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Examining a novel, parent child interaction therapy-informed, behavioral treatment of selective mutism

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ABSTRACT

Background: The purpose of this study was to evaluate a new therapy for children with selective mutism (SM) that combines Parent-Child Interaction Therapy principles and behavioral techniques.
Method: Children aged 4–10 with a primary diagnosis of SM were eligible to participate. Comorbidity was allowed with the exception of autism spectrum disorder, intellectual disability, mania or psychosis. Of 54 potentially eligible participants, 33 met inclusion/exclusion criteria of which 31 families consented (94%). Following assessment, children were waitlisted for an average of 4 months before receiving 16 sessions of weekly therapy at an outpatient psychiatry clinic of a children's hospital in Vancouver, Canada; all children completed treatment. Assessments were conducted at time of referral (baseline), pre-treatment, post-treatment, 3 month follow up, and 1 year follow up. Two did not complete follow up assessments (93% retention).
Results: Results showed significant and large (Cohen's $d = 1.80$) gains in speaking behaviors across contexts from pre- to post-treatment. Gains were maintained at 3-months and 1-year post-treatment. Statistically significant and large improvements were also found in post-treatment teacher and parent reports of child anxiety as well as a behavioral measure of the child speaking to an unknown adult. Parents reported high satisfaction with treatment. None of the potential predictors of treatment response examined were found to be significant.
Conclusions: PCIT-SM appears to be an effective treatment for children aged 4–10 with SM.

1. Introduction

Selective mutism (SM) is an anxiety disorder in which children consistently fail to speak in situations where speaking is expected despite generally normal speaking in other situations (American Psychiatric Association, 2013). SM occurs in about 0.75% of children (Bergman, Piacentini, & McCracken, 2002) and persists for many without treatment (Remschmidt, Poller, Herpertz-Dahlmann, Hennighausen, & Gutenbrunner, 2001). SM negatively impacts children's educational achievement, language development, emotional well-being, and social competence (Cunningham, McHolm, & Boyle, 2006). While the reported mean age of onset ranges from 2.7 to 4.1 years, the average referral age is 6 to 9 years (Viana, Beidel, & Rabian, 2009).

Although the specific etiology of SM is still debated, several risk factors have been identified, including behaviorally inhibited temperament (Muris, Hendriks, & Bot, 2016) and a history of speech or expressive language difficulties (Klein, Armstrong, & Shipon-Blum, 2013). SM is also more prevalent in immigrant children (Elizur & Perednik, 2003). While some children with SM appear to have behavior problems as well as anxiety, a review of the available evidence suggests that oppositionality is an unlikely etiological path to SM (Muris & Ollendick, 2015). Regardless of the specific etiology for a given child to develop SM, it is important that interventions target factors that maintain the disorder. One such factor is the use of "non-speaking behavior" as an avoidance strategy to reduce anxiety in stressful situations, such as school or other social situations (Scott & Beidel, 2011). The commonly seen tendency of others to answer for the child with SM or avoid situations in which the child is asked to talk potentially strengthens this avoidance strategy by creating a negative reinforcement cycle in which the child is asked a question, feels anxious and avoids answering, and is then 'rescued' or withdrawn from speaking demands. This cycle leads to short term relief from anxiety that ultimately strengthens the avoidance and, given the frequency of speaking demands, becomes well practiced over time (Young, Bunnell, & Beidel, 2012).

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Research into SM treatment is growing with recent RCTs of behavioral interventions showing particular promise (Bergman, Gonzalez, Piacentini, & Keller, 2013; Oerbeck, Johansen, Lundahl, & Kristensen, 2012; Oerbeck, Overgaard, Stein, Pripp, & Kristensen, 2018; Oerbeck, Stein, Wentzel-Larsen, Langsrud, & Kristensen, 2014). While these studies indicate that behavioral treatments can be effective, more research with larger sample sizes, predictors of response, and multimodal outcome measures are needed. Additionally, given the theorized role of parental accommodation in maintaining SM (Roslin, 2013), parent-child interaction is a potentially important yet under researched component of therapeutic intervention for SM.

1.1. Parent-child interaction therapy for selective mutism (PCIT-SM)

Parent-child interaction therapy (PCIT) is a well-established treatment for disruptive behavior disorders (Funderburk & Eyberg, 2011) that focuses on restructuring parent-child interaction patterns that maintain or worsen child symptoms. PCIT has been successfully adapted and shown to be effective in the treatment of anxiety disorