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Reduced Restricted and Repetitive Behaviors after Pivotal Response Treatment

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Abstract

Children with ASD show high frequency of restricted and repetitive behaviors (RRBs); however, higher-order RRBs, such as restricted interests, have remained largely resistant to treatment. This study evaluated change in severity of RRBs following a 16-week open trial of Pivotal Response Treatment (PRT). Participants included 15 children with ASD ages 4 to 7 years. RRBs, as measured by the Repetitive Behavioral Scales- Revised (RBS-R) and Aberrant Behaviors Checklist (ABC), decreased significantly after treatment. These reductions remained significant after controlling for change in social communication skills. PRT shows promise in reducing RRBs; although PRT explicitly addresses pivotal social communication skills, there is a secondary and less direct effect on RRBs.

Keywords

Restricted and repetitive behaviors; Pivotal Response Treatment (PRT); behavior therapy; Repetitive Behavior Scale-Revised (RBS-R)

Restricted repetitive behaviors (RRBs) are core symptoms of Autism Spectrum Disorder (ASD), and these behaviors interfere with the daily functioning of the individual as well as the family (Gabriels, Cuccaro, Hill, Ivers, & Goldson, 2005; Harrop et al., 2014; Lounds, Seltzer, Greenberg, & Shattuck, 2007; Nadig, Lee, Singh, Bosshart, & Ozonoff, 2010; Richler, Bishop, Kleinke, & Lord, 2007; Shattuck et al., 2007). RRBs are a diverse set of behaviors that are often divided into two factors: repetitive sensory motor (RSMs) behaviors and insistence on sameness (IS) behaviors. RSM behaviors include lower-level behaviors, such as repetitive mannerisms and sensory seeking behaviors, while IS behaviors include higher-order behaviors, such as compulsions, rituals, difficulty shifting, and circumscribed

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Compliance with Ethical Standards

This work was approved by Yale University Institutional Review Board (IRB). All participants provided informed consent for participation.

The authors declare that they have no conflict of interest.

interests (Cuccaro et al., 2003; Honey, McConachie, Turner, & Rodgers, 2012; Szatmari et al., 2006). RRBs are not unique to individuals with ASD and are observed across individuals with other developmental disorders and intellectual disabilities; however, children with ASD have higher frequency of RRB compared to children with non-spectrum developmental disorders and intellectual disabilities (Bodfish, Symons, Parker, & Lewis, 2000; Hattori et al., 2006; Kim & Lord, 2012; Richler et al., 2007; Werner & Dawson, 2005).

Severity and type of RRB correlate with age and functioning level in individuals with ASD. For example, stereotyped movements are more common in younger individuals with ASD, and ritualistic behavior, compulsions, and restricted interests often emerge or increase in severity later in development (Bishop, Richler, & Lord, 2006; Esbensen, Seltzer, Lam, & Bodfish, 2009; Lam & Aman, 2007; Militerni, Bravaccio, Falco, Fico, & Palermo, 2002). Similarly, repetitive sensory motor behaviors are negatively correlated with nonverbal IQ (NVIQ), while circumscribed interests are positively correlated with NVIQ (Bishop et al., 2006; Cuccaro et al., 2003; Goldman et al., 2009; Militerni et al., 2002; Mooney, Gray, Tonge, Sweeney, & Taffe, 2009; Richler, Huerta, Bishop, & Lord, 2010; Schultz & Berkson, 1995; Szatmari et al., 2006). Additionally, individuals with lower levels of language skills exhibit higher frequency of RRBs overall (Ray-Subramanian & Ellis Weismer, 2012). RRBs are also associated with higher levels of anxiety in children with ASD (Sukhodolsky et al., 2008).

