



A review on measuring treatment response in parent-mediated autism early interventions

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Abstract: This review describes and evaluates measures aimed at assessing treatment response in parent-mediated early interventions targeting autism spectrum disorder (ASD). In order to be able to evaluate the relative utility of different interventions, it is critical to have access to tools that identify treatment effects. Measures across domains of child outcomes, parent or family outcomes, and parent-child interaction outcomes are evaluated by summarizing their roles in previous studies, and describing the consistency of treatment effects. In particular, some variables included in parent-child interaction outcomes generated relatively consistent treatment effects. Considerations for future research are discussed, including cultural considerations, challenges associated with the current set of outcome measures, and an emphasis on enhancing ecological validity.

Keywords: Autism spectrum disorder (ASD); early intervention; treatment outcome

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For years, parent-mediated interventions have been recognized as pivotal treatments for young children with autism spectrum disorder (ASD), targeting developmental and social communication skills (1). In particular, these models of intervention are especially relevant for very young children on the spectrum. The importance of intervening early in this age range cannot be understated given the neuroplasticity associated with infancy and toddlerhood (2). Summary statements on best practices for early intervention in children with ASD (3) and the American Academy of Pediatrics Council on Children with Disabilities (4) both contend that a family/caregiver component should be included in effective early intervention, and emphasize the importance of culturally competent care that focuses “on the family as well as on the child.” In order to evaluate the therapeutic value of such interventions, it is important to examine outcomes of studies aimed at investigating effectiveness. Subsequently, it is critical that outcome measures are sufficiently sensitive so as to assess response to treatment in a clinically meaningful way. In this review, methods for measuring treatment response in parent-

mediated early interventions for ASD are examined and discussed, before providing recommendations for future studies.

ASD is a neurodevelopmental disorder characterized by deficits in social communication and social interaction, as well as the presence of restricted, repetitive patterns of behavior or interests (5). As the prevalence of ASD has steadily risen, so has the number of individuals seeking to access services, in the absence of a comparable rise in the supply of relevant services. This has contributed to shortages in availability of evidence-based services for ASD [e.g., (6)] as well as long wait lists. In particular, caregivers of children with ASD (as compared to caregivers of children with other developmental disabilities/mental health concerns) endorse burdens including difficulty using services, lack of source of care, sub-optimal insurance coverage, and adverse family impact (7). Given the barriers and challenges associated with gaining access to early interventions for ASD, parent-mediated interventions represent an avenue of treatment that can facilitate a wider reach, increasing access to care. Parent-mediated interventions involve professionals

teaching parents therapy techniques that they can use with their child in the home and other settings. Some of these interventions have demonstrated the ability to improve the child's communication and social interactions (8).

Three relatively recent systematic reviews and meta-analyses spanning treatment approaches have identified a set of parent-mediated early interventions for ASD (i.e., children between the ages of 1 and 6) that have been evaluated for efficacy of the intervention including a control or comparison group (8-10). These reviews have consisted of studies where parents or caregivers primarily mediated the intervention, and where the intervention focused on parent implementation. In order to adequately evaluate outcome, a control or comparison group (e.g., no treatment or treatment as usual) was necessary. In the current paper, *Table 1* depicts parent-mediated early interventions for ASD (listed with corresponding outcome studies) to be explained in greater detail below.

These studies included outcomes consisting of child, parent or family, and parent-child measures. The following sections describe a non-comprehensive sample of tools which have been employed to measure response to treatment in parent-mediated early interventions for ASD, as well as treatment response reported by studies that have used these measures. The sample is meant to represent a range of options for outcome measures that cover several domains of functioning. Oono and colleagues' systematic review (10) describes significantly more details regarding treatment response for some of the measures mentioned herein, including forest plots which summarize effects of targeted outcomes across studies (e.g., assessment of child receptive language, parent report of child communication, autism severity).

One important limitation of this body of research relates to cultural applicability. The studies described were conducted nearly exclusively in Western settings, in the English language. Additionally, the outcome measures have been developed and standardized in Western countries. This unfortunately limits the ability to generalize findings to other settings and cultures. It has been well established that high income countries, and individuals of high socioeconomic status, are overrepresented in autism research, and this particular area of research is no exception (37). Various barriers have been identified as making it difficult for individuals from low-resource settings to access evidence-based therapies, but it would nonetheless be essential to conduct similar studies in diverse settings to be able to speak to the generalizability of findings discussed

below, and more generally to improve access to services worldwide.

Another key point is that not all parent-mediated interventions are created equally. Some programs exclusively involve parent coaching, for multiple sessions per week over months or years, while other programs embed a smaller dose of parent coaching into a program also including therapist delivered intervention. A common duration and intensity of parent coaching is one session per week for approximately 12 weeks, though there is a wide range of dosage in interventions that have been tested, which is a known factor that can impact outcomes. While specifically evaluating interventions is outside the scope of this paper, it is an important consideration when comparing outcome measures.

Child outcomes

Child measures have generally sought to characterize treatment response in terms of language development (receptive and expressive; directly tested and parent- or teacher-reported), social communication, developmental/cognitive ability, adaptive behavior, and child behavior (maladaptive behavior and ASD symptoms).

Language development

Receptive language has been directly assessed through the Reynell Developmental Language Scales, Third Edition [(38); current version is the New Reynell Developmental Language Scales], whereas it has been measured via parent report with the MacArthur Communicative Development Inventory [(39); current version is the MacArthur Bates Communicative Development Inventories]. The Reynell Developmental Language Scales, Third Edition measures both expressive and receptive language in children between the ages of 1–6, and takes between 35 and 60 minutes to administer. The MacArthur Communicative Development Inventory is designed to assess communication in infants and toddlers (to age 2 years, 6 months) via parent report using a checklist of 89 to 100 words.

For receptive language on the Reynell Developmental Language Scales, Third Edition, Roberts and colleagues (12) reported that all groups (home-based, center-based combined with parent training, and a non-treatment comparison group) showed non-significant improved raw scores, while the center-based group also showed a non-significant increase in standard score. The center-based

Table 1 Parent-mediated early interventions for young children with ASD

Intervention	Study assessing efficacy/ effectiveness	Outcome measures described in current paper
ABA ¹ vs. PT ²	(11)	Child Behavior Checklist
Building Blocks	(12)	Reynell Developmental Language Scales, Third Edition; Pragmatics Profile of Everyday Communication; Developmental Behavior Checklist; Parenting Stress Index; Parent Perception Questionnaire; Beach Family Quality of Life Questionnaire
Child's Talk Project	(13)	MacArthur Communicative Development Inventory; Vineland Adaptive Behavior Scales; Autism Diagnostic Observation Schedule; Parenting Stress Index; parent-child interaction
Day care plus parent training vs. day care alone	(14)	Early Intervention Developmental Profile; Preschool Developmental Profile; Autism Behavior Checklist; Stress-Arousal Checklist; Client Satisfaction Questionnaire
DIR ³ /Floortime	(15,16)	Preschool Language Scales; Comprehensive Assessment of Spoken Language; Child Behavior Rating Scale; parent fidelity; Functional Emotional Developmental Questionnaire; Childhood Autism Rating Scale; Functional Emotional Assessment Scale
Early Start Denver Model (ESDM)	(17-19)	Autism Diagnostic Observation Schedule; Mullen Scales of Early Learning; Vineland Adaptive Behavior Scales; Child Behavior Checklist; parent satisfaction; parent fidelity; parent-child interaction
Focused Playtime Intervention	(20)	Parent-child interaction
Hanen's More Than Words	(21)	Early Social Communication Scales; Mullen Scales of Early Learning; Vineland Adaptive Behavior Scales; Parent Interview for Autism—Clinical Version; Consumer Satisfaction Survey; parent-child interaction
Home-based program	(22)	Preschool Behavior Checklist
Joint Attention Symbolic Play Engagement and Regulation (JASPER)	(23-25)	Parent self-rated confidence and comfort; parent-child interaction; Parenting Stress Index
Parent Focus Training	(26)	Autism Diagnostic Interview-Revised
Parent-mediated Communication-focused Treatment (FACT)	(27)	Preschool Language Scales; MacArthur Communicative Development Inventory; Communication and Symbolic Behavior Scales-Developmental Profile; Vineland Adaptive Behavior Scales; Autism Diagnostic Observation Schedule; parent-child interaction
PLAY: Play and Language for Autistic Youngsters	(28)	Child Behavior Rating Scale; Autism Diagnostic Observation Schedule; Parenting Stress Index; Functional Emotional Assessment Scale
Preschoolers with Autism	(29,30)	Reynell Developmental Language Scales, Third Edition; Psychoeducational Profile-Revised; Vineland Adaptive Behavior Scales; Developmental Behavior Checklist-Autism Screening Algorithm; Childhood Autism Rating Scale; McMaster Family Assessment Device; General Health Questionnaire
Pivotal Response Training (PRT)	(31,32)	Vineland Adaptive Behavior Scales; parent satisfaction; parent confidence; parent fidelity; parent-child interaction
Social ABCs	(33)	Parent-child interaction
Social Communication, Emotional Regulation, and Transactional Supports (SCERTS)	(34)	Vineland Adaptive Behavior Scales; Autism Diagnostic Observation Schedule
Treatment and Education of Autistic and Related Communication Handicapped Children (TEACCH)	(35)	Parenting Stress Index; Mullen Scales of Early Learning
VIPP-AUTI: Video Intervention to promote Positive Parenting for children with Autism	(36)	Early Social Communication Scales

¹Applied Behavior Analysis, ²Parent Training, ³Developmental, Individual-differences, & Relationship-based. ASD, autism spectrum disorder.

intervention consisted of 40 weekly 2-hour sessions (a playgroup of 4–6 children, alongside a concurrent parent support and training group). The home-based intervention consisted of a 2 hour visit every other week, over a period of 40 weeks (20 sessions total), where staff worked with the family to address individual needs such as speech, sensory, and social skills. Somewhat similarly, in other studies, both the treatment and control groups showed improvement on the MacArthur Communicative Development Inventory, with no significant effect of group in a treatment program involving monthly parent-child treatment sessions for 6 months followed by 6 months of less frequent sessions (13). Additionally, improvement was shown on the Reynell Developmental Language Scales, Third Edition, with no significant group effect following a comparison of Preschoolers with Autism intervention of ten 90-minute small group sessions, and ten 60-minute individual family sessions, to active control and control groups (29).

Expressive language has been directly assessed through the Preschool Language Scales, Fourth Edition (40), Comprehensive Assessment of Spoken Language (41), and Reynell Developmental Language Scales, Third Edition. Additionally, the MacArthur Communicative Development Inventory has been used as a parent report measure. The Preschool Language Scales, Fourth Edition measures developmental language skills in young children (birth through age 6 years, 11 months) and takes between 20 and 45 minutes to administer. Finally, the Comprehensive Assessment of Spoken Language is administered to children as young as 3 years of age (through age 21) and measures a range of language processing skills, including comprehension, expression, and retrieval. The Comprehensive Assessment of Spoken Language takes approximately 30 to 45 minutes to administer.

