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Brief report: Reduced anxiety following Pivotal Response Treatment in young children with Autism Spectrum Disorder

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Abstract

Up to 40% of children with Autism Spectrum Disorder (ASD) exhibit co-occurring anxiety symptoms. Despite recent success in mitigating anxiety symptoms in school-aged children with ASD (mean age >9 years) using adapted versions of Cognitive Behavioural Therapy, little is known about potential treatment outcomes for younger children. To address the gap in the literature, this open-label study evaluated change in anxiety following a 16-week open-label trial of Pivotal Response Treatment (PRT) in children with ASD aged 4–8 years. PRT is a behavioural treatment based on the principles of Applied Behaviour Analysis and has a primary aim of increasing social communication skills in children with ASD through natural reinforcements. To minimise conflation of anxiety and other co-occurring symptoms such as disruptive behaviour and attention-deficit hyperactivity disorder, we measured anxiety using the autism anxiety subscale of the Child and Adolescent Symptom Inventory (CASI) devised by Sukhodolsky et al. (2008). We observed significant anxiety reduction over 16-weeks of PRT. Furthermore, anxiety reduction was independent of changes in autism symptom severity. This study shows promising results for PRT as an intervention for reducing anxiety in young children with ASD.

Keywords

Pivotal Response Treatment (PRT); Autism Spectrum Disorder (ASD); anxiety

1. Introduction

Autism Spectrum Disorder (ASD) is a pervasive developmental disorder characterised by social communication difficulties, and restricted and repetitive behaviour (American

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Conflicts of interests

The authors declare that they have no conflict of interest.

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Psychiatric Association, 2013). Up to 40% of individuals with ASD experience co-occurring symptoms of anxiety (Simonoff et al., 2008; Steensel, Bögels, & Perrin, 2011; White & Roberson-Nay, 2009). Developing and adapting existing interventions to simultaneously target social communication difficulties as well as anxiety in children with ASD can help to optimise children's long-term outcome, since heightened anxiety is associated with more behavioural outbursts (Kim, Szatmari, Bryson, Streiner, & Wilson, 2000; White, Oswald, Ollendick, & Scahill, 2009) and greater social withdrawal and avoidance (Markram & Markram, 2010), further compromising the development of social competency.

One type of intervention that has been adapted to treat anxiety in children with ASD is Cognitive Behavioural Therapy (CBT) (Barrett, 2000; Brewin, 2006; Wood & McLeod, 2008). Recent meta-analysis has shown large effects of CBT for anxiety reduction in children with ASD across randomised-controlled trials (Sukhodolsky, Bloch, Panza, & Reichow, 2013), though empirical studies have mostly focused on children with an average age of 9 years and up (Drahotka, Wood, Sze, & Van Dyke, 2011; Storch et al., 2013; Wood, Drahotka, Sze, Har, et al., 2009; Wood, Drahotka, Sze, Van Dyke, et al., 2009). Therefore, uncovering effective interventions for reducing anxiety in a younger (4–8 years) group of children with ASD requires further examination.

Interventions aimed to reduce anxiety in younger children with ASD need further reduced cognitive and language load due to even greater difficulty in higher-level cognition including perspective taking, metacognition, self-reflection, and cognitive flexibility than their older counterparts (Baron-Cohen, 1989). Scaffolding highly structured social interactions with more predictable patterns of action-outcome contingency during behavioural interventions may be particularly beneficial for young children with ASD and co-occurring anxiety, who may otherwise struggle with the unpredictability of the social world (Van de Cruys et al., 2014). Pivotal Response Treatment (PRT; Koegel & Koegel, 2012) is a behavioural intervention based on Applied Behaviour Analysis (ABA; Lovaas, 1987) with a large empirical evidence-base accumulated from both behavioural and neuroscience research (R L Koegel, Dyer, & Bell, 1987; Robert L. Koegel & Williams, 1980; Lei & Ventola, 2017a, 2017b; Pierce & Schreibman, 1995; Schreibman & Koegel, 1996), and endorses more natural rewarding contingencies, such as directly enabling the child to gain access to a desirable toy following successful attempt/response upon social bid. Direct and natural reinforcements in a structured manner serves to form a predictable style of social interaction, creating highly motivating circumstances to entice children with ASD to increase social engagement (Koegel & Mentis, 1985; Koegel & Williams, 1980).