RRBs have been directly targeted as a primary outcome within pharmacological trials (Carrasco, Volkmar, & Bloch, 2012; Soorya, Kiarashi, & Hollander, 2008; King et al., 2009), but with inconsistent results and notable adverse effects. Focused behaviorally-based intervention strategies based on principles of Applied Behavioral Analysis (ABA) have shown reductions in some lower-order sensory-oriented and repetitive behaviors (Kuhn, Hardesty, & Sweeney, 2009; Rapp & Vollmer, 2005; see Boyd, McDonough, & Bodfish, 2012; Harrop, Gulsrud, Shih, Hovsepyan, & Kasari, 2015 for comprehensive reviews). The majority of these studies, however, have included small sample sizes, limiting the generalizability of the results. All studies also followed principles of ABA with highly intensive, individualized intervention.

Although there have been positive results in reduction of some RRBs, namely lower order repetitive sensory motor behaviors, following targeted behavioral treatment, there is significant need for treatments to also target higher-order behaviors, particularly insistence on sameness behaviors, such as rituals, difficulty with flexibility, preoccupations, and circumscribed interests. Additionally and more specifically, there is great need for behavioral treatments, such as Pivotal Response Treatment (PRT), that are appropriate across the range of social functioning and intellectual levels found in individuals with ASD (Boyd et al., 2012; Harrop et al., 2015).

PRT is a particularly appropriate approach for investigating RRB reduction, since in PRT, it is common practice to functionally incorporate RRBs into the treatment, as for example, using circumscribed interests as natural reinforcement to motivate the child (e.g., Koegel & Koegel, 2012). Most studies using PRT have focused on improvement in social communication or cognitive ability. For example, several recent RCTs of PRT have shown

significant improvement in language and social functioning (Hardan et al., 2014; Mohammadzaheri, Koegel, Rezaee, & Rafiee, 2014; Mohammadzaheri, Koegel, Rezaei, & Bakhshi, 2015). To date, though, no studies have investigated the effects of PRT on RRB reduction. Therefore, the purpose of the current study was to explore the differential effects of PRT with a focus on reduction in RRB following a 16-week trial of PRT.

Methods

Participants

Participants included fifteen children (5 female, 10 male), aged 4 to 7 years diagnosed with an autism spectrum disorder (ASD) using "gold-standard" diagnostic procedures. All children entered the study with a prior diagnosis of ASD, and the diagnosis was confirmed by a highly experienced licensed clinical child psychologist. Diagnostic impressions were informed by the Autism Diagnostic Observation Schedule (ADOS; Lord et al., 1999) and the Autism Diagnostic Interview-Revised (ADI-R; Rutter, LeCouteur, & Lord, 2003). Clinicians administering these measures were research reliable. Refer to Table 1 for characterization information.

Fourteen of the fifteen children participating in the study were enrolled in public school with both typically developing children and children with special needs. One child attended a special education school. All of the children received speech and language therapy and social skills instruction in school, and six of the fifteen received occupational therapy. None of the children were taking psychotropic medications. The families were asked not to add any behavioral treatments during the course of the study participation. No changes in educational placement or major changes in educational services were reported by the parents while their children were in the study.

Design

The current study is an open study of PRT that is part of a larger project on PRT aimed at investigation of feasibility, effects on social function, and brain mechanisms (Ventola et al., 2014; Ventola et al., 2015). This paper describes the differential effects of PRT with a focus on the change in RRBs before and after PRT. Baseline measures were taken immediately prior to treatment (t1), and post treatment measures were taken at the conclusion of 16 weeks of treatment (t2).

Treatment Approach

PRT is a naturalistic, behaviorally-based treatment approach based on principles of Applied Behavior Analysis (ABA). It involves specific components (child choice, child attending, clear opportunity, contingent reinforcement, natural reinforcement, reinforcement of attempts, and interspersed maintenance/acquisition tasks) designed to increase the child's social motivation. Additionally, the approach is highly naturalistic, so for the context of the current study, the sessions were play-based and used materials such as craft supplies, balls, blocks, and 'play-doh.'

