The Effects of Parent Training in Pivotal Response Treatment (PRT) and Continued Support through Telemedicine on Gains in Communication in Children with Autism Spectrum Disorder

A thesis submitted to the University of Arizona College of Medicine-Phoenix
in partial fulfillment of the requirements for the Degree of Doctor of Medicine

Namrata Singh
Class of 2014

Mentor: Raun Melmed, MD

Acknowledgements

I would like to express my deepest appreciation to my mentor, Dr. Raun Melmed, for all of his support and guidance over the last four years. Without his mentorship this scholarly project would not have been possible.

I would also like to sincerely thank Dr. Daniel Openden, Dr. Christopher Smith, and Alexis Boglio for their contributions to my project. With their support, I have learned so much about how to conduct research in the field of autism. I very much appreciate the time that all three of these individuals took to help me design and implement my project, answer my questions throughout the course of my project, and the time they set aside to go over my data analysis with me.

Finally, I would like to thank the Southwest Autism Resource and Research Center in Phoenix and the patients and families who attend SARRC for their permission to conduct this research project. I hope that this project will be helpful in guiding further studies using telemedicine to aid in the therapy of children with autism spectrum disorder.

Abstract

Introduction: With the increase in children diagnosed with autism spectrum disorders (ASD) each year comes the need for individualized interventions necessary for children with autism. Many of the treatments currently available are time consuming, costly, and rely heavily on behavioral interventionists despite the lack of qualified interventionists to provide these services. Therefore, there is a growing recognition of the need for efficient, cost-effective treatment models that involves families in the intervention. This study examined the effects of continuous parent training and feedback for 3 months in Pivotal Response Treatment (PRT) through telemedicine on responsivity to language opportunities in children with ASD versus a control group of parents receiving one week of PRT training alone. We hypothesized that with telemedicine support, the child's verbal communication would increase.

Methods: 30 child-parent dyads from Tucson, AZ were enrolled in this randomized control trial. Subjects were 24-60 months of age and met DSM-IV criteria for autism at the time of enrollment. All subjects received one week of intensive parent training at the Southwest Autism Research and Resource Center (SARRC) in PRT. The support group received telemedicine feedback three times weekly for three months. Data was analyzed using two sample t-tests and Wilcoxon rank sum tests.

Results: The control group had a mean responsivity in function verbal utterances of 64.3% and the telemedicine group had a 62.7% verbal responsivity rate prior to initiation of telemedicine support. At three month follow-up, the control group had a mean responsivity rate of 58.6% and the telemedicine group had a mean responsivity of 64.3%. A two sample t-test showed a no significant difference between the two groups with a p-value of 0.51.

Conclusions: This study did not find any significant difference between the telemedicine and control groups. However, there is a trend towards increased verbal communication in the telemedicine group. We therefore recommend further studies to determine the utility of telemedicine and parent training in PRT in the treatment of children with ASD.

Table of Contents

List of Figures and Tables

Introduction/Significance

The number of children diagnosed with autism is increasing steadily. Estimates of prevalence rates for autism spectrum disorders (ASD) vary among different studies, with the most recent estimates of prevalence being 1 in 88 children worldwide.[1] With the increase in children diagnosed with autism spectrum disorders (ASD) comes the need for individualized interventions necessary for children and families of children with autism. Many of the treatments currently available are time consuming, costly, and rely heavily on behavioral interventionists instead of parents as interventionists.[2] There are not enough qualified interventionists to provide services to many of the families with autism. This problem will only be exacerbated in the future as more children are diagnosed earlier with autism as more sensitive detection methods are developed. Therefore, there is a growing need for efficient, cost-effective treatment models that includes families in the intervention process.

