LANGUAGE IMPROVEMENT FOLLOWING PIVOTAL RESPONSE TREATMENT FOR CHILDREN WITH DEVELOPMENTAL DISORDERS

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for Children with Developmental Disorders

Abstract

Given the high prevalence of communication deficits in developmental disorders, there is need for effective and efficient early interventions. The aim of this pilot study is to examine benefits of Pivotal Response Treatment (PRT) for improving language in young children with developmental disorders without autism spectrum disorder. Parents of fifteen children with developmental disorders received weekly PRT parent training for 12 weeks. Standardized parentrated assessments were administered at baseline and post-treatment to measure changes in language. Structured laboratory observation indicated children demonstrated significantly greater frequency of utterances and improvement on standardized questionnaires measuring expressive language and adaptive communication skills following PRT. Findings suggest that PRT may be efficacious in improving language abilities among children with developmental disorders.

Keywords: Developmental Disorders, Pivotal Response Treatment, language, communication

Children with developmental disorders (DD) are a growing population in the United States with national prevalence rates estimated as high as 5.81%, including both children with Autism Spectrum Disorder (ASD) and other developmental disabilities (Zablotsky et al., 2015). While there are several ways to define DD, the literature broadly defines DD as a group of conditions that originate in childhood and involve prominent delays in one or more domains of development including language, motor, cognition, social communication, and adaptive functioning (McDonald et al., 2006; Shevell, 2008). The etiologies of some DD may be known, such as the chromosomal abnormality in Down syndrome or genetic mutations in 22q11 deletion syndrome, while the etiologies of some DD are yet to be determined, as in the case of idiopathic intellectual disability or global developmental delay (GDD; Shevell, 2008). Importantly, many children with DD exhibit prominent deficits in language ability, as well as cognitive delays, when compared to same-age peers (Moeschler & Shevell, 2014).

Some studies report improvements in language, motor, and cognitive outcomes among children with DD following behavioral interventions (Brosnan & Healhy, 2011; McIntyre, 2008) and early therapies targeting language deficits in children with DD remain an important ongoing area of research (Matson, Mahan, & LoVullo, 2009; Reed, Hirst, & Hyman, 2012). The strong evidence base for early intervention in children with Autism Spectrum Disorder (ASD; Reed & Hirst, 2012; Warren et al., 2011; Reichow, 2012) supports the idea that behavioral treatment can substantially improve functional language for young children. In particular, treatment studies in children with ASD suggest that early interventions are associated with the best long-term outcomes (Estes et al., 2015). One class of early interventions, Naturalistic Developmental Behavioral Interventions (NDBI; Schreibman et al., 2015), usually recommended for children with ASD, combines behavioral principles of learning with naturalistic and developmental

teaching strategies to target a variety of skills including language, social communication, and joint attention (Schreibman et al., 2015). Several randomized controlled trials of NDBI report improvements in language, social communication, play, joint attention, and emotion regulation among children with ASD (Dawson et al., 2010; Kasari et al., 2006; Yoder & Stone, 2006; Wetherby et al., 2014; Hardan et al., 2015; Gengoux et al., 2015). Given that children with ASD and children with DD without ASD share similar difficulties with language and social communication, it is possible that NDBI may also be effective for children with DD without ASD for improving language and other developmental domains. Despite the growing prominence of NDBI in improving outcomes for children with ASD, the potential effectiveness of these interventions in children with DD without ASD (i.e., global developmental delay with or without known genetic etiology) has not received enough scientific attention.

Pivotal Response Treatment (PRT) is an established NDBI that targets language and social communication skills and has a strong evidence base for use with children with ASD (Bradshaw, Koegel, & Koegel, 2017; Gengoux et al., 2019; Hardan et al., 2015; Wong et al., 2015). Across many studies, children with ASD who received PRT sessions exhibited substantial improvements in language and social communication (Gillett & LeBlanc, 2007; Koegel, Camarata, Koegel. Ben-Tall, & Smith, 1998; Koegel, Carter, & Koegel, 2003; Koegel, O'Dell, & Koegel, 1987; Hardan et al., 2015), and these treatment benefits appear to persist over time (Gengoux et al., 2015). A critical component of PRT is parent training in which parents receive explicit instruction in PRT strategies as well as coaching and feedback on PRT implementation through regular meetings with a trained PRT therapist (Hardan et al., 2015). Research evidence suggests that parents can become proficient in PRT strategies and achieve high fidelity of implementation ratings (Minjarez et al., 2011; Hardan et al., 2015). Parents can become skilled

in implementing PRT strategies in the home and community on a daily basis, and this generalization of training has been associated with improved outcomes among children with ASD (Coolican, Smith, & Bryson, 2010; Koegel, Symon, & Koegel, 2002; Hardan et al., 2015; Minjarez et al., 2011; Nefdt, Koegel, Singer, & Gerber, 2010; Gengoux et al., 2015). Although the effects of PRT and parent training on child outcomes have been well-studied in children with ASD, little research has been conducted on expanding the use of PRT to other DD. Given that deficits in language skills are common among children with DD without ASD, it is likely that these children would also benefit from participation in PRT. However, to our knowledge, no investigations have been published to date examining the efficacy of PRT as delivered by parents for children with DD without ASD.