Although PRT is not directly focused on anxiety reduction, the structured delivery of PRT that allow reciprocal social interactions to unfold in predictable manners may support anxiety reduction in young children with ASD. First, by teaching children social initiations, children's responses during reciprocal interactions are reinforced in predictable, yet naturalistic ways. Natural reinforcements contingent upon children's responses also help increase exposure to predictable action-outcome contingencies, further increasing the predictability of social interactions (Golan et al., 2009; Baron-Cohen, 2009). Second, anxiety in children with ASD is a form of emotion dysregulation associated with greater levels of social communication deficits (Sukhodolsky 2008, Hallett 2013). Improvements in

social communication targeted by PRT may also produce improvements in communication-based emotion regulation strategies and reduce anxiety. We therefore present the argument that the structured delivery of PRT, in combination with the explicit teaching of social communication skills to young children with ASD, may elicit therapeutic gains in anxiety reduction.

The current study aimed to evaluate reduction in anxiety in young children (aged 4–8 years) with ASD following a 16-week open-label trial of PRT. Given the paucity of literature in this area, the study is a preliminary, open-label trial to assess possible reduction of anxiety symptoms in this younger age group following the trial of PRT. We also sought to explore how anxiety reduction is related to improvement in social communication skills. Anxiety reduction independent from co-occurring changes in autism symptom severity would support our hypothesis that it can be fully considered as a primary positive therapeutic outcome, rather than a consequential change following social gains made over the course of PRT. Finally, we examined whether the magnitude of anxiety reduction may be a function of development and cognitive capacities in young children with ASD, by partitioning the variance associated with change in anxiety when taken into consideration the effects of age and IQ.

2. Methods

2.1 Participants

Twenty-one children (9 female, 12 male) aged 4–8 years old were recruited from the community using public announcements and from clinic referrals. This study protocol was approved by the university Institutional Review Board (IRB), and Human Investigation Committee (HIC) #1106008625, and is in line with the Declaration of Helsinki as revised in 2000. Written informed consent was obtained from each participant's parent(s), and assent was obtained from each participant.

All children entered the study with a prior diagnosis of ASD. Inclusion criteria included using the “gold standard” diagnostic procedures: Autism Diagnostic Observation Schedule (ADOS; Lord et al., 2000) and the Autism Diagnostic Interview-Revised (ADI-R; Lord, Rutter, & Couteur, 1994) to further inform diagnosis, as well as IQ ≥ 70 as measured by the General Conceptual Ability (GCA) of the Differential Abilities Scale-II (DAS-II; Elliott, 2007). Clinicians administering the ADOS and ADI-R were research reliable. See Table 1 for participants' characterisation and demographics. All children had IQ ≥ 70 (GCA: M 102, SD 16.31), and received special education in school, including speech and language therapy and social skills support. No children were taking any psychotropic medications. Upon commencing PRT, parents were asked not to start any other behavioural interventions during the 16-week period of the study. No major changes in educational placement or services received were reported during the study.

2.2 Treatment Approach and Fidelity of Implementation

PRT is a behaviourally-based treatment designed to increase social motivation to enhance quality of social interaction in children with ASD (Koegel, Carter, & Koegel, 2003). A more

detailed description of PRT is provided in the updated instruction manual (Koegel & Koegel, 2012), and see *Supplementary Material A*. For the current study, each child received three treatment sessions (7h total) weekly for 16 weeks. Treatment per week consisted of 5h of direct intervention with the child, and 2h of parent guidance. During each PRT session, clinicians delivered activities in a structured manner following the antecedent-behaviour-consequence (ABC) model, delivering bids of varying levels of demand (antecedent) to prompt the child to engage in social interaction (behaviour) followed by natural reinforcement (consequence) contingent upon child's appropriate reciprocal social response. At the lowest level of scaffolding, the clinician may withhold the object of interest from the child during an activity and wait for the child to make an appropriate attempt to request for the material at hand. When no response is provided, the clinician will explicitly draw the child's attention to the opportunity for reciprocal social interaction by asking a question such as "How can you ask? / What could you say?" (Intermediate scaffolding). Finally, the clinician will model the appropriate response for the child to follow, such as saying "You could say..." (highest scaffolding). In the current project, by using child-preferred activities, children were highly motivated to engage in reciprocal social interchanges with the clinicians. In turn, clinicians were able to scaffold many learning opportunities within each activity to help children gain perspective taking skills, as well as increasing functional use of language, thus building upon their basic language skills in this sample of children aged 4–8.