With regard to expressive language findings, Aldred and colleagues (13) reported that the treatment group showed significant progress after one year of intervention relative to the control group (treatment program involving monthly parent-child treatment sessions for 6 months followed by 6 months of less frequent sessions), across various levels of severity and age. However, in several other studies, improvement was present across groups and did not differentiate the intervention from control. For example, no significant effect was observed in a standardized, directly assessed measure of language (Preschool Language Scales), although parent report (MacArthur Communicative Development Inventory) indicated that the Preschool Autism Communication Trial intervention group (18

sessions over 6 months, plus parents were asked to do 30 minutes of home practice per day) demonstrated an improved outcome relative to the control group [similar results were reported for receptive language (27)]. For those assigned to either an intervention focused on socialization and parent-child interaction (DIR-based approach involving 2 hours of therapy and coaching per week for 12 months) compared to a community treatment group, both groups showed improvement according to either the Preschool Language Scales or Comprehensive Assessment of Spoken Language, with no significant group effect (15). Finally, and similarly, one study indicated that there was no distinct benefit of adding a parent education component to an intervention targeting language development (Preschoolers with Autism intervention of ten 90-minute small group sessions, and ten 60-minute individual family sessions), as there was no group effect (29).

Social communication

Several studies have considered aspects of a child's social communication as primary outcomes. Social communication has been directly assessed with the Early Social Communication Scales (42) and the social composite of the Communication and Symbolic Behavior Scales-Developmental Profile (43), whereas it has been assessed through parent report with the Vineland Adaptive Behavior Scales (VABS) (44) and the Pragmatics Profile of Everyday Communication (45). Additionally, a modified version of the Child Behavior Rating Scale (46,47) has also been used to assess a child's social communication ability, which additionally characterizes behavior problems and parent-child interaction/reciprocity.

The Early Social Communication Scales is a structured assessment of joint attention skills used with young children whose developmental skills fall between 8 and 30 months. It takes approximately 15–25 minutes to administer. The Communication and Symbolic Behavior Scales-Developmental Profile is appropriate for use with infants and toddlers whose functional communication skills range between 6 and 24 months, and evaluates social communication, expressive language, and symbolic functioning. The VABS allows parents to report on their child's adaptive skills across domains of Communication, Daily Living Skills, Socialization, and Motor Skills, and can be used with individuals ranging from birth to 90 years of age. The primary goal of the Pragmatics Profile of Everyday Communication is to better understand how a child

communicates in everyday life, and can be administered to parents of children up to age 10. The four sections of the interview include Communicative Functions, Response to Communication, Interaction and Conversation, and Contextual Variation. Finally, the Child Behavior Rating Scale is used to assess children's social interactive behavior with their parents, including attention to the activity during joint interactions with the parent, involvement, cooperation, joint attention, and enjoyment of the activity.

At outcome, one study observed a significant group effect favoring the Preschool Autism Communication Trial intervention (18 sessions over 6 months, plus parents were asked to do 30 minutes of home practice per day) for parent report of their child's social communication (using the Communication and Symbolic Behavior Scales-Developmental Profile social composite) (27). Additionally, a different study showed that children who received Hanen's 'More Than Words' intervention of eight weekly group sessions and three individual sessions, which focused on teaching parents skills, showed greater frequency of increasing joint attention and requesting behaviors (measured via Early Social Communication Scales), but only for a set of children (i.e., those with low object interest at baseline) (21). The Early Social Communication Scales detected improvement in initiating joint attention at the group level across time for those receiving the Video-feedback Intervention to Promote Positive Parenting adapted to Autism, involving five home visits of 60–90 minutes each over three months, but again only for a subset of children (i.e., those who did not attend school) (36).

Aldred and colleagues (13) failed to demonstrate a treatment effect in child's social communication skills, as defined by the Communication domain of the VABS, although they reported a trend in the direction of the treatment group (monthly parent-child treatment sessions for 6 months followed by 6 months of less frequent sessions) showing a stronger effect. The Communication domain of the VABS was also used in a study comparing Pivotal Response Treatment Parent Training Group to a psychoeducation group. In this study, a significant treatment effect was demonstrated, where those in the Pivotal Response Treatment Parent Training Group, consisting of eight 90-minute parent only group sessions and four 60-minute parent-child dyad sessions, demonstrated greater improvement (31).

Roberts and colleagues used the Pragmatics Profile of Everyday Communication and concluded that all three groups in the study (center-based which included parent

training and support, home-based which sought to train and support parents, and waitlist) made statistically significant improvements over the course of the study, with no effect for group (12). In Casenhiser *et al.* (15), participants in the study's treatment group, which emphasized social and parent-child interaction using DIR principles in 2 hours of therapy and parent coaching per week for 12 months, showed significantly greater improvement than the community treatment control group over one year's time on the majority of Child Behavior Rating Scale items (e.g., cooperation, joint attention, and enjoyment of the activity). Children in the PLAY Project intervention group, whose parents received monthly coaching for 12 months, showed improved interaction skills in the home setting in the areas of increased shared attention and initiation (28). Overall, several studies have shown improvements in child social communication, although sometimes only in a set of participants, and other times while not differentiating treatment and control groups.

Developmental/cognitive ability

With regard to developmental/cognitive assessments, the Mullen Scales of Early Learning (MSEL) (48), Wechsler Preschool and Primary Scale of Intelligence-Revised (49), Bayley Scales of Infant Development, Second Edition (50), Early Intervention Developmental Profile (51), Preschool Developmental Profile (51), and Psychoeducational Profile-Revised (52) have assessed cognitive/developmental outcomes including nonverbal reasoning, language, and motor skills. These measures assess similar constructs, although the age ranges differ somewhat. Appropriate age ranges for administration are as follows: MSEL, birth to 68 months; Wechsler Preschool and Primary Scale of Intelligence-Revised, 30 months to 91 months; Bayley Scales of Infant Development, Second Edition, 1 to 42 months; Early Intervention Developmental Profile, birth to 36 months; Preschool Developmental Profile, 36 to 72 months; Psychoeducational Profile-Revised, 6 to 84 months.