The present study is a 12-week pilot trial of a PRT parent training program targeting language deficits in young children with DD without ASD. In the present study, DD are defined as prominent delays in major domains of development (including cognition, motor, and language skills) of both known and unknown etiologies. The primary goal of the study was to evaluate the feasibility of implementing a 12-week PRT intervention with parents of children with DD without ASD to target language impairments. Secondary goals of the study included assessing whether children exhibit significant improvement in language skills on video recorded samples of child language, standardized assessments, and parent-report questionnaires following 12 weeks of PRT.

Method

Participants

Fifteen children with DD without ASD participated in this study at a university clinic. Recruitment occurred between September 2012 and September 2017 through distribution of

fliers, referrals by local professionals, and word of mouth. Following informed consent, a comprehensive intake evaluation was conducted to determine study eligibility. The intake evaluation included a review of medical and psychiatric histories and administration of standardized developmental and language assessments (i.e., Mullen Scales of Early Learning; MSEL; Preschool Language Scale, Fourth Edition; PLS-4) to establish eligibility.

Fifteen subjects (7 males, 8 females; mean age 40 months \pm 10.4; range: 24-60) with DD were included in the final sample. DD represented in this sample included the following: cerebral palsy (n=2; 13.3%), Cri du chat syndrome (n=2; 13.3%), Down syndrome (n=1; 6.67%), Fragile X (n=1; 6.67%), Klinefelter syndrome (n=1; 6.67%), and idiopathic global developmental delay (n=8; 53.39%). A small portion (33%; 5/15) of children in the sample were reported by parents to be in the Mild/Moderate range for social impairment on the SRS; however none of these participants met ADOS and ADI-R autism criteria or had a formal diagnosis of ASD. All participants were in school (100%), primarily in special day classes (53%) and attending an average of 15.1 hours of school per week (range = 0-27). None of the participants received inhome Applied Behavior Analysis (ABA) treatment, but all participants (100%) received weekly speech therapy with an average of 75 minutes of individual therapy per week. Additional data on child demographic and clinical baseline measures are included in Table 1.

INSERT TABLE 1

Inclusion and Exclusion Criteria

This study was approved by the University's institutional review board. Participants included children (a) 2-6 years old, (b) with either global developmental delay or a medical/genetic disorder associated with risk for intellectual disability, and (c) significant impairment on standardized tests of global development and language. Significant impairment in

development was defined as MSEL Early Learning Composite score \leq 70. Given the goal of evaluating PRT for children with limited functional communication, the PLS-4 criteria for significant language impairment was age-based and more stringent for older children (2- and 3year olds were \geq 1 SD below age level, 4-year olds \geq 2 SDs, and 5–6-year olds \geq 3 SDs). Children with a diagnosis of autism spectrum disorder (ASD) were excluded from the study and families with prior PRT treatment were excluded from this study. The Social Responsiveness Scale (SRS; Constantino, 2013) was obtained from all participants and if scores were above the clinical cutoff, (i.e., 70 total T-score) the Autism Diagnostic Interview-Revised (ADI-R; Le Couteur et al., 1989) and Autism Diagnostic Observation Schedule (ADOS; Lord et al., 2000) were administered to rule out ASD. No changes in inclusion or exclusion criteria were made over the duration of the study. Additionally, participants were required to have at least one caregiver available to consistently participate in treatment and to maintain all existing community treatments stable during the investigation (e.g., occupational therapy, speech therapy). See Table 1 for participant characteristics.

Procedures

After completion of screening procedures, eligible participants and their parents attended a baseline visit during which a structured laboratory observation (SLO) was collected. Parents also completed questionnaires related to child language, adaptive, and social skills. Following completion of baseline measures, participants and their parents immediately began the 12-week PRT parent training program. At the end of the trial, the same battery of assessments from baseline was repeated at post-treatment (week 12). All data collected were managed using REDCap electronic data tools (Harris et al., 2009) hosted at the University's Center for Clinical Informatics.

Intervention

The intervention consisted of 12 weeks of Pivotal Response Treatment (PRT) parent training. Sessions were one hour per week and focused on training parents in implementing PRT strategies to target their child's functional communication skills (i.e., use of utterances, words, or phrases for requesting or commenting). The parent training curriculum was based on the PRT manuals, Pivotal Response Treatment: Using Motivation as a Pivotal Response (Koegel, 2011) and Using Pivotal Response Treatment to Teach First Words to Children with Autism (Koegel, 2011), and utilized PRT training materials similar to those used in previous studies of PRT parent training (Hardan et al., 2015; Minjarez et al. 2011). Of the 12 sessions, three were parentonly sessions and nine were parent-child dyadic sessions. During the first parent-only session (week 1), therapists used didactic instruction and video examples to teach the six motivational PRT techniques including: (a) establishing the child's attention and providing a clear prompt, (b) following the child's lead and incorporating child choice of teaching materials, (c) interspersing maintenance tasks with acquisition tasks, (d) providing contingent reinforcement, (e) reinforcing the child's attempts, and (f) utilizing natural reinforcers (Koegel et al., 2001). During the parentchild sessions (weeks 2-8 and 11-12), the therapist demonstrated PRT procedures and provided in-vivo coaching while the parent practiced PRT with their child. For the other two parent-only sessions (week 9 and 10), parents reviewed video of their own PRT implementation in the clinic and during home practice sessions and received feedback from the therapist. In between training visits, parents were encouraged to implement PRT strategies with their child in the natural environment (home and community settings). Parent questions related to daily implementation in natural settings were discussed during the weekly parent training visit.