One way to address this increasing need is to train parents to implement interventions through natural language interactions with their child. Training parents as primary intervention agents requires less time from highly trained staff, while increasing the intensity and frequency of the treatment. Evidence demonstrates that outcomes for children with autism are substantially improved with intensive behavioral intervention early on.[2] Pivotal Response Treatment (PRT) is an evidence based model that uses both a developmental approach and the science of Applied Behavioral Analysis (ABA) procedures to target areas that underlie the core symptoms of autism, including impairments in social interaction, communication, and repetitive/restrictive behaviors.[3] Research has shown that children with autism learned question asking skills, conversational skills, and social skills when taught using PRT.[4] Research also supports the use of PRT in parent education programs, as it is typically delivered through a parent training model and inherently occurs in natural settings.[4]

There are four phases of verbal communication: preverbal communication, first words, word combinations, and sentences.[5] In the preverbal communication phase, children communicate

through vocal babble and gesture. In typically developing children, this occurs during the first 6-12 months of life.[6] In the first words phase (ages 12-18 months in typically developing children), children use non-imitated spontaneous single words referentially and symbolically to communicate about objects and events, including those outside the immediate context.[7,8] The third phase, word combinations, occurs in typically developing children ages 18 -30 months when children have a vocabulary that is rapidly increasing in size and includes a variety of parts of speech (nouns, verbs, descriptors). Children are able to make two to three word combinations.[6] Children in phase four, combine words into clausal structures or sentences. They also use some morphological markers such as plurals, prepositions, and some verb endings. Their vocabulary is sufficiently large to serve their communicative needs in everyday situations. They communicate a wide range of functions in different settings with both familiar and unfamiliar people.

Failure to develop useful speech is one of the more common consequences of neurodevelopmental disorders, one of the most debilitating being autism. Classically, it was estimated that up to 50% of individuals with autism would never develop useful speech, though the current estimate seems to be lower.[9] If a child, with autism or any other condition, has not developed useful speech by age 5 years, it is unlikely that he or she will do so.[10] Studies of parent training in PRT have been shown to increase the functional verbal utterances in children with ASD [1,3] and Coolican, Smith & Bryson also looked at the nature of the verbal utterances. None of these studies looked at the effects of continuous parent training over an extended period of time.

The field of telemedicine is relatively new and is just starting to be used in the diagnosis and treatment of autism. While more research has been conducted in the use of telemedicine for diagnosis of behavioral disorders such as autism, less research has looked at using telemedicine as a therapeutic tool in pediatric patients with autism. One study showed that telemedicine was comparable to or even superior to traditional therapy sessions.[11] This study looked at various aspects of communication including both gestures and verbal communication. However, the

sample size of this study was only two patients and parents were not the main conductors of the therapy. Currently, there are no known large scale studies that utilize telemedicine in the delivery of PRT using the parent as the primary therapist.

This randomized control trial examined the effects of continued parent training and support in PRT via telemedicine over a three month period on verbal language responsivity after 1 week of intensive parental training versus only one week of parental training in PRT. We hypothesized that there would be a significantly greater increase in responsivity in the children of parents who received continued supportive training in PRT through telemedicine.

Research Materials and Methods

In order to comply with federal regulations, this prospective randomized controlled trial received IRB approval from an outside review board as part of a larger investigation at SARRC. To ensure compliance through the University of Arizona for this particular investigation, the project was subjected to internal review by Dr. Joan Shapiro and an F309 form was completed for the University of Arizona.

Thirty parent-child dyads who reside in Tucson, Arizona participated in this study. Selection criteria for parents included: a) being the primary caregiver for a child with ASD between the ages of 24 and 60 months and b) no previous training in or experience with implementing PRT. Children were diagnosed with ASD within the six months prior to enrollment in the study and DSM-IV criteria for autism were confirmed at the time of enrollment, based on information from the Autism Diagnostic Interview-Revised (ADI-R)[12] and the Autism Diagnostic Observational Scale-General (ADOS-G).[13] To maintain comparability of experimental groups, an equal number of individuals from each diagnostic category of ASD (autism, Asperger's Syndrome, and PDD-NOS) were matched and randomly assigned to either the support group or control group. The parent trainer(s) remained blind to the initial assessment results. Parent trainer(s) included B.A. level or higher clinical interventionists with more than 3 years of experience implementing applied behavior analysis (ABA) based interventions and PRT with families of children with ASD.