Additionally, the PATH Curriculum Checklist (53) has been used in a similar manner, although it is not a standardized instrument. The PATH Curriculum Checklist is a criterion-based tool for assessing child development, delivered in a play-based format. It covers many domains associated with ASD including receptive language, expressive language, joint attention, cognition, and play skills.

Results from a standardized measure of developmental functioning (MSEL) ranged from demonstrating a significant treatment effect, to a significant overall effect, to no significant findings. For children who received high intensity intervention via the Early Start Denver Model (ESDM) which also included parent coaching (20/hours per week delivered by therapist, 5 hours/week of parent coaching, for two years), they showed a significantly greater increase in MSEL score relative to the community treatment group at both 1 year and 2 year outcome (17). In a comparison of ESDM delivered via standard parent coaching (P-ESDM) *vs.* enhanced parent coaching with an additional 90-minute session per week, multimodal materials, and motivational interviewing, both groups showed improvement in MSEL developmental quotient score, with no group by time interaction (19). Participants in a Home TEACCHing Program intervention (training parents for 12 90-minute sessions) showed gains in developmental skills (MSEL) over the treatment period, particularly in expressive language, with the waitlist group showing similar improvement (35). However, Carter and colleagues (21) did not identify an effect of Hanen's 'More Than Words' intervention (eight weekly group sessions and three individual sessions, over three months, focused on teaching parents skills) on child's developmental functioning measured with the MSEL.

Jocelyn and colleagues (14) used the Early Intervention Developmental Profile and Preschool Developmental Profile to evaluate developmental outcome. In both the treatment (caregiver-based intervention in community day-care settings, where caregivers received psychoeducation and support) and control groups, developmental skills increased across domains. One significant group by time interaction emerged whereby the intervention group showed significantly greater change in the language domain.

No significant group differences on the Psychoeducational Profile-Revised emerged for those who received parent education and counseling *vs.* parent education and behavior management (Preschoolers with Autism 20-week program of ten 90-minute small group sessions, and ten 60-minute individual family sessions) *vs.* control (29). With regard to the ESDM PATH Curriculum Checklist, both the standard P-ESDM and enhanced P-ESDM groups showed significant improvements in overall score, with no group by time interaction effect. Importantly, children of parents who demonstrated better fidelity to the treatment showed relatively more

developmental gains.

Adaptive behavior

In addition to measuring social communication skills as mentioned above (through Communication domain), the VABS (44) has also been used extensively to capture changes in adaptive functioning more broadly, often as the primary outcome measure in these studies. In one such example, at two-year follow-up, Dawson and colleagues (17) showed that the ESDM group (20/hours per week delivered by therapist, 5 hours/week of parent coaching, for two years), had gained adaptive skills at the same rate as typical peers, while the community treatment group continued to fall even more behind over time. Tonge and colleagues (29) also noted positive findings, whereas after controlling for baseline scores and child age, the Preschoolers with Autism intervention group of ten 90-minute small group sessions, and ten 60-minute individual family sessions showed significantly greater improvements in communication, socialization, and daily living skills for children who were initially very delayed, relative to the counseling and control groups. Additionally, individual intervention (parent-implemented Early Social Interaction, two to three times per week for 9 months) led to relatively stable or a slight increase in VABS Daily Living and Socialization scores, while group intervention (parent-implemented Early Social Interaction, once weekly for 9 months) led to a decrease in scores (34).

In a study testing the Hanen's 'More Than Words' intervention consisting of eight weekly group sessions and three individual sessions, focused on teaching parents skills, no significant group effect was found for the VABS, which in part contributed to the authors' question regarding the appropriateness of Hanen's 'More Than Words' for very young toddlers with ASD (21). Similarly, for teacher report on the VABS, Green and colleagues (27) did not identify a significant group difference (Preschool Autism Communication Trial; 18 sessions over 6 months) in the Adaptive Behavior Composite change score. Finally, the enhanced parent coaching intervention in Rogers and colleagues (19), which added an additional weekly P-ESDM session, multimodal materials, and motivational interviewing to a standard course of P-ESDM, did not result in greater gains in VABS scores relative to standard parent coaching. As such, some treatments/programs have reported significant findings as reflected on the VABS, while

others have not detected effects.

Child behaviors

Studies assessing treatment response in parent-mediated early interventions for ASD have considered two general categories of child behaviors: maladaptive or difficult behaviors, and autism symptoms/characteristics. The following parent-report measures have sought to identify changes in maladaptive behaviors in response to treatment: Preschool Behavior Checklist (54), Developmental Behavior Checklist (55), and Child Behavior Checklist (56). The Preschool Behavior Checklist is a screening tool for emotional and behavioral problems in preschool age children, frequently completed by teachers and other school professionals. The Developmental Behavior Checklist is designed to assess behavioral and emotional problems in individuals with developmental and intellectual disabilities, and can be completed by caregivers or teachers. Finally, the Child Behavior Checklist is a caregiver report measure which identifies problem behaviors in children across a range of domains, and can be used with children ranging from 18 months (preschool version) through 18 years (school-age version).