Therapist training

All therapists conducting PRT parent training sessions were trained in PRT by the senior author through didactic instruction, video review, observation of treatment sessions, and in-vivo coaching. The two primary therapists were master-level clinicians with at least one year of experience implementing PRT for children with ASD. All therapists demonstrated greater than 80% PRT fidelity of implementation based on video review of teaching interactions prior to beginning treatment sessions.

Measures

Feasibility Outcome Measures

Several indices were used to assess the feasibility of implementing a parent-training model of PRT to improve functional communication abilities of children with DD including recruitment, attrition, and success of outcome data collection.

Preliminary Efficacy Outcome Measures

The following primary outcome measures were obtained at baseline and post-treatment (week 12) to estimate the preliminary efficacy of PRT in improving functional communication for this population.

Functional Utterances in Structured Laboratory Observation (SLO). To assess the effects of PRT on functional communication skills of children with DD, behavioral data were collected on the frequency of functional utterances made by participants in a 10-minute videotaped structured laboratory observation (SLO; Hardan et al., 2015; Gengoux et al., 2019). Functional utterances were broadly defined as intentional utterances made by the child to the parent for the purposes of requesting or commenting. For each 10-minute SLO, parents were provided with a standard set of toys and instructed to play with their child and to elicit communication as they typically would at home (Hardan et al., 2015; Gengoux et al., 2019).

These videotaped parent-child interactions were coded by trained raters, blind to study timepoint, according to established procedures (Hardan et al., 2015; Gengoux et al., 2019). Specifically, frequency counts were generated for the total number of functional utterances made within 10 minutes, as well as for each unique type of utterance: (a) unintelligible, (b) imitative, (c) verbally prompted (d) nonverbally prompted, and (e) spontaneous. Unintelligble utterances were defined as difficult-to-understand utterances directed at the parent with a clear intent of requesting or commenting (e.g., child utters "gha" to request a nearby ball), while imitative utterances were defined as intelligible utterances made by the child to intentionally mimic the parent's language (e.g., child utters "ball" to mimic the word ball said aloud by the parent). Verbally prompted utterances were non-imitative responses to a parent's question or statement, in which the child generated a new word or phrase (e.g. child says "ball" in response to parent asking what child would like). Nonverbally prompted utterances were defined as child utterances that immediately followed a clear, nonverbal prompt from the parent (e.g., parent holds up a ball to wait for the child to make an utterance to request the ball), while spontaneous utterances were defined as a functional utterance in the absence of any prompt or effort from the parent to elicit a response (e.g., child utters "ball" spontaneously to request a nearby ball; Hardan et al., 2015). Utterance types are mutually exclusive. Raters were trained until they consistently reached target interobserver agreement (IOA; 80%) and intraclass correlation coefficients (ICC) also indicated excellent agreement for total utterance frequency [ICC(2,1) = 0.94] and acceptable agreement for utterance type (unintelligible = 0.83; imitative = 0.98; verbally prompted = 0.97; nonverbally prompted = 0.89; spontaneous = 0.74).

Preschool Language Scale-Fourth Edition (PLS-4). The PLS-4 (Zimmerman et al. 2002) is a standardized assessment used to measure expressive and receptive language.

Vineland Adaptive Behavior Scales, Second Edition (Vineland-II). The Vineland-II (Sparrow et al 2005), is a measure of adaptive functioning in natural contexts. The Vineland provides information about functioning in the following domains: Communication, Daily Living, Socialization and Motor Skills.

MacArthur-Bates Communicative Development Inventories (CDI). The CDI Words and Gestures and Words and Sentences versions (Fenson et al., 2007) are parent report instruments that provide information about a child's early communicative skills, including vocabulary comprehension, production, gestures, and grammar.

Social Responsiveness Scale (SRS). The Social Responsiveness Scale is a parent-report questionnaire that assesses the degree of a child's social impairments in natural social settings (SRS; Constantino and Gruber, 2005).

Statistical analyses

Preliminary Efficacy. To examine the hypothesis that PRT would increase the frequency of child functional utterances in the SLO, paired sample *t*-tests were conducted to compare mean total utterances, as well as specific utterance types, between the baseline and post-treatment (week 12) timepoints. Change between baseline and post-treatment on secondary outcome measures (PLS-4, Vineland-II Communication domain, CDIs, and SRS) was also analyzed using paired sample *t*-tests across timepoints. Lastly, mixed effects regression models were employed to analyze potential predictors of improvement from baseline for the primary outcome measure (SLO total utterances). Type 1 error rate of 0.05 was used for analyses of the primary and secondary outcome measures. Multiple comparison correction was not performed as the purpose of the secondary analyses was to better understand the specificity of the improvements from baseline in this small initial study in order to inform future study planning.

