 751 substitute) to clinician-delivered intervention. We believe 752 for these severely involved preschoolers, direct, focused 753 treatment-which requires carefully shaping vocal behav-754 755 ior into intelligible speech, choosing words to introduce that are within the child's phonological zone of proximal 756 development, and withholding reinforcement when targets 757 are not accurately met-necessitates the skill of a trained 758 intervention agent. However, we also believe that 759 expanding upon the gains made in these clinician-delivered 760 761 sessions, by providing multiple opportunities for practice of spoken communication in an enjoyable interaction with 762 highly positive affective valence wit peatly enhance the 763 effect of the clinician-delivered intervention. Although pre-764 765 treatment parent responsiveness did not moderate treatment in our study, perhaps due to the small sample size, Aldred 766 et al. (2012) recently reported that when parent respon-767 siveness to children with ASD improved, children were 768 more likely to show a positive effect of communication 769 intervention. This finding supports our intuition that pro-770 viding parent responsiveness training within our interven-771 772 tion packages led to greater improvement in children's 773 response to the clinician-delivered treatment.

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#### 774 Limitations and Future Research

A primary limitation of the current study was the small 775 776 sample size. The strict entry criteria, involving very limited 777 spoken language in conjunction with at least a 12 month 778 level in nonverbal cognition, as well as the logistical dif-779 ficulties of providing an intervention that required clinic 780 attendance in addition to the child's ongoing school pro-781 gram, made recruitment difficult. The power to find dif-782 ferences in outcomes between the two treatments, as well 783 as to identify moderators of response was thus limited. 784 Future research with larger samples may enable more 785 precise information about moderators that assist in 786 matching children to specific treatment approaches. A 787 second limitation was the decision to assign children non-788 randomly to the MCT condition if they could not master 789 the motor imitation skills necessary for RMIA treatment. 790 This resulted in our inability to use a completely random-791 ized design, which limits the generalizability of the find-792 ings. Despite this difficulty, we believe that comparisons of 793 differing treatment methods, including contrasting DTT 794 methods with more naturalistic approaches, is an essential 795 element of treatment research. Without such direct com-796 parisons, more effective matching of children to treatments 797 will not be feasible.

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