In Rickards *et al.* (22), comparing center-based (children received 5 hours of intervention spread over two sessions weekly during the school year) *vs.* home-based (additional weekly 1.5 hour sessions with a specialist preschool teacher for 40 weeks, who worked with the child and demonstrated strategies to parents) *vs.* control groups, early intervention teachers rated children in the home-based treatment group as showing improved behavior on the Preschool Behavior Checklist relative to those in the control group. Roberts and colleagues (12) did not identify significant group differences in outcomes on the Developmental Behavior Checklist (home-based which sought to train and support parents, center-based combined with parent training, and a non-treatment comparison group). However, when using a modified version of the Developmental Behavior Checklist (a subset of items related to symptoms of autism; Developmental Behavior Checklist-Autism Screening Algorithm), Tonge and colleagues (29) reported a significant main effect of their Preschoolers with Autism intervention (ten 90-minute small group sessions, and ten 60-minute individual family sessions), where participants in the parent education and behavior management group showed significantly lower scores than those in the control group. Rogers and colleagues (19), in a comparison of standard

12-session P-ESDM to enhanced P-ESDM adding: (I) a second weekly 1.5 hour in-home session; (II) materials offered in multiple modalities including text, website, visuals; and (III) motivational interviewing) used the Child Behavior Checklist to consider associated symptoms such as tantrums, sleep, and self-injury, and found that enhanced P-ESDM was not associated with greater improvement in associated symptoms. Finally, Smith and colleagues (11) found no significant differences in maladaptive behavior according to teacher report on the Child Behavior Checklist after comparing a year of intense therapist-delivered intervention of 30 hours/week to parent training of 5 hours/week.

In addition, the Functional Emotional Developmental Questionnaire (57) is a caregiver report form assessing emotional and intellectual functioning which involves functional development levels of shared attention and regulation, engagement/relating, purposeful emotional interaction, social problem solving, creating ideas, and thinking logically. When parents estimated their child's development via the Functional Emotional Developmental Questionnaire, Pajareya and colleagues (16) demonstrated that there was a significantly greater gain for the DIR/Floortime™-based intervention group (parents were trained in DIR/Floortime™ and asked to carry out techniques for 20 hours/week with their child) compared to the control group.

Changes in autism symptoms and characteristics have been measured via clinician observations with the Autism Diagnostic Observation Schedule, Second Edition (ADOS-2) (58) and Childhood Autism Rating Scale (CARS) (59), and via parent report with the Autism Diagnostic Interview-Revised (ADI-R) (60), Parent Interview for Autism-Clinical Version (61), and Autism Behavior Checklist (62). The ADOS-2 and ADI-R are gold standard tools for assessing and characterizing symptoms of ASD. The ADOS-2 involves a play-based or interview-based (depending on the individual's level of functioning) direct assessment of an individual's social communication skills, as well as restricted and repetitive behaviors. The ADI-R is a semi-structured interview which allows caregivers to report on their child's symptoms. The Parent Interview for Autism-Clinical Version is also a caregiver interview which was developed to identify behavioral change in young children with ASD. The CARS is a clinical rating scale which allows a clinician to make observations of a child's behavior which are specific to ASD. Finally, the Autism Behavior Checklist is a questionnaire completed by a caregiver or clinician which asks the rater to report on scales of relating, sensory, body and object use, language, and social

and self-help behaviors.

Rogers *et al.* (19) showed that although there was not a significant group difference (comparing standard 12-session P-ESDM to enhanced P-ESDM with additional 90-minute session per week, multimodal materials, and motivational interviewing), when the P-ESDM and enhanced P-ESDM groups were combined, a significant decrease emerged in pre- to post-treatment ADOS-2 scores. In studies that used a previous version of the ADOS-2, called the ADOS-G (63), Green and colleagues (27) reported that there was only a small effect of Preschool Autism Communication Trial intervention (18 sessions over 6 months) on ADOS-G score as it related to diagnostic threshold, and Dawson and colleagues (17) indicated that groups did not differ in ADOS severity score following either 1 or 2 years of treatment (treatment group involving 20 hours per week delivered by therapist, and 5 hours/week of parent coaching, for two years). Conversely, Aldred and colleagues (13) showed that the treatment group (monthly parent-child treatment sessions for 6 months followed by 6 months of less frequent sessions) showed a greater effect in change in ADOS-G score than did the control group after controlling for baseline score; most of the observed effect emerged within the Reciprocal Social Interaction subdomain. A Social Communication, Emotional Regulation, and Transactional Supports (SCERTS)-based intervention program (Early Social Interaction for 9 months: individual, 2–3 sessions per week *vs.* group, one session per week) demonstrated that children in both groups showed improvement in ADOS social affect, as well as increased symptoms in ADOS restricted and repetitive behaviors (34). Further, Solomon and colleagues (28) demonstrated improvement in autism symptomatology (ADOS-G) in response to monthly parent-mediated intervention over a 12-month controlled study. Although there is mixed evidence with some reflecting significant change in ADOS score over time, the ADOS was designed primarily as a diagnostic tool and may not pick up on subtle changes, especially in the short term.

On the CARS, Tonge and colleagues (29) found no treatment effect (parent education and counseling *vs.* Preschoolers with Autism parent education and behavior management *vs.* control), while Pajareya and colleagues (16) indicated that a CARS change score reflecting symptom improvement was significantly greater in the intervention group (parents received training in DIR/Floortime™ and were asked to carry out techniques for 20 hours/week) when compared to routine care. On the Autism Behavior Checklist in Jocelyn and colleagues' study (14) comparing

caregiver-based intervention in community day-care settings (consisting of caregivers receiving psychoeducation and support) to a control group involving no intervention, the psychologist's rating of autism symptoms did not differ by group following treatment; parents of children in both groups reported symptom improvement.