Results

Feasibility Outcomes

Fifty parents were screened after contacting the clinic to inquire further about the study and this high number indicates that this population can be recruited as participants for a parent training intervention. Following screening, 18 participants met inclusion criteria and signed consent, while 32 participants did not meet criteria for a variety of reasons including severe autism symptomology (SRS > 70) or a diagnosis of ASD (n = 24), and/or could not commit to participation requirements (n = 8). Two of the 18 participants enrolled in the trial terminated prematurely and 16 participants completed the 12-week trial; however, one participant who completed treatment did not complete post-treatment assessments. The resulting attrition rate (17%) is acceptable and within the range of other published studies of similar interventions (Bearss et al., 2018; Bearss et al., 2015). With regards to data collection for efficacy outcome measures, 100% (16/16) of standardized assessments (PLS-4), laboratory observations (SLO), and parent-report measures (Vineland-II, CDI, SRS) were completed at the baseline timepoint. Following the 12-week trial, the majority of participants (94%; 15/16) completed the standardized assessments, laboratory observations, and parent-report measures.

Preliminary Efficacy Outcomes

Table 2 summarizes the findings of this study including total utterances and distinct utterance types from the SLO, standardized assessments, and parent-reported outcomes.

Results show that the average frequency of child utterances increased from baseline to post-treatment as assessed during the SLO. Figure 1 displays the total number of functional utterances and different utterance types across time points. On average, children demonstrated significant improvement in total number of utterances with an average increase of 28 total

utterances between baseline and week 12. The improvement from baseline was most apparent for imitative utterances with an average increase of 15 imitative utterances over 12 weeks. Nonverbally prompted and spontaneous utterances showed a nonsignificant trend toward improvement. The improvement from baseline was not significant for unintelligible utterances.

A significant improvement from baseline was observed for the expressive raw scores on the PLS-4 with an average increase of four points over time. Although there were no significant changes for the expressive standard and total standard scores on the PLS-4, children increased on average by three standard score points on the expressive subscale and two standard score points on the total standard score following participation in PRT. Significant changes were also observed for the Vineland-II Communication domain with an average increase of five standard score points over 12 weeks. Additionally, a significant improvement from baseline was also observed in phrases and words understood as well as words produced and total gestures on the CDI Word & Gestures. Additionally, a significant improvement from baseline was observed in words produced and word forms on the CDI Words & Sentences; however, no significant changes were observed for word endings. There were also no significant changes for the SRS total T-score or the social communication or restricted and repetitive behavior subscales.

INSERT TABLE 2

Predictors of Response

There were no effects of gender or age on improvements with PRT treatment based on independent samples t-tests. However, regression analysis indicated that children with higher expressive t-scores on the Mullen and higher total utterances at baseline on the SLO had more improvement in total utterances at the post-treatment timepoint (F(2,13) = 14.02, p = 0.001). Baseline Mullen expressive t-scores were a significant predictor of improvement from baseline

for total and imitative utterances, accounting for 34% of the variance in improvement for total utterances. Improvement from baseline was not modified by other language indices including baseline PLS total language standard scores, CDI words out of 396 or 680, or CDI sentence length.

INSERT FIGURE 1

Discussion

In this pilot study, PRT parent training appeared to be a feasible intervention for parents of children with diverse DD without ASD. Although children with diverse developmental disabilities were included in this pilot trial, all children in this study were considered minimally verbal due to significant language impairment on the PLS-4. Preliminary efficacy results indicate improvements in language abilities in children with DD without ASD following participation in a 12-week trial of PRT. The results of this trial suggest that PRT may have promise for increasing language abilities in children with a wide range of DD. To our knowledge, no studies to date have assessed the effects of PRT in children with DD other than ASD. Furthermore, our observations provide additional support for the potential benefit of NDBI in targeting a wide range of deficits in individuals with DD including language abilities.

Information gleaned from study recruitment, attrition, and rate of successful outcome data collection support the feasibility of a parent-mediated trial of PRT for children with DD without ASD. A high number of parents expressed interest in this study and this number was similar to those reported by previous feasibility studies of interventions with children with ASD (Matheson, Drahota, & Boutelle, 2019), and higher than some studies of children with ASD (Bearss et al., 2018) or other DD (Fodstad, Kirsch, Faidley, & Bauer, 2018). Additionally, the attrition rate of the present study was similar to anticipated benchmarks reported by previous studies (Bearss et al., 2018), which also supports the feasibility of a parent-mediated trial of PRT for children with DD without ASD. Cumulatively, findings from this pilot trial suggest that PRT can be delivered to parents of children with DD without ASD with high rates of engagement and participation.