Treatment goals focused on improving social initiation, responsiveness, and reciprocity (Koegel & Koegel, 2012). All clinicians were bachelor and master-level students who received training and supervision from the lead clinician (licensed clinical psychologist) to deliver PRT. All clinicians passed fidelity assessment prior to starting PRT sessions for the current study, and maintained fidelity for the duration of the study. See *Supplementary Material B* for details on criteria for maintenance of fidelity of implementation.

2.3 Outcome Measures

Child and Adolescent Symptom Inventory–Autism Anxiety Subscale (CASI-Aut-Anx; Gadow & Sprafkin, 2002)—CASI-5 is a 132-item parent-report questionnaire, which evaluates a range of DSM-V referenced disorders. Symptom severity is calculated by the sum of total raw score across the items within any specific disorder category. CASI-5 has good sensitivity (0.64–0.8) and specificity (0.65–0.96) in detecting a broad range of psychiatric disorders ranging from disruptive behaviour, to anxiety (Gadow & Sprafkin, 2002). The current study uses the 20-item CASI-5 autism anxiety subscale (CASI-Aut-Anx), previously shown to be able to sensitively and reliably detect anxiety in children with ASD (Sukhodolsky et al., 2008). Detailed explanations for the selection of items are provided by Sukhodolsky et al. (2008), and see *Supplementary Materials C*.

Child Behaviour Checklist (CBCL; Achenbach & Dumenci, 2001)—The CBCL is a 113-item parent-report questionnaire, widely used to assess emotional and behavioural problems in children. CBCL scoring is broadly divided into Internalising, and Externalising domains as well as factor-analytically derived subscales of specific forms of psychopathology. The current study focused on using CBCL's Internalising Problems domain, which includes symptoms of anxiety, depression and somatic complaints.

Social Responsiveness Scale-Second Edition (SRS-2; Constantino, 2012)—

SRS-2 is a 65-item parent-report questionnaire, assessing autism symptom severity, and primarily social communication competency in individuals with ASD. It includes five sub-domains: Social Awareness, Social Cognition, Social Communication, Social Motivation, and Autistic Mannerisms, with the sum yielding a total raw score. SRS-2 has high test-retest reliability ($r = .88$) in clinical populations, and has high concurrent validity with ADI scores ($r = .6$ to $.79$) (Constantino et al., 2003).

2.4 Data Analyses

Mothers of all twenty-one children completed CASI-5, CBCL, and SRS-2 both at the beginning (t1), and the end (t2) of a 16-week open-label trial of PRT. Raw scores were utilised for analyses with CASI-Aut-Anx and SRS-2 because they provided a greater range of scores to better capture the spectrum in symptom severity across our sample. T-scores from CBCL's internalising symptom domains were utilised for analyses to increase comparability across the different age-appropriate forms.

Changes in anxiety and other outcome measures were evaluated in three steps. In step one, paired sample t-tests were conducted to evaluate changes in anxiety (CASI-Aut-Anx), internalising symptoms (CBCL), and autism symptom severity (SRS-2) over the course of treatment. We assessed changes in broader internalising behaviour (CBCL) as a secondary outcome measure to examine concurrent validity with CASI-Aut-Anx. To control for effects of variation in initial symptom severity at t1 on magnitude of change over time (i.e., regression towards the mean), residualised change scores for each outcome measure were calculated by taking the predicted value at t2 (based on t1 value) from the actual observed value at t2.