For each child, treatment lasted 16 weeks, with a total of seven hours of treatment per week. Sessions were held in the clinic as well as in the child's home. Five hours per week were direct intervention with the child, and two hours per week consisted of parent guidance. The treatment targeted pivotal areas, including social initiation and responsivity, with the premise that improvements in these areas should lead to more widespread and generalized improvements in multiple areas of development and represent core changes in social motivation.

RRBs were targeted in this trial but within the context of a primary focus on pivotal social communication skill development. For instance, reciprocity was directly targeted as a pivotal area of PRT, but a focus on reciprocity also addressed restricted interests and flexibility. As an example, when taking turns within a conversation, the child needed to flexibly shift between topics. As another example, treatment focused on social initiations as a pivotal area, but within this context, stereotyped speech was addressed. If a child used stereotyped language (e.g., always used the exact same language to request), the child was prompted to use more flexible speech and make the request in different ways with different vocabulary and structure to the language. Therefore, PRT did address RRBs, particularly higher order RRBs, but within the context of social communication skill development.

Additionally, of importance, clinicians were able to utilize topics and activities related to circumscribed interests as a means of motivating the child. For example, clinicians utilized materials, such as games and craft activities, related to a child's circumscribed interest to increase their motivation and engagement in the interaction. Using this approach allowed clinicians to make the child's strong interests more functional, as the child was able to practice reciprocal engagement while using content related to their strong interest. As another example, access to materials related to a child's circumscribed interest was used as natural reinforcement for appropriate social communication; access to a highly desired material related to a circumscribed interest would be withheld until the child appropriately requested the material. A more detailed description of PRT can be found in the original instruction manual (Koegel, Schreibman, Good, Cerniglia, Murphy, & Koegel, L.K., 1989) and in an updated guide (Koegel & Koegel, 2012).

Fidelity of Implementation

The lead clinician was trained in PRT by faculty from the University of California Santa Barbara (UCSB), the research institution where PRT was developed. The lead clinician sent two separate videotaped sessions (of different children) to the trainer to ensure maintenance of treatment fidelity. Both videos met the standard fidelity criteria. To ensure that the bachelors-level clinicians were correctly implementing PRT during their sessions, they met with the lead clinician (licensed clinical psychologist) for two hours per week. During these meetings, clinicians discussed the children's progress, current presentation, and specific activities for the treatment sessions that would be motivating and foster skill development. Additionally, the lead clinician observed sessions live and via videotape at least once weekly for each participant.

Formal fidelity of implementation was assessed for two randomly coded treatment sessions for each subject. Two randomly selected five-minute segments per session were used for this

fidelity assessment. The standard fidelity assessment published by the developers of the approach was used, and per convention, fidelity was defined as demonstrating the treatment components (child choice, child attending, clear opportunity, contingent reinforcement, natural reinforcement, reinforcement of attempts, and interspersal of maintenance/acquisition tasks) in 80% of opportunities (Koegel & Koegel, 2012). The scoring was dichotomous; if the therapist demonstrated the component, a checkmark was used, and if not, a minus was used. All therapists maintained the defined treatment fidelity across the duration of the study.

Subject Characterization Assessment

Differential Ability Scales-II (DAS-II), Early Years Battery (Elliott, 2007)—The DAS-II is a standardized assessment of cognitive abilities that measures verbal, nonverbal and spatial reasoning abilities. The DAS-II was used in the current study for subject characterization at t1.

Autism Diagnostic Interview-Revised (ADI-R; Rutter, 2003)—The ADI-R is a comprehensive diagnostic parent-report interview that focuses on language/communication, reciprocal social interactions and restricted, repetitive and stereotyped behaviors and interests. The ADI-R was administered to the mothers of each subject at t1 for characterization.

Autism Diagnostic Observation Schedule (ADOS; Lord, 1999)—The ADOS is a semi-structured diagnostic assessment that allows clinicians to observe and assess social, communication and repetitive behaviors associated with ASD. The ADOS was performed on each subject at t1 by expert clinicians with no other involvement in the current study. The ADOS was used for subject characterization.