Results from preliminary efficacy measures revealed significant increases in the total number of utterances with parent implementation of PRT with their children. Results collected through the SLO indicate that children displayed a significant increase in total functional communicative utterances, particularly imitative utterances, following PRT intervention. The amount of change in utterances over time in the present study is consistent with previouslyreported improvements in functional communicative utterances in a related study with PRT with young children with ASD (Hardan et al., 2015). The observed increase in total utterances is likely driven by the significant increase in imitative utterances observed in this sample. The increases in imitative utterances may be attributed to the PRT technique for minimally verbal children of frequent modeling of words for the child to imitate as a maintenance task. Although some increase in unintelligible utterances may also be expected given that minimally verbal children often exhibit poor articulation as they learn to talk, the change in unintelligible utterances observed in this trial was not statistically significant. In addition, the fact that the greatest improvement was observed in the imitative utterance type is consistent with an expected developmental sequence, as young children may find it easier to imitate adult language models before they are capable of responding to other types of prompts or using un-prompted communication (Demchak, 1990; Koegel & Koegel, 2019; MacDuff, Krantz, McClannahan, 2001). Findings here also demonstrated that nonverbally prompted and spontaneous utterances showed a nonsignificant trend towards greater improvement. Future research will be needed to

determine whether these more independent utterance types could show significant improvement over a longer treatment duration. Additionally, it would be important to include fidelity measures of parent PRT implementation in future trials.

Improvements on other secondary outcome measures were also observed with PRT. Children in the present sample exhibited significant increases in raw scores on the expressive subscale of the PLS-4. This increase was observed to be a moderate effect size in this sample, and is particularly interesting as prior studies of early language interventions for children with ASD have not consistently demonstrated change on the PLS-4 (Hardan et al., 2015; Gengoux et al., 2019). This finding highlights the promising nature of PRT in improving language among children with DD; however, further investigations through randomized controlled trials are warranted. Consistent with previous research on PRT in children with ASD, results from this study indicated that children with DD receiving PRT displayed gains in expressive language as measured by the Vineland-II Communication domain (Baker-Ericzen, Stahmer, & Burns, 2007; Coolican, Smith, & Bryson, 2010; Gengoux et al., 2015; Koegel & Koegel, 2019). Changes in the standard and v-scale scores between baseline and week 12 are particularly meaningful given that these are norm-referenced scores and therefore positive numeric change indicates greater progress than the normative sample over a similar time period. Raw score improvement was also observed on the MacArthur-Bates Communicative Development Inventories, providing additional evidence of quantitative improvement in expressive vocabulary. Together, these data suggest preliminary efficacy for children with DD as has been established for PRT with children with ASD. Evidence that children with stronger baseline expressive language made greater progress is also congruent with previous research indicating that children who display some language and higher expressive communication at baseline will show greater treatment gains and outcomes (Lotter, 1974; Venter, Lord, & Schopler, 1992). Further research on predictors of response to intervention is warranted.

This study also provides support for the recommended use of parent training in treatment programs (Brookman-Frazee, Vismara, Drahota, Stahmer, & Openden, 2009; Koegel & Koegel, 2019; National Research Council, 2001). The present intervention consisted of a 12-week program of weekly parent training sessions. In spite of the relatively low number of treatment hours provided (12 hours total), children demonstrated significant gains in functional communication. These findings are consistent with the assertion that parent training can be both effective and an efficient strategy for boosting child communication (Hardan et al., 2015). Although parent fidelity of PRT implementation was not directly measured in this study, child improvements in language abilities across measures suggest that PRT parent training was effective in teaching parents how to elicit more functional communication from their child.

There are several limitations to this study that warrant a discussion. First, this pilot study was an uncontrolled trial. It will be important to conduct a randomized controlled trial of PRT with children with DD without ASD to ensure a systematic and controlled investigation into the language and social communication outcomes reported in this study. Second, this study included children with a variety of developmental disorders. As there may be differential effects of treatment depending on underlying etiology, subsequent studies of more homogeneous groups will be important. Third, this pilot trial had a limited sample size and data collection occurred over a five-year period due to budget constraints and therapist availability. It will be essential to include a larger number of participants in a future trial to replicate findings and better understand predictors of treatment response.

In light of the limited number of PRT investigations in individuals with DD, it is crucial to examine different service delivery models in this population. Much research has been conducted on different doses of treatment and treatment modality (e.g., individual treatment, in-home treatment, individual parent training, group parent training) of PRT for children with ASD, and determining the effectiveness of various forms of this intervention for children with DD would be valuable (Baker-Ericzen et al., 2007; Hardan et al., 2015; Schreibman & Koegel, 2005). Additionally, it would be helpful to investigate the effect of PRT with non-autism DD on other variables, such as nonverbal communicative behaviors, social interactions, and cognitive skills. Lastly, further research is warranted into long-term outcomes of children with DD without ASD who receive PRT.

In summary, this preliminary study suggests that PRT may be a feasible intervention for children with DD without ASD and a potentially effective treatment for increasing expressive communication. Specifically, a short-term parent training intervention may produce significant improvements in both total utterances and overall expressive communication skills. Further research including controlled studies will be necessary to build on these findings and to investigate the effectiveness of NDBIs like PRT for children with diverse developmental disabilities.