In step two, Pearson's correlations between the residualised change scores of anxiety, and autism symptom severity were evaluated, to evaluate whether any change observed in anxiety was independent of change in autism symptom severity. We also conducted repeated-measures ANCOVA, to assess independence of changes observed in anxiety and autism symptom severity.

In step three, we sought to partition the variance associated with change observed in anxiety over the course of treatment, by including change observed in autism symptom severity, IQ, and age at t1 as independent variables in a step-wise hierarchical linear regression analysis. In both steps two and three, residualised change scores for anxiety (CASI-Aut-Anx) was used, as the primary focus of the paper was change in anxiety.

3. Results

Mean scores of all outcome measures across the sample at both t1 and t2 are shown in Table 2. Parents reported significant reductions in anxiety ($p = .02$) and internalising symptoms ($p = .003$) following PRT. CASI-Aut-Anx significantly correlated with CBCL's internalising subscale t-score ($r = .76$, $p = .00$), indicative of high concurrent validity. Significant reduction in autism symptom severity ($p = .001$) is consistent with our previous reports (Ventola et al., 2014; Ventola et al., 2016; Yang et al., 2016).

Residualised change score of anxiety and autism symptom severity were not correlated ($r=.14$, $p=.552$). When controlling for the residualised change in autism symptom severity (SRS-2) as a continuous covariate, participants still showed a significant reduction in anxiety from pre- to post-treatment, $F(1,19)=6.13$, $p=.023$, partial $\eta^2=.24$. In a step-wise hierarchical linear regression model (Table 3), residualised change in autism symptom severity was not associated with residualised change in anxiety symptom severity ($\beta=.14$, $t(19)=.61$, $p=.55$) and did not account for much of the variance associated with change reported in anxiety ($p=.55$). Adding IQ (GCA) measured at t1 as a covariate did not change the model's ability to account for variance associated with changes in anxiety over PRT ($p=.99$). Further addition of age at t1 as a covariate did not alter the model's ability to account for changes observed in anxiety symptom severity over PRT ($p=.75$).

4. Discussion

Over a 16-week open-label trial of PRT, parents reported significant reduction in anxiety among young children with ASD. Anxiety reduction was independent of autism symptom severity reduction and was not associated with participants' age or IQ. Therefore, anxiety reduction may be an additional therapeutic component of PRT, beyond supporting social communication competency in young children with ASD. The significant anxiety reduction has many notable theoretical and clinical implications. By increasing the predictability of social interactions in PRT (e.g., through structured delivery of social bids, setting clear expectations), children with ASD may find social exchanges less anxiety provoking and more engaging. Our finding that the magnitude of anxiety reduction could not be accounted for by reductions in autism symptom severity alone supported our idea that the structured PRT delivery style may be helpful to reduce anxiety symptoms, and may be an active therapeutic element that is less frequently discussed.

However, the structured antecedent-behaviour-consequence model is an integral part of behavioural interventions in general, and thus it is important to conjecture which mechanisms specific to PRT might be helpful in reducing anxiety in young children with ASD. First, by increasing exposure to natural reinforcements during PRT, children with ASD may be able to learn to adopt functional social schemas by increasing their ability to predict how social interactions can unfold in an ecologically valid manner, similar to how typically developing children may acquire similar skills through natural play activities. Second of all, by using child-directed and preferred activities during PRT, not only can it help young children with ASD to better sustain their social motivation during intervention, but may also help control some uncertainty associated with the task at hand as children are more familiar with the sequence of events that typically unfold during their preferred activities. Therefore, PRT may make use of the zone of proximal development as proposed by Vygotsky (1978) by slowly scaffolding new and more complex levels of social interactions embedded into children's preferred activities, to slowly enable children to master social skills in a less anxiety-provoking and natural manner.

Finally, the independence of autism symptom severity and anxiety reduction suggest the changes are unlikely due to a halo effect for positive change from parental expectancy, as parents did not report similar levels of improvement on all measures. Anxiety reduction over

PRT was not predicted by children's age or cognitive capacity, indicating that the intervention effect was not necessarily specific to the age range or functioning level included in the current study.