With regard to information on autism symptoms gleaned from parent interview, Drew and colleagues (26) reported no significant differences in symptom severity on ADI-R domain scores following one year of treatment, consisting of one 3-hour parent coaching session every 6 weeks. Children across both Hanen's 'More Than Words' intervention (eight weekly group sessions and three individual sessions, over three months, aimed at teaching parents skills) and control groups showed large improvements in nonverbal communication according to the Parent Interview for Autism-Clinical Version (21). Across these various measures of autism symptoms and characteristics, evidence is relatively mixed on whether positive outcomes result from parent-mediated early interventions for children with ASD. It is possible that these measures do not detect potential changes, the interventions do not produce substantial changes in this area, or the time of measurement did not allow for sufficient change to transpire in order to detect it. Additionally, the composition of the samples may have played a role [e.g., Tonge and colleagues (29) following the Preschoolers with Autism intervention noted that the CARS might have proven less useful in their particular age range].

Parent or family outcomes

Outcomes more broadly characterizing the parent/family include parent stress level, parent satisfaction with therapy, parent confidence (observed, and in managing disability/behavior problems), family functioning, and parent mental health. Additionally, some intervention programs have sought to characterize parent skill level in delivering the intervention, also known as fidelity.

Parent level of stress

Parent's level of stress has been measured via self-report through the Stress-Arousal Checklist (64) and the Parenting Stress Index (65). The Stress-Arousal Checklist is a self-report measure containing 45 adjectives which respondents rate as consistent/inconsistent with their current feelings (e.g., active, tense, peaceful). The Parenting Stress Index is designed for parents of children between the ages of 1

month and 12 years, and consists of 101 items which address stress related to child characteristics, parent characteristics, and situational/demographic factors.

In Jocelyn and colleagues' (14) comparison of caregiver-based intervention in community day-care settings (where caregivers received psychoeducation and support) to a control group involving no intervention, neither the treatment nor the control group experienced a significant change in Stress-Arousal Checklist scores over the course of treatment. Similarly, Aldred and colleagues (13) showed no group difference (active treatment of monthly parent-child treatment sessions for 6 months followed by 6 months of less frequent sessions *vs.* control) in change in total Parenting Stress Index score, nor did Welterlin and colleagues [Home TEACCHing Program, consisting of 12 90-minute parent training sessions *vs.* control (35)]. Although parent stress levels decreased over time for those in both the PLAY Project Home Consultation, involving monthly caregiver coaching for 12 months, and usual community services groups, there was no main effect for group (28). After adjusting for baseline differences, Roberts and colleagues (12) reported no significant change in stress level on the Parenting Stress Index across home-based (training and supporting parents) *vs.* center-based including parent training intervention group following the course of treatment, whereas the waitlist group showed a significant decrease in stress (although differences between groups were not present). And in one study comparing a naturalistic, developmental behavioral intervention (10 weekly hour-long sessions of parent training in Joint Attention, Symbolic Play, Engagement and Regulation) to a psychoeducational intervention, results actually indicated that parents in the psychoeducational group showed a relatively larger reduction in child-related (Parenting Stress Index) stress over time (25). Across these findings, it is not clear that parent-mediated early interventions for children with ASD impact parent stress level in a meaningful way, at least when assessing at short-term follow-up.

Parent satisfaction with therapy

In some studies, a secondary outcome has consisted of satisfaction with the therapy program. Jocelyn and colleagues (14) compared caregiver-based intervention in community day-care settings (where caregivers received psychoeducation and support) to a control group involving no intervention, using a set of four questions assessing satisfaction (e.g., "How much did the help you received

assist you in understanding your child's needs?"), and found that the intervention group reported higher levels of satisfaction with how the program increased knowledge of ASD, contributed to better understanding the child's needs, and contributed to better meeting the child's needs. Nefdt and colleagues (32) assessed ease of understanding, usefulness, and entertainment value, following a self-directed learning program in pivotal response treatment for parents, and reported high satisfaction ratings overall. Rogers and colleagues (19) used a questionnaire to gauge parents' perception of the intervention's utility in a number of areas, including promoting child language, ease of use, competence of staff members, and barriers to participation. Overall, parents were highly satisfied and there was no significant group difference in mean score across the standard treatment (12-session P-ESDM) and enhanced treatment (with additional 90-minute session per week, multimodal materials, and motivational interviewing) groups, although the medium effect size favored the enhanced treatment group. In their telehealth parent training study comparing 12 weekly 1.5-hour videoconference sessions with online access to P-ESDM materials to monthly 1.5-hour videoconference sessions with online access to alternative resources, Vismara and colleagues (18) administered a 20-item questionnaire to parents which evaluated the ease of use of the website, intervention content/learning tools, therapist support, and parent confidence in teaching new skills to children. Results indicated that parents in the P-ESDM group reported significantly higher satisfaction, confidence, and use of website than did parents in the community treatment-as-usual group. Finally, Carter and colleagues' (21) study testing Hanen's 'More Than Words' intervention developed and administered a Consumer Satisfaction Survey which consisted of 11 questions aimed at evaluating perception of the group leader, and 17 questions querying the group's climate (e.g., support, cohesion). Overall, consumer satisfaction was reported to be extremely high on both group leader experience and group climate.

Parent confidence-observed

One study coded a parent's level of confidence through observing his or her interactions with the child (32). The measure of observed parent confidence was adapted from Brookman-Frazee (66), and assessed whether the parent appeared certain of how to teach/interact with his or her child, completed following a 10-minute observation.

Results suggested that parents in the self-directed learning program in pivotal response treatment were rated to be more confident in interacting with the child as compared to parents in the wait-list control group.

Parent confidence in coping with disability and behavior problems

Kasari and colleagues (23,24) have examined parent competence in trials aimed at testing a caregiver-mediated intervention for toddlers with autism (Joint Attention Symbolic Play Engagement and Regulation) by inquiring about parent's level of a) confidence in carrying out intervention strategies, and b) comfort in carrying out intervention strategies. They found that neither question regarding parent competence predicted engagement level, type of play, or joint attention from a coded caregiver-child play interaction, and that self-rated competence was generally high across the board following 24 caregiver-mediated sessions (23).