Clinical Evaluation of Language Fundamentals-Preschool-2 (CELF-P-2: Wiig, 2004); Clinical Evaluation of Language Fundamentals-Fourth Edition (CELF-4; Wiig, 2006)—The CELF-P-2 is a standardized language assessment for children 3 to 6 years, and the CELF-4 is a standardized language assessment for children 5 to 21 years. These measures evaluate receptive and expressive syntactic and semantic skills. They are useful for providing specific information regarding amounts and types of information the child understands and is capable of using. The CELF-P-2 or CELF-4 was administered to each subject at t1 for characterization. Test selection was based on the child's age. Children ages 4–6 completed the CELF-P-2 (n= 13), and children aged 7 completed the CELF-4 (n= 2).

Outcome Measures

Repetitive Behavior Scale-Revised (RBS-R; Bodfish et al., 2000)—The RBS-R is a 43-item parent report questionnaire assessing a range of restricted and repetitive behaviors in ASD. The scale includes six subscales: Stereotyped Behavior, Self-injurious Behavior, Compulsive Behavior, Ritualistic Behavior, Sameness Behavior, and Restricted Behavior. Items are rated from 0–3, and parents are instructed to report on frequency and interference of specific behaviors. The measure yields subscale scores and a total score.

Mean scores from prior studies range widely (e.g., M 26.4, SD 17.0, Kanne & Mazurek, 2011; M 8.88, SD 5.22 for high NVIQ sample, Gabriels et al., 2005; 24.02 for low RRB group and 62.24 for high RRB group, Gabriels et al., 2008). It is a commonly used and well-accepted measure to assess RRB (Honey et al., 2012). The mothers of participating children completed the RBS-R at t1 and t2 as an outcome measure.

Aberrant Behavior Checklist- Community Version (ABC; Aman, 1985)—The ABC is a 58-item parent report scale assessing maladaptive behaviors. It includes 5 empirically-derived subscales: Irritability, Lethargy/Social Withdrawal, Stereotypic Behavior, Hyperactivity, and Inappropriate Speech. Items are scored from 0–3, and parents are instructed to consider the frequency of the behavior and the degree to which is interferes with adaptive functioning. The scale yields subscale scores. The mothers of participating children completed the ABC at t1 and t2 as an outcome measure.

Social Responsiveness Scale-Second Edition (SRS-2; Constantino, 2012)—The Social Responsiveness Scale-Second Edition is a 65-item parent report scale designed specifically for use in ASD to quantitatively measure an individual's ability to engage in reciprocal social behavior in a naturalistic social setting. Each item on the scale inquires about an observed aspect of reciprocal social behavior that is rated on a scale from 0 (never true) to 3 (almost always true). Behaviors are assessed over five domains: Social Awareness, Social Cognition, Social Communication, Social Motivation, and Autistic Mannerisms. The total score generated serves as an index of severity of social deficits. The mothers of participating children completed the SRS-2 at t1 and t2 as an outcome measure.

Results

RRBs were measured using two parent-report measures, RBS-R and the Stereotypy subscale of the ABC. Parents of all 15 children completed the ABC and parents of 13 children completed the RBS-R due to changes in study protocol (RBS-R was added to the protocol following enrollment of the first two children). RRBs were assessed before (t1) and after (t2) a 16-week trial of PRT. Social functioning was assessed using the SRS-2. To control for possible effects of initial severity (e.g., regression toward the mean), change scores were modeled using residualized change scores, that is, the post-treatment severity scores minus the predicted post-treatment severity scores as predicted by the pretreatment severity scores. Reduction of RRB was defined as a statistically significant decrease in reported RRB symptoms as assessed by the RBS-R and ABC Stereotypy Domain. The measures assess RRBs by considering frequency, intensity, and amount of interference in daily functioning, as described in detail in the Methods section.