It should be noted that the range of anxiety symptom severity scores within our sample (CASI-Aut-Anx; $M=7.81$, $SD=5.9$) overlap and fall on the lower end of the range evaluated by Sukhodolsky et al. (2008) in the validation study of the 20-item CASI-Aut-Anx scale ($M=13.5$, $SD=8.5$), conducted in a sample of 172 children and adolescents with autism. Given our younger sample ($M=5.94$, $SD=1.11$) with a narrower age range (4–8 years) than Sukhodolsky et al. (2008) ($M=8.2$, $SD=2.6$, range 5–18 years), the difference in scores is likely due to inclusion of older participants in the validation study. Future studies can examine effectiveness of PRT at anxiety reduction in younger children with ASD and more severe anxiety symptoms to evaluate the generalisability of current findings.

Our current study also provides further evidence supporting the abridged version of CASI-Aut-Anx as an example of successfully adapting questionnaire to reliably detect anxiety symptoms in young children with ASD (Sukhodolsky et al., 2008). By removing items that might measure overlapping symptom constructs with other co-occurring symptoms in children with ASD, CASI-Aut-Anx allow parents' reports to better identify and distinguish behaviours associated only with anxiety, and avoid "diagnostic overshadowing" (MacNeil, Lopes, & Minnes, 2009; Mason & Scior, 2004; Mayes, Calhoun, Murray, Ahuja, & Smith, 2011; White et al., 2009). Therefore, our study raises the broader issue of the need to develop and validate questionnaires that can reliably detect anxiety in young children with ASD, taking into account differences in clinical manifestation to help effectively evaluate treatment outcome.

Several limitations of the current study should also be mentioned. A major limitation of the current study is the lack of well-matched control group of high-functioning children with ASD who served as either waitlist controls, or randomised to receive more traditional forms of ABA or alternative behavioural treatments. Without a control group, it is difficult to identify the mechanism by which PRT might specifically be able to support anxiety reduction in young children with ASD. Although we discussed potential benefits of utilising child-preferred activities, using natural reinforcements, and delivering social prompts to set up antecedents in a structured manner as all possible viable mechanisms of action for anxiety reduction in young children with ASD via PRT, it is important to acknowledge that whereas the first two points are unique to PRT, the latter is a more commonly used approach across many behavioural-based interventions. Therefore, in order to distil the active ingredients of PRT that are key to anxiety reduction, future studies can look to conduct randomised-controlled trials comparing PRT to more traditional behavioural approaches that are less natural and utilise fewer child-directed activities to explicitly test the viability of the mechanisms proposed that are unique to PRT.

Three additional limitations are also outlined. First, the sample size ($N=21$) was appropriate for a preliminary open-label trial, but results require replication in a larger sample. With a larger sample, future studies may also investigate gender effects on treatment response. Second, our outcome measures were based solely on parent report, and lack more objective

measures such as clinician ratings or qualitative observation, and potential rater-bias needs to be taken into consideration when interpreting the current results. However, the finding that anxiety reduction occurred independent of reduction in autism symptom severity is important to note, as it suggests that the patterns of reductions reported by parents are not uniform across different symptoms, and that anxiety reduction reported by parents is not a mere reflection of overall improvement in symptom severity over the course of treatment. Future studies should seek to obtain and integrate reports from multiple informants including parent and clinician ratings to help better quantify, and assess the convergence on ratings of anxiety reduction following PRT. Finally, further investigation is needed to fully evaluate whether PRT may be an effective intervention for children across the autism spectrum, including children with more limited language and social interaction skills than the current sample.

5. Implications

Our study is the first to address the gap in the literature on developing, adapting, and evaluating treatment outcomes on anxiety in young children with ASD. Our study provides initial evidence suggesting PRT, a well-established evidence-based treatment for children with ASD, may bear significant additional therapeutic potential in anxiety reduction among young children with ASD. Although preliminary, these positive results are very encouraging. Given the prevalence and long-term negative impacts of anxiety on the daily functioning of children with ASD, interventions that can effectively target these symptoms at a younger age are necessary to maximise long-term treatment outcome. Future studies can investigate with more breadth and depth the extent of existing intervention's effectiveness on reducing anxiety, and identify other potential mediators and moderators that may predict therapeutic outcome, to provide more individualised and cost-effective treatments for children with ASD.