References

- Baker-Ericzén, M. J., Stahmer, A. C., & Burns, A. (2007). Child demographics associated with outcomes in a community-based pivotal response training program. *Journal of positive behavior interventions*, *9*(1), 52-60.
- Bradshaw, J., Koegel, L., Koegel, R. (2017). Improving Functional Language and Social Motivation with a Parent-Mediated Intervention for Toddlers with Autism Spectrum Disorders. *Journal of Autism and Developmental Disorders*, 47, 2443-2458.
- Brookman-Frazee, L & Koegel, R. L. (2004). Using parent/clinician partnerships in parent education programs from children with autism. *Journal of Positive Behavior Interventions*, 6(4), 195-213.
- Brookman-Frazee, L., Vismara, L., Drahota, A., Stahmer, A., & Openden, D. (2009). Parent training interventions for children with autism spectrum disorders. In *Applied behavior analysis for children with autism spectrum disorders* (pp. 237-257). Springer, New York, NY.
- Brosnan, J., & Healy, O. (2011). A review of behavioral interventions for the treatment of aggression in individuals with developmental disabilities. *Research in developmental disabilities*, *32*(2), 437-446.\

Constantino, J. N. (2013). Social responsiveness scale (pp. 2919-2929). Springer New York.

Coolican, J., Smith, I. M., & Bryson, S. E. (2010). Brief parent training in pivotal response treatment for preschoolers with autism. *Journal of Child Psychology and Psychiatry*, 51(12), 1321-1330.

- Dawson, G., Rogers, S., Munson, J., Smith, M., Winter, J., Greenson, J., et al. (2010).
 Randomized, controlled trial of an intervention for toddlers with autism: The Early Start
 Denver Model. Pediatrics, 125, 17–23.
- Demchak, M. (1990). Response prompting and fading methods: A review. *American Journal on Mental Retardation*, 94(6), 603-615.
- Estes, A., Munson, J., Rogers, S. J., Greenson, J., Winter, J., & Dawson, G. (2015). Long-term outcomes of early intervention in 6-year-old children with autism spectrum disorder.Journal of the American Academy of Child & Adolescent Psychiatry, 54(7), 580-587.
- Fenson, L., Marchman, V.A., Thal, D.J., Dale, P.S., Reznick, J.S., & Bates, E. (2007). *MacArthur-bates communicative development inventories*. Baltimore, MD: Paul H. Brookes Publishing Co.
- Gengoux, G. W., Berquist, K. L., Salzman, E., Schapp, S., Phillips, J. M., Frazier, T. W.,
 Minjarez, M.B. & Hardan, A. Y. (2015). Pivotal response treatment parent training for autism: Findings from a 3-month follow-up evaluation. *Journal of Autism and Developmental Disorders*, 45(9), 2889-2898.
- Gerenser, J., & Forman, B. (2007). Speech and language deficits in children with developmental disabilities. In J. W. Jacobson, J. A. Mulick & J. Rojahn (Eds.), Handbook of intellectual and developmental disabilities; handbook of intellectual and developmental disabilities (pp. 563-579, Chapter xx, 726 Pages) Springer Publishing Co, New York, NY.
- Gillett, J.N., & LeBlanc, L.A. (2007). Parent implemented natural language paradigm to increase language and play in children with autism. *Research in Autism Spectrum Disorders*, 1(3), 247-255.

- Hardan, A. Y., Fung, L. K., Libove, R. A., Obukhanych, T. V., Nair, S., Herzenberg, L. A., . . . Tirouvanziam, R. (2012). A randomized controlled pilot trial of oral N-acetylcysteine in children with autism. *Biological Psychiatry*, 71(11), 956-961.
- Hardan, A. Y., Gengoux, G. W., Berquist, K. L., Libove, R. A., Ardel, C. M., Phillips, J., Frazier, T., Minjarez, M. B. (2015). A randomized controlled trial of pivotal response treatment group for parents of children with autism. Journal of Child Psychology and Psychiatry, 56(8), 884-892.
- Hastings, R. P. (2002). Parental stress and behaviour problems of children with developmental disability. *Journal of Intellectual and Developmental Disability*, 27(3), 149-160.
- Kasari, C., Freeman, S., & Paparella, T. (2006). Joint attention and symbolic play in young children with autism: A randomized controlled intervention study. Journal of Child Psychology and Psychiatry, 47, 611–620
- Koegel, L. K. (2011). Pivotal Response Treatment: Using Motivation as a Pivotal Response: A training manual. Santa Barbara, CA: UCSB Koegel Autism Center.
- Koegel, L. K. (2011). Using Pivotal Response Treatment to Teach First Words to Children with Autism: A training manual. Santa Barbara, CA: UCSB Koegel Autism Center.
- Koegel, L.K., Carter, C.M., Koegel, R.L. (2003). Teaching children with autism self-initiations as a pivotal response. *Topics in language disorders*, *23*, 134-145.
- Koegel, R. L., Bimbela, A., & Schreibman, L. (1996). Collateral effects of parent training on family interactions. *Journal of autism and developmental disorders*, 26(3), 347-359.
- Koegel, R. L., Camarata, S., Koegel, L. K., Ben-Tall, A., & Smith, A. E. (1998). Increasing speech intelligibility in children with autism. *Journal of autism and developmental disorders*, 28(3), 241-251.