The program consisted of two phases. The first phase was consistent with previous studies measuring the efficacy of a 25 hour training program designed to teach parents to implement the basic motivational procedures of PRT with fidelity.[14,15] As in the studies described above, the initial one-week intensive parent training was implemented at SARRC for five consecutive days at 5 hours per day. Motivational procedures included interspersing maintenance tasks and task variation, shared control and child choice, and using direct reinforcers. On the first day of intervention, parents were provided with *How to Teach Pivotal Behaviors to Children with Autism: Training Manual.*[16] Parent trainers reviewed the manual and modeled the procedures

for approximately 60 minutes. During the remaining 4 hours and throughout the remaining 4 days, parents practiced implementing PRT with their child while the parent trainer provided in-vivo, continual coaching and feedback. Sessions were provided in small clinic playrooms that included a table and chairs and a variety of toys. Additional sessions took place in nearby community settings including restaurants, parks, shopping malls, and other natural environments.

The second phase of the study was conducted in the training group only. To extend the findings of the previous studies,[14, 15] this project utilized the Arizona Telemedicine Program[17] to provide enhanced parent training and continued support. Following completion of the initial 1-week intensive parent training program at SARRC, parent child dyads returned home and "met" three times weekly with the parent trainer via telemedicine for 12 weeks. For each session, parents were asked to bring a 15 minute video of the child in his natural environment while he was receiving PRT by the parent. Similar to the initial parent training program at SARRC, parent trainers were provided continuous coaching and feedback on parent implemented PRT during each of the telemedicine intervention sessions.

Diagnostic, cognitive, and adaptive behavior assessments were conducted at enrollment with four measures: ADI-R[12], ADOS-G[13], Stanford-Binet Intelligence Scale v. 5[18], and the Vineland Adaptive Behavior Scales[19]. Please see Table 1 for a description of the various assessments. Verbal responsivity was examined at baseline, post in-vivo parent training program, and follow-up at 3 months. Data was collected through video submission, de-identified and coded by an un-blinded researcher. The first 10 minutes of each videotaped probe was coded and the child's responsivity to verbal cues was documented using a 15 second interval recording system. An interval was scored as appropriate if the child produced one or more functional verbal utterances. A functional verbal utterance was operationally defined as a verbalization that included the following: the child's use of at least normal vocal loudness, body and facial orientation towards the parent and/or relevant stimulus materials, and vocalization that appeared functional or related to the task. Functional verbal utterances included requests,

refusals, comments, responses, initiations, and questions. The type of prompting was also recorded. Different prompts recorded were model prompts (child repeated word modeled by the therapist/parent), indirect prompts (choice, fill in the blank, response to question) or child initiated. Overall responsivity was calculated as the percentage of times the child responded appropriately, either via model prompt, indirect prompt, or child initiation. Following collection of data, results were analyzed using a two sample t-test with unequal variances and a Wilcoxon rank sum test. Significance of results was determined by a p-value of less than 0.05.

Table 1. Enrollment Assessment Protocols

Protocol	Description
Autism Diagnostic Interview-Revised (ADI-R)	Semi-structured, investigator based informant interview; used both as a key assessment for diagnosis and to evaluate the breadth of symptoms associated with each of the core autism domains
Autism Diagnostic Observation Scale-General (ADOS-G)	Uses standard presses and observation; used in conjunction with the ADI-R to provide corroborating information for the diagnosis of autism
Stanford-Binet Intelligence Scale v.5	Incorporates the use of two routing subtests which increases the precision of measurement by tailoring the difficulty of the items to level of cognitive functioning
Vineland Adaptive Behavior Scale (VABS)	Semi-structured interview that evaluates four areas of adaptive behavior: daily living, communication, motor skills, and socialization; can be used to assess almost all levels of functioning in children from normal to severely impaired

Results

A total of 17 parent-child dyads were enrolled in the telemedicine support group and 13 parent-child dyads were enrolled in the control group based on the inclusion criteria. At baseline, the control group and the telemedicine group had a mean responsivity of 29.2% and 28.4%, respectively. The control group had a mean responsivity of 64.3% at the completion of the 5 day in-house parental training in PRT and the telemedicine support experimental group had a 62.7% verbal responsivity rate prior to initiation of telemedicine support. Responsivity was determined by the presence of a functional verbal utterance including requests, refusals, comments, modeling, initiations and questions. In order for a communication to be recorded as a response, an utterance also needed to include normal vocal loudness, body and facial orientation towards the parents and/or relevant stimulus materials, and vocalization that appeared functional or related to the task. At three month follow-up, the control group had a mean responsivity rate of 58.6% and the telemedicine group had a mean responsivity of 64.3%. (Figure 1, Table 2).