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Highlights

- Pivotal Response Treatment (PRT) is a natural behavioural intervention based on principles of Applied Behavioural Analysis, targeting social communication skills in young children with Autism Spectrum Disorder.
- Following an open-label 16-week trial of PRT, parents reported significant anxiety reduction in 21 young children (aged 4–8) with ASD.
- Anxiety reduction occurred independent of reductions in autism symptom severity.
- Scaffolding highly structured and predictable social interactions during PRT may be an additional and independent therapeutic component for anxiety reduction.

Table 1

Sample demographics and clinical characteristics.

	Mean	SD	Range
Age in years (N=21)	5.94	1.11	4–8
DAS-II (IQ) (N=21)			
Verbal	96.67	16.32	67 – 123
Non-Verbal	105.14	16.34	78 – 136
Spatial	102.29	16.08	70 – 137
GCA	102.29	16.31	70 – 128
ADOS 2-Module 2 (n=9)			
Social Affect	11.11	2.89	6 – 16
Repetitive Behaviour	3.89	1.76	2 – 8
Total (CSS)	7.11	1.05	5 – 8
ADOS 2-Module 3 (n=12)			
Social Affect	10.75	4.88	4 – 20
Repetitive Behaviour	3.08	2.15	0 – 7
Total (CSS)	7.08	2.43	3 – 10
ADI-R (N=21)			
Social	18.48	6.89	5 – 29
Verbal Communication	16.52	4.81	5 – 22
Repetitive Behaviour	7.00	2.68	1 – 12

Note. DAS-II[†] = Differential Ability Scales–II; GCA = General Conceptual Ability; ADOS = Autism Diagnostic Observation Schedule; CSS = Core Severity Score; ADI-R = Autism Diagnostic Interview–Revised

[†]DAS-II is a standardised cognitive assessment of functioning in verbal, nonverbal, and spatial reasoning skills. DAS-II is commonly used in studies for children with ASD to assess cognitive abilities, as it has a lower demand on use of functional language compared to other forms of cognitive testing (Frazier, Georgiades, Bishop, & Hardan, 2014; Kanne et al., 2011), and has high convergent validity with Mullen Scales of Early Learning (Bishop, Guthrie, Coffing, & Lord, 2011).

Table 2

Changes in outcome measures before and after treatment (N=21).

	Pre-Treatment		Post-Treatment		<i>t</i> (20)	<i>p</i> Value	Cohen's <i>d</i>
	Mean	SD	Mean	SD			
CBCL Int (t-score)	57.24	11.18	52.00	10.18	-3.41	.003	.49
CASI-Aut-Anx	7.81	5.90	6.24	5.61	-2.54	.020	.55
SRS-2 Total Raw	87.52	26.12	71.61	23.71	-3.71	.001	.81

Note. CBCL Int = Child Behaviour Checklist Internalising problems; CASI-Aut-Anx = Child and Adolescent Symptom Inventory Autism Anxiety Subscale; SRS = Social Responsiveness Scale

Table 3

Summary of step-wise hierarchical regression analysis for variables accounting for variances associated with residualised change observed in anxiety (CASI-Aut-Anx) over Pivotal Response Treatment.

Step/Variable	Step 1		Step 2		Step 3	
	<i>B</i> (<i>SE</i>)	β	<i>B</i> (<i>SE</i>)	β	<i>B</i> (<i>SE</i>)	β
1. SRS-2 total raw	.02 (.04)	.14	.02 (.04)	.14	.02 (.04)	.15
2. IQ (GCA)	-	-	.00 (.04)	.00	.01 (.04)	.04
3. Age	-	-	-	-	.02 (.05)	.05
<i>R</i> ²	.02		.02		.03	
<i>F</i>	.37		.00		.11	

Note. CASI-Aut-Anx = Child and Adolescent Symptom Inventory Autism Anxiety Subscale; SRS-2 = Social Responsiveness Scale-2; GCA = General Conceptual Ability