- Koegel, R. L., & Koegel, L. K. (2019). Pivotal response treatments for autism spectrum disorder. Paul H Brookes Publishing: Baltimore, MD.
- Koegel, R. L., O'dell, M. C., & Koegel, L. K. (1987). A natural language teaching paradigm for nonverbal autistic children. *Journal of autism and developmental disorders*, 17(2), 187-200.
- Koegel, R. L., Symon, J. B., & Kern Koegel, L. (2002). Parent education for families of children with autism living in geographically distant areas. *Journal of Positive Behavior Interventions*, 4(2), 88-103.
- Krakovich, T. M., McGrew, J. H., Yu, Y., & Ruble, L. A. (2016). Stress in parents of children with autism spectrum disorder: An exploration of demands and resources. *Journal of autism and developmental disorders*, 46(6), 2042-2053.
- Le Couteur, A., Rutter, M., Lord, C., Rios, P., Robertson, S., Holdgrafer, M., & McLennan, J. (1989). Autism diagnostic interview: a standardized investigator-based instrument. *Journal of Autism and Developmental Disorders*, *19*(3), 363-387.
- Lord, C., Risi, S., Lambrecht, L., Cook, E. H., Leventhal, B. L., DiLavore, P. C., ... & Rutter, M. (2000). The Autism Diagnostic Observation Schedule—Generic: A standard measure of social and communication deficits associated with the spectrum of autism. *Journal of Autism and Developmental Disorders*, 30(3), 205-223.
- Lotter, V. (1974). Factors related to outcome in autistic children. *Journal of Autism & Childhood Schizophrenia*, 4(3), 263-277.
- MacDuff, G. S., Krantz, P. J., & McClannahan, L. E. (2001). Prompts and prompt-fading strategies for people with autism. Making a difference: Behavioral intervention for autism, 37-50

- Majnemer, A. (1998, March). Benefits of early intervention for children with developmental disabilities. In *Seminars in Pediatric Neurology* (Vol. 5, No. 1, pp. 62-69). Elsevier.
- Matson, J. L., Mahan, S., & LoVullo, S. V. (2009). Parent training: A review of methods for children with developmental disabilities. *Research in Developmental Disabilities*, 30(5), 961-968.
- McCracken, J. T., McGough, J., Shah, B., Cronin, P., Hong, D., Aman, M. G., . . . Hollway, J. (2002). Risperidone in children with autism and serious behavioral problems. *New England Journal of Medicine*, 347(5), 314-321.
- McDonald, L., Rennie, A., Tolmie, J., Galloway, P., & McWilliam, R. (2006). Investigation of global developmental delay. *Archives of Disease in Childhood*, *91*(8), 701-705.
- McIntyre, L. L. (2008). Parent training for young children with developmental disabilities: Randomized controlled trial. *American Journal on Mental Retardation*, *113*(5), 356-368.
- Moeschler, J. B., & Shevell, M. (2014). Comprehensive evaluation of the child with intellectual disability or global developmental delays. *Pediatrics*, peds-2014.
- Mohammadzaheri, F., Koegel, L. K., Rezaee, M., & Rafiee, S. M. (2014). A randomized clinical trial comparison between pivotal response treatment (PRT) and structured applied behavior analysis (ABA) intervention for children with autism. *Journal of autism and developmental disorders*, *44*(11), 2769-77.
- National Research Council. (2001). *Education children with autism*. Washington, DC: National Academy Press.
- Nefdt, N., Koegel, R., Singer, G., & Gerber, M. (2010). The use of a self-directed learning program to provide introductory training in pivotal response treatment to parents of children with autism. *Journal of Positive Behavior Interventions*, *12*(1), 23-32.

- Reed, F. D. D., Hirst, J. M., & Hyman, S. R. (2012). Assessment and treatment of stereotypic behavior in children with autism and other developmental disabilities: A thirty year review. *Research in Autism Spectrum Disorders*, 6(1), 422-430.
- Reichow, B. (2012). Overview of meta-analyses on early intensive behavioral intervention for young children with autism spectrum disorders. *Journal of Autism and Developmental Disorders*, 42(4), 512-520.
- Rogers, S. J. (1996). Brief report: Early intervention in autism. *Journal of autism and developmental disorders*, 26(2), 243-246.
- Schreibman, L., Dawson, G., Stahmer, A. C., Landa, R., Rogers, S. J., McGee, G. G., ...
 Halladay, A. (2015). Naturalistic developmental behavioral interventions: Empirically validated treatments for autism spectrum disorder. *Journal of Autism and Developmental Disorders*, 45(8), 2411–2428.
- Schreibman, L., & Koegel, R. L. (2005). Training for Parents of Children with Autism: Pivotal Responses, Generalization, and Individualization of Interventions. In E.D. Hibbs & P.S. Jensen (Eds.), *Psychosocial treatments for children and adolescent disorders: Empirically based strategies for clinical practice* (2nd ed., pp.605-631). Washington, DC: American Psychological Association.
- Shevell, M. (2008). Global developmental delay and mental retardation or intellectual disability: conceptualization, evaluation, and etiology. *Pediatric Clinics of North America*, 55(5), 1071-1084.
- Sparrow, S.S., Cicchetti, D.V., & Balla, D.A. (2005). *Vineland adaptive behavior scales* (2nd edition). San Antonio, TX: Pearson.