To analyze the data, a two sample t-test with unequal variances was conducted at baseline, post-in vivo parent training and at three month follow-up. At three month follow-up, a two sample t-test showed no significant difference between control and telemedicine groups (p-value= 0.51). Two sample t-tests were also conducted using the difference in responsivity between post-5 day training and baseline, three month follow up and baseline, and between three month follow-up and post-5 day training. These tests also showed no significant differences with p-values of 0.93, 0.49, and 0.28, respectively. Given the large variability within groups as evidenced by large standard deviations, Wilcoxon rank sum tests were run to analyze differences in responsivity between post-5 day training and baseline, three month follow up and baseline, and between three month follow-up and post-5 day training in the control and treatment groups. Similar to results obtained by the two sample t-tests, there was no significant difference between the two groups with p-values of 0.95, 0.43, and 0.22, respectively.

Figure 1. Mean Verbal Responsivity Prior to and After Telemedicine Support

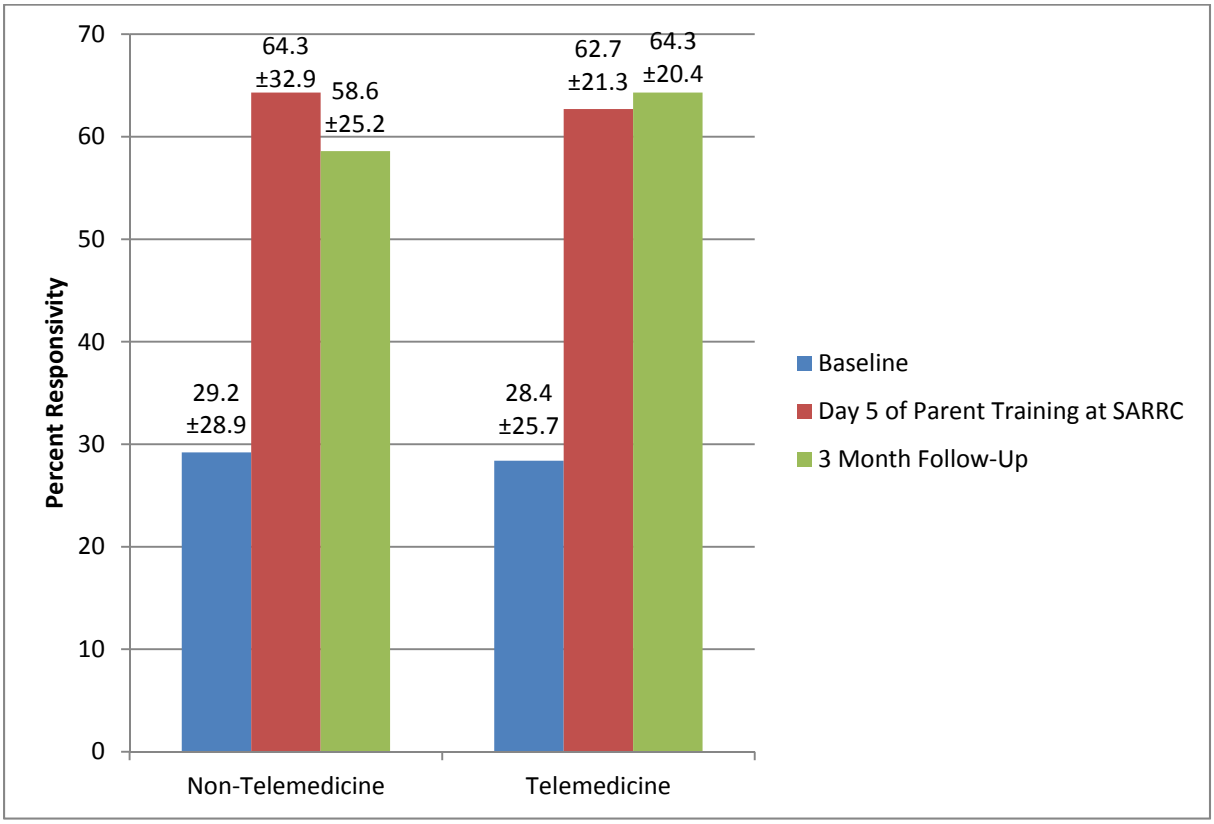

Figure 1. Figure 1 shows the percent responsivity in verbal communication at baseline, day 5 of intensive parental training just prior to the experimental portion of the study, and at the completion of three months of telemedicine feedback. Responsivity in the support group remains high and increases slightly, while the control group has a slight decrease in responsivity at 3 month follow up, which was not, however, statistically significant.

Table 2. Comparison of Responsivity Between Control and Treatment Groups

Group	*Responsivity*					
	Baseline	**Post-5 Day Training**	**3 month Follow-Up**	**5 Day Training-Baseline**	**3 Month F/U - Baseline**	**3 Month F/U - Day 5**
Control (N=13)	29.2±28.9[a]	64.3±32.9	58.6±25.2	35.1±30.6	29.5±25.5	-5.6±15.4
Treatment (N=17)	28.4±25.7	62.7±21.3	64.3±20.4	34.3±23.9	35.9±24.2	1.6±21.1
p-value[b]	0.94	0.88	0.51	0.93	0.49	0.28
p-value[c]	--	--	--	0.95	0.43	0.22

[a] Standard deviation

[b] Derived from two-sample t test with unequal variances

[c] Derived from Wilcoxon rank sum test

Discussion

With the increase in numbers of children being diagnosed with autism and the lack of resources to deliver therapy to them, the use of telemedicine has important therapeutic implications. This randomized control trial aimed to examine the effects of continued parent training and support in PRT via telemedicine over a three month period on verbal language responsivity after 1 week of intensive parental training versus only one week of parental training in PRT. We hypothesized that our support group would have a significant increase in verbal communication and responsivity to verbal cues compared to our control group. Our results do show a slight increase in responsivity in the telemedicine support group compared to the control group in which there was a slight decrease in responsivity at three month follow-up. However, no significant differences were apparent between the control and telemedicine support groups related to increases in verbal communication.