A non-standardized questionnaire titled the Parent Perception Questionnaire (12) asked parents to rate their confidence, coping skills, knowledge, understanding, family issues, and planning. Roberts and colleagues reported that the center-based plus parent training intervention group showed the greatest gains in perception of confidence, as compared to home-based parent training intervention and waitlist groups, which led them to hypothesize that perhaps the small group environment in the center-based setting was more effective in improving parent understanding of how to manage behaviors associated with ASD. In this setting, parents were able to share knowledge with each other and with professionals within the center. The authors also acknowledged that there are aspects of the center-based program which aim to improve parent knowledge, while the educational component in the home-based program is less structured.

Family functioning

The McMaster Family Assessment Device (67) is a self-administered scale measuring general family function. In their randomized controlled trial examining the role of parent mental health in an education and skills training program (Preschoolers with Autism; ten 90-minute small group sessions, and 10 60-minute individual family sessions) for parents of young children with ASD, Tonge and colleagues (30) reported that the effect of treatment was

dependent upon level of family functioning at baseline.

The Beach Family Quality of Life Questionnaire (68) has been used to assess perceived quality of life across a number of domains, including family interaction, parenting, emotional wellbeing, physical wellbeing, and disability support. Roberts and colleagues (12) revealed that after controlling for baseline differences, the center-based intervention group (which involved parent training and support) showed the most gains in quality of life overall, followed by the waitlist group and finally by the home-based (parent training) intervention group.

Parent mental health

The General Health Questionnaire (69) was the primary outcome measure for Tonge and colleagues (30), who examined how participation in a parent education and skills training program for parents of children with ASD (Preschoolers with Autism; ten 90-minute small group sessions, and 10 60-minute individual family sessions) affected parental mental health and adjustment. The General Health Questionnaire has four subscales including somatic symptoms, social dysfunction, anxiety and insomnia, and depression. In Tonge and colleagues' study, both the treatment (parent education and behavior management intervention) and the active control (parent education and counseling) resulted in significant improvement in overall parental mental health at 6-month follow-up, with the parent education and behavior management intervention effectively reducing a greater percentage of anxiety, insomnia, and somatic symptoms and family dysfunction relative to the active control condition.

Parent skill level/fidelity

Many studies testing parent mediated early interventions for children with ASD have included a measure of parent fidelity, or parent adherence to the components of the intervention. This could be used both to indicate how well the program conveyed the skills to the parent, as well as help to account for the child's subsequent improvement. Although specific interventions would employ an individualized measure of parent fidelity, many interventions with similar principles share common characteristics in their assessment of parent fidelity.

Rogers and colleagues (19) demonstrated that they were able to improve parent fidelity through an enhanced version of P-ESDM (with additions consisting of motivational

interviewing, multimodal learning tools, and a weekly 90-minute home visit) relative to the basic model of P-ESDM. Further, they indicated that parent fidelity was positively associated with child's improved skills. For those receiving P-ESDM via telehealth (12 weekly 1.5-hour videoconference sessions with online access to P-ESDM materials *vs.* monthly 1.5-hour videoconference sessions with online access to alternative resources), parent fidelity was significantly better than in the control group (18). Casenhiser and colleagues (15) described a parent fidelity scale which consisted of behaviors such as joining, use of affect, and support for reciprocity in their comparison of a DIR-based intervention focused on socialization and parent-child interaction (2 hours of therapy and coaching per week for 12 months) and a community treatment group. Post-treatment, the intervention group in this study showed significantly more improvement in fidelity ratings relative to the community treatment group for nearly all items. Similarly, Nefdt and colleagues (32) showed significantly better fidelity of implementation of pivotal response treatment procedures for those in the self-directed learning program compared to the wait-list control group.

Parent-child outcomes

Outcomes capturing the interaction between the parent and child include codes generated from semi-structured parent-child interaction tasks, some of which are shared or joint attention, child initiations, and parent synchrony. Semi-structured parent-child interaction tasks afford the opportunity to observe parent and child behaviors in a play setting, which has the potential to be more naturalistic than a standardized testing environment. Shared or joint attention is characterized by instances where the parent and child experience mutual attentional focus. Child initiations involve the frequency with which the child initiates an interaction with the parent. Parent synchrony involves a parent contributing comments, statements, or other types of social interaction which support the child's responses.

Green and colleagues (27) noted clear treatment effects (18 sessions over 6 months of parent-mediated communication-focused treatment in children with autism; Preschool Autism Communication Trial) for measures of parent-child interaction, including parent's synchronous response, child initiations, and parent-child shared attention. Additionally, Aldred and colleagues (13) showed significantly greater improvement in parental synchronous communication and child communicative acts in the active

treatment group (educating parents and training them in how to adapt communication based on child skill level; Child's Talk Project), although no group difference was observed in outcome level of shared attention.

A primary focus of Siller and colleagues' (20) study was to evaluate the effect of a parent-mediated intervention (Focused Playtime Intervention, a parent education program involving one weekly 90-minute session for 12 weeks) on responsive parental communication, also known as synchronization [i.e., whether the parent's communication was synchronized with the child's actions, which has been shown to predict language development, e.g., (70)]. Results revealed that parents who were assigned to the Focused Playtime Intervention condition demonstrated more gains in responsive communication than those in the control group. This study also identified parent characteristics that appeared to be predictive of a greater improvement in responsive communication (i.e., how differences in learning styles contribute to acquiring this particular skill), and reported that parents classified as "insightful" (into the child's behavior and the parent-child relationship) were more likely to benefit from improvements in responsive communication. However, no effect was identified for the association between short-term gains in parental responsiveness and long-term child language development.