On average, the participants showed significant reduction in RRB symptom severity from pretreatment ($M = 26.67$, $S.D. = 19.69$) to post-treatment ($M = 15.58$, $S.D. = 12.25$), $t(11) = 3.53$, $p = .005$, Cohen's $d = 0.69$. Table 2 shows mean and standard deviation values for the measures of RRB symptoms before and after treatment. There was also significant reduction in the six subdomains on the RBS-R: Stereotyped Behavior (e.g. repetitive movements and sensory seeking behavior) ($p < .01$), Self-injury (e.g. hitting self) ($p < .05$), Compulsive Behavior subdomain (e.g., ordering objects, counting in a certain way) ($p < .05$), Ritualistic

Behavior (e.g., insisting on a certain order of events or certain repetitive topics or responses within conversations) ($p < .05$), Sameness (e.g. difficulty with transitions and changes in routines) ($p < .02$) and Restricted Behaviors (e.g., preoccupation with one subject matter or object) ($p < .02$). On the ABC, there was a significant reduction in the Stereotypy subdomain ($p < 0.001$) (e.g., repetitive movements).

We also tested whether reduction in RRB symptoms is associated with improvement in the social domain. Consistent with our previous reports (Ventola et al., 2014), the participants showed significant reduction in autism symptom severity indexed by the SRS-2 Total Raw score from pretreatment ($M = 86.75$, $S.D. = 22.25$) to post-treatment ($M = 73.50$, $S.D. = 17.83$), $t(11) = 3.25$, $p = .008$, Cohen's $d = 0.66$. (SRS-2 Total Raw score was utilized in the analyses as it provided a greater range of scores as well as finer gradients than T-Score equivalents.) The residualized change scores of RRB symptom severity, as assessed by the RBS-R, and those of autism symptom severity, as assessed by the SRS-2, were uncorrelated, $r = .12$, $p = .71$. We also conducted repeated-measures ANCOVA with the pre- to post-treatment change in the SRS-2 Total Raw score as a covariate to test whether change in RRBs was independent from the change in social communication symptoms. When covarying out the effects of the residualized change of the autistic symptom severity, repeated-measures ANCOVA revealed that the participants still showed significant reduction in RRB symptom severity from pre-treatment to post-treatment, $F(1, 11) = 12.06$, $p = .006$, partial $\eta^2 = .55$.

Discussion

The purpose of the current pilot study was to assess the differential effects of PRT, with a particular focus on reduction of restricted and repetitive behaviors (RRB) following a 16-week trial of Pivotal Response Treatment (PRT). Overall, children exhibited a significant reduction of a variety of RRBs, regardless of severity, and this improvement was independent of change in social communication skills.

RRBs are a diverse group of behaviors, and there was significant reduction in symptoms within all of the RRB domains assessed: stereotyped behavior, self-injury, compulsive behavior, ritualistic behavior, insistence on sameness, and restricted behavior. These results are particularly notable given that improvements were found in both lower order behaviors as well as higher order behaviors. As discussed, most behavioral treatments focus on only lower-order sensory behaviors, and there is great need for treatment approaches to focus on higher order behaviors, such as the ones reported here (e.g., ritualistic behavior, insistence on sameness, compulsive behavior). Additionally, reduction in RRBs was independent of improvement in social communication skills, as assessed by the SRS-2, indicating that PRT has separable effects on the overall autism severity and more domain-specific RRB symptom severity. Reduction of RRB was not solely a function of improved social communication. Also, of importance, the independence of these results indicates that the reduction of RRB was likely not a result of parental expectancy bias across parent report measures.

Interestingly, the PRT trial primarily and explicitly focused on improving social communication skills thought to be pivotal areas of development, including initiation,

responsiveness, and reciprocity. RRBs were addressed as well, yet less explicitly and directly. As such, a primary focus on core social communication may also have a secondary effect on RRB reduction. Often children demonstrate RRB when either overwhelmed or under-stimulated, so when children are more socially engaged and more able to participate in reciprocal interactions as a result of PRT, they then also demonstrate fewer RRB, respond more flexibly, shift between topics/activities more readily, and use more flexible, generative speech. For example, when a child is taking turns within a natural play scenario, it is unlikely that he/she can focus exclusively on an area of circumscribed interest, engage in repetitive behaviors, or insist on ritualistic behaviors. Furthermore, as treatment focused on social communication skills, children may have experienced less anxiety and uncertainty, which could also have related to a reduction in RRB.