In evaluating this study, we recognize certain limitations. Although participants were well randomized, neither the families nor the researchers were blinded as to which group participants were in. This could have led to researcher bias. Similarly, participants could have tailored their compliance with therapy according to their group assignation. These biases could lend themselves to falsely elevated increases in communication in the experimental group. Our study did aim to minimize bias, however, by randomization and matching participants. Secondly, we recognize that the sample size in this study is small, which lends to a low power. The small sample size may increase the likelihood of a type II error. This study had large variability within groups as evidenced by the large standard deviation within both the control and treatment group at baseline, post in-vivo training and at three month follow-up. This is likely because we used children with disorders along the entire spectrum of autism spectrum disorder, from mild autism to severe. If we had focused on one subset of children within autism spectrum disorder, there would likely have been less variability within groups making the results more significant. Finally, this study did not examine the degree of adherence to the correct implementation of pivotal response therapy training that parents received. The degree of adherence to correctly implementing pivotal response therapy should be examined to

determine if this has an impact on the results that we obtained as incorrect implementation could negatively impact the study results.

Future Directions

Though this study does not show a significant difference in the acquisition and maintenance of verbal communication between the telemedicine and control groups, there are limitations to this study as described above. Therefore, we would recommend conducting a similar study with a larger sample size. This is warranted given the trend in increased communication observed in the treatment group. Telemedicine should also be explored as a medium for initial parental training in PRT, especially for those without access to a behavioral therapist. With further research using continued parent training along with telemedicine as a means for training and therapy, there is the potential for many more children with ASD to receive the interventions that they need for improved communication and overall functional outcomes. Given the increasing numbers of children being diagnosed with ASD and the lack of sufficient trained therapists, this research is crucial.