Kasari and colleagues (23) similarly demonstrated that those in the immediate treatment (caregiver-mediated joint engagement intervention for toddlers with ASD; 24 sessions of Joint Attention Symbolic Play Engagement and Regulation) showed increased joint engagement at post-treatment relative to those in the waitlist group. Further, children in the immediate treatment group showed significantly better responsiveness to joint attention bids than did those in the waitlist group. Pajareya and colleagues (16) showed that children in the home-based DIR/Floortime™ intervention group demonstrated significantly greater gains on the Functional Emotional Assessment Scale (71), which is a DIR-specific rating scale that can be applied to interactions between parents and children. This measure was also used in a study comparing the effectiveness of the PLAY Project Home Consultation model of monthly caregiver coaching for a year to usual community services, which showed that children in the treatment group showed significant improvements in social-emotional development (28).

From parent-child play sessions, Vismara and colleagues (18) coded a child's social communication utterances, including spontaneous functional verbal utterances and spontaneous joint attention behaviors; however, they did not identify

a significant effect when comparing 12 weekly 1.5-hour videoconference sessions with online access to P-ESDM materials to monthly 1.5-hour videoconference sessions with online access to alternative resources. Similarly, in testing Hanen's 'More Than Words' intervention (eight weekly group sessions and three individual sessions, over three months, aimed at teaching parents skills) Carter and colleagues (21) failed to identify a significant effect for change in parental responsivity from a parent-child free play task, although effect sizes were reported to range from medium to large.

Other studies have coded videotaped probes of parents interacting with their child in typical play interactions to identify the frequency with which the parent provided language opportunities, and how many functional verbal utterances were produced by the child (32). Consistent with their hypotheses, parents in the self-directed learning program in pivotal response treatment showed increased use of language opportunities relative to the community treatment group, and children in the intervention group demonstrated significantly more functional utterances following treatment. In another study, parent-child dyads assigned to the Social ABCs intervention (12 weeks of parent training, and 12 weeks of implementation) showed significantly greater improvement in vocal responsiveness to parent prompting, child vocal initiations, and parent smiling (33).

Overall, a number of parent-child measures reflect a treatment effect, particularly in the areas of parent synchrony, child initiations, and shared attention. These effects are noted across studies and models of intervention, including the Preschool Autism Communication Trial, Child's Talk Project, DIR/Floortime™, Joint Attention Symbolic Play Engagement and Regulation, and others. This shows that parent-mediated intervention models have been particularly well equipped to demonstrate progress in the setting of a parent interacting with his or her child.

Discussion

Overall, studies investigating the efficacy of parent-mediated early interventions for ASD employ varied approaches to examining treatment outcomes. Most studies use a number of measures to assess treatment response, including measures focused on the child, parent or family, and/or parent-child interaction. When summarizing results across studies, findings were generally mixed, as there was no one measure that consistently showed a treatment effect.

Some measures or categories of measurement, however, appeared to perform better than others. As noted earlier, there is a wide range in duration/intensity and delivery modality of interventions, which could also contribute to the mixed results.

Of the child outcomes, measures of social communication were relatively more likely to show a treatment effect based on the studies reviewed, though not consistently. Adaptive behavior skills showed a treatment effect in a handful of studies. Finally, parent-child interaction variables generally proved valuable in measuring response to treatment, including parent synchrony, child initiations, and shared attention. In fact, of all the sub-categories of potential indicators of treatment response, those that fell under the category of parent-child interaction variables appeared to perform best, in terms of the number of studies that reported significant results. Since a core feature of many of the parent-mediated interventions is parent-child interaction, it would follow that skills gained through intervention would transfer well to a play session. Some of the skills resulting from parent-mediated intervention may include a child's ability to initiate more communication and follow joint attention, both of which could be measured through parent-child interaction tasks. Further, as a parent shows increased responsivity to the child's communication, or demonstrates greater synchrony by commenting on the child's actions, it serves to reinforce and promote the child's engagement. Once a child has developed more functional social and communication skills through parent-mediated intervention, it may become more natural for a parent to demonstrate responsive communication, engage the child, and share interest in the child's actions.

The importance of culture cannot be understated when designing research studies and seeking to identify appropriate measures of treatment response. Cultural considerations necessitate the selection of measures that are relevant to the population of interest, or that are adapted as appropriate to better meet the needs of the population. Interpretability of results is at significant risk when tools developed for a specific culture or set of experiences are applied to disparate settings. As more tools are translated and culturally adapted, rich opportunities for cross-cultural research efforts expand.

One challenge of evaluating this body of research is that a wide range of measures have been used to assess outcomes. Additionally, some included measures were not originally intended to detect change. Thus, this makes it difficult to combine results across studies and to generate meaningful

conclusions.

Another challenge is the subjectivity associated with some of the common measures evaluating treatment response, and/or the fact that some raters (e.g., parents) are not able to stay blind to treatment condition. In the future, it will be important to continue to develop tools that can objectively measure treatment response, including biomarkers that are sensitive to change (72). Promising tools might include heart rate variability, eye tracking, and electroencephalography, although some of these measures may be hindered by their limited transportability to naturalistic settings (72,73).

Finally, when evaluating treatment response in parent-mediated autism early interventions, it may be important to consider ecological validity. As many of these interventions are meant to be used in naturalistic settings, measures that are specific to one setting or one person's interaction with a child may be limited in ecological validity. Siller and colleagues sought to address this by including "samples of parent-child interaction... videotaped on three separate days, both in the research lab and the families' home, using two parallel sets of toys" (20). If more studies employed this approach, treatment effects could be more easily and realistically evaluated, allowing for a greater emphasis on generalization of skills, real world impact of intervention, and assessing functioning across settings in which the child interacts with his or her environment.

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Footnote

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