The PRT trial included a high level of parent training (2 hours weekly) in addition to direct work with the child, so following the trial, parents were able to engage and support their children more effectively, which also may have led to reduction in RRB. For example, parents learned how to structure opportunities for their child to engage in flexible behavior (e.g., providing the child two choices of motivating activities that do not involve a circumscribed interest or ritualized behavior). Parents also learned how to support more flexible speech by prompting their child to use different ways of relaying ideas, as opposed to responding to stereotyped language.

Additionally of importance, circumscribed interests were used within the context of the treatment to motivate the child. Therefore, placing contingencies on RRBs, particularly circumscribed interests, may decrease the frequency of the behaviors, and the use of RRBs as natural reinforcement may make these behaviors more functional. Furthermore, practicing reciprocal interactions while engaging in a topic of strong interest may increase the child's ability to play and converse around a circumscribed interest more functionally.

Overall, the results thus far are promising, but there are clear limitations. This study is a pilot, so the sample size was small, and there was not a control group. Additionally, inclusion criteria included a diagnosis of ASD and overall cognitive abilities of IQ > 50, but only one child had an IQ < 70 (Full Scale IQ= 55 for the one child). Therefore, the results cannot generalize to all children with ASD. Furthermore and importantly, children were not selected based on severity of RRB, and the scores on the RBS-R and ABC stereotypy scales were lower than in studies that evaluated effects of medication on these domain (King et al., 2009; Scahill et al., 2015), although they were consistent with other studies on the clinical features of high-functioning children with ASD (Gabriels et al., 2008; Gabriels et al., 2005; Kanne & Mazurek, 2011). With these initial positive results, though, we have shown differential effects of PRT on a variety of RRBs, and PRT can be considered a promising treatment modality for reducing RRB. Future directions can include a study that selects children based on severity of RRB to assess symptom reduction in a group of children with high levels of RRB and also assesses RRB severity based on multiple sources, not just parent report. Future studies will allow a broader investigation of reduction in RRB, including assessing clinical meaningfulness, evaluating moderating variables on reduction of these behaviors, controlling for concomitant treatments, and assessing further RRB domains that are more/less responsive to treatment.

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Table 1

Participant demographic and clinical characteristics.

Male/Female	10/5
Age in years	6.11 (1.13)
General Conceptual Ability (DAS-II) *	98.60 (20.20), Range 55 to 128
Verbal *	94.00 (20.80)
Nonverbal *	105.40 (17.09)
Spatial *	97.20 (17.75)
ADOS Calibrated Severity Score (CSS)	15.33 (5.72)
CELF Core Language *	88.75 (23.13)
SRS-2 Total Raw Score	84.53 (21.80)

* Mean 100, s.d. 15

Table 2

Mean scores for Repetitive Behavioral Scales- Revised (RBS-R) and Aberrant Behaviors Checklist (ABC) before and after treatment.

	Pre-treatment		Post Treatment		
	Mean	**Std. Deviation**	**Mean**	**Std. Deviation**	**p-value**
RBS-R subdomains					
Stereotyped	4.58	4.03	2.42	2.57	0.009
Self-Injurious	1.83	2.12	0.83	1.47	0.020
Compulsive	4.33	3.26	2.33	2.42	0.046
Ritualistic	4.75	3.89	3.08	2.81	0.041
Sameness	7.50	6.50	4.67	4.19	0.012
Restricted	3.67	2.84	2.25	2.18	0.014
RBS-R Total Score	26.67	19.69	15.58	12.25	0.005
ABC Stereotypy subdomain	3.57	3.23	1.43	2.71	0.001