Conclusions

This study is consistent with prior studies suggesting that parental training in pivotal response treatment is beneficial to increased communication in children with ASD. This study does not however show a significant difference in verbal communication with the addition of continued parental training and support through telemedicine, despite showing a trend toward increased communication in patients whose families received continued support. Further research in this area is recommended in order to explore the use of telemedicine as an adjunct to the treatment of children with ASD in the home environment.

References

1. Morbidity and Mortality Weekly Report. Prevalence of Autism Spectrum Disorders-Autism and Developmental Disability. Center for Disease Control. 2012.
2. National Research Council. Educating Children With Autism. Washington DC: National Academy Press. 2001.
3. Minjarez, M.B., Williams, S.E., Mercier, E.M., & Hardan, A.Y. Pivotal Response Group Treatment Program for Parents of Children with Autism. *Journal of Autism and Developmental Disorder* 2011; 41:92–101.
4. Coolican, J., Smith, I.M., Bryson, S.E. Brief parent training in pivotal response treatment for preschoolers with autism. *Journal of Child Psychology and Psychiatry* 2010; 51(12): 1321–1330.
5. Tager-Flushberg et al. Defining Spoken Language Benchmarks and Selecting Measures of Expressive Language Development for Young Children With Autism Spectrum Disorders. *Journal of Speech, Language, and Hearing Research* 2010; 52: 643-652.
6. Koegel, R.L., Openden, D., Fredeen, R.M., & Koegel, L.K. The basics of pivotal response treatment. In R. Koegel L., & L.K. Koegel (Eds.), [Pivotal Response Treatments for Autism Communication, Social, and Academic Development] (pp.25) Baltimore, MD: Brookes. 2001.
7. Oller,K. The emergence of the speech capacity. Mahwah, NJ: Erlbaum. 2000.
8. Stoel-Gammon, C. Sounds and words in early language acquisition: The relations between lexical and phonological development. In R. Paul (Ed.), Exploring the speech-language connection (pp. 25–52). Baltimore: Brookes. 1998.
9. Pickett, E., Pullara, O., O'Grady, J., & Gordan, B. Speech Acquisition in Older Nonverbal Individuals With Autism. *Cognitive Behavioral Neurology* 2009; 22:1.
10. Ornitz, E.M. Childhood Autism: A review of the clinical and experimental literature. *California Medicine* 1973; 118: 21-47.
11. Baharav, E & Reiser C. Using Telepractice in Parent Training in Early Autism. *Telemedicine and e-Health* 2010; 16(6): 727-731.

12. Lord, C., Rutter, M, & Couteur, A. Autism Diagnostic Interview-Revised: a revised version of a diagnostic interview for caregivers of individuals with possible pervasive developmental disorders. *Journal of Autism and Developmental Disorders* 1994; 24 (5): 659-685.
13. Lord, C, Risi, S, Lambrecht, L, et al. 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* 2000; 30:205–223.
14. Koegel, R.L., Symon, J.B., & Koegel, L.K. Parent education for families of children with autism living in geographically distant areas. *Journal of Positive Behavior Interventions* 2002; 4(2), 88-103.
15. Symon, J.B. Expanding interventions for children with autism: Parents as trainers. *Journal of Positive Behavior Interventions* 2005; 7(3): 159-173.
16. Koegel, R.L., et al. How to teach pivotal behaviors to children with autism: A training manual. Santa Barbara, CA: University of California, Santa Barbara, Dept. of Speech and Hearing Sciences. 1988.
17. Distance Education. *Arizona Telemedicine Program*. www.telemedicine.arizona.edu. Date accessed: June 2010.
18. Dombrowski, S., DiStefano, C., & Noonan, K. Review of the Stanford-Binet intelligence scales: Fifth edition. *Journal of Psychoeducational Assessment* 2006; 24: 123-136.
19. Sparrow, S., Balla, D., & Cicchetti, D. Vineland Adaptive Behavior Scales: Second edition. Circle Pines, MN: American Guidance Service, Inc. 2005.