- Venter, A., Lord, C., & Schopler, E. (1992). A follow-up study of high-functioning autistic children. *Child Psychology & Psychiatry & Allied Disciplines*, 33(3), 489-507.
- Warren, Z., McPheeters, M. L., Sathe, N., Foss-Feig, J. H., Glasser, A., & Veenstra-VanderWeele, J. (2011). A systematic review of early intensive intervention for autism spectrum disorders. *Pediatrics*, 127(5), 14-21.
- Wetherby, A. M., Guthrie, W., Woods, J., Schatschneider, C., Holland, R. D., Morgan, L., & Lord, C. (2014). Parent implemented social intervention for toddlers with autism: An RCT. *Pediatrics*, 134, 1084–1093.
- Wong, C., Odom, S. L., Hume, K. A., Cox, A. W., Fettig, A., Kucharczyk, S.,...& Schultz, T. R. (2015). Evidence-based practices for children, youth, and young adults with autism spectrum disorder: A comprehensive review. *Journal of Autism and Developmental Disorders*, 45(7), 1951-1966.
- Yoder, P., & Stone, W. (2006). A randomized comparison of the effect of two prelinguistic communication interventions on the acquisition of spoken communication in preschoolers with ASD. Journal of Speech, Language, & Hearing Research, 49, 698–711.
- Zablotsky, B., Black, L. I., Maenner, M. J., Schieve, L. A., & Blumberg, S. J. (2015). Estimated prevalence of autism and other developmental disabilities following questionnaire changes in the 2014 National Health Interview Survey. Center for Disease Control, 2015 Annual Report.



Figure 1. Amount and type of child utterances on the structured laboratory observation (SLO) at the baseline and week 12 timepoints.

Table 1

Participant Characteristics

Child Characteristics (N=15)	Mean (SD)
Mean age (months)	40 (10.0) : Range: 24-60
Male / Female	7 / 8
Mullen Early Learning Composite	51.0 (6.0) : Range: 49-70
Ethnicity	4 Caucasian / 5 Asian / 6 Biracial
Education	
School type	8 Special Education / 2 Regular Education / 5 Private
School hours/week	15.1 (7.6)
Concomitant treatment minutes/week	
ABA	0 (0)
Speech Therapy	75.0 (35.9)
Other therapeutic interventions	93.1 (48.5)

Table 2

Performance on Primary and Secondary Outcome Measures Between Baseline and Week 12

	Mean (SD)		Main effect		Effect Size
	Baseline	Week 12	t	р	d
SLO					
Total utterances	57.4 (41.33)	85.1 (33.8)	-3.40	0.005*	0.77
Unintelligible utterances	44.9 (31.8)	54.5 (21.9)	-1.13	0.277	0.35
Imitative utterances	8.1 (9.9)	22.6 (18.5)	-3.55	0.004*	0.94
Verbally prompted utterances	4.0 (6.4)	5.6 (6.6)	-1.19	0.267	0.25
Non-verbally prompted utterances	0.07 (0.27)	1.57 (2.8)	-2.09	0.057	0.71
Spontaneous utterances	0.28 (0.61)	0.71 (0.99)	-2.10	0.054	0.52
Vineland					
Communication Domain standard score	71.1 (10.4)	75.9 (12.3)	-3.01	0.010*	0.45
Expressive V-scale score	8.00 (1.5)	8.79 (2.2)	-3.30	0.006*	0.37
Expressive raw score	26.7 (8.5)	32.4 (10.2)	-3.93	0.002*	0.50
PLS-4					
Expressive raw score	24.5 (4.1)	28.3 (4.8)	-4.45	<0.001*	0.85
Expressive standard score	60.8 (8.2)	63.8 (10.6)	-1.32	0.206	0.32
Total standard score	57.7 (9.2)	59.3 (10.3)	-0.97	0.346	0.16
CDI – Words & Gestures					
Phrases understood out of 280	21.8 (5.0)	24.2 (3.3)	-2.70	0.017*	0.57
Words understood out of 396	221.1 (101.7)	262.5	-2.74	0.016*	0.43
		(100.5)			
Words produced out of 396	70.9 (39.6)	105.7 (90.7)	-3.59	0.003*	0.40
Total gestures out of 63	41.9 (14.2)	45.8 (13.1)	-2.44	0.029*	0.24
CDI – Words & Sentences					
Words produced out of 680	91.9 (126.2)	142.0	-3.08	0.009*	0.41
		(152.7)			
Word forms out of 25	0.64 (1.2)	1.14 (1.3)	-2.19	0.047*	0.46
Word endings out of 45	0.07 (0.27)	0.14 (0.4)	-1.00	0.336	0.21
SRS					
Total T-scores	61.1 (8.9)	60.1 (8.7)	0.84	0.42	0.11
Social communication T-scores	61.1 (9.3)	60.4 (8.6)	0.64	0.53	0.08
RRB T-scores	59.7 (10.9)	57.8 (10.3)	1.06	0.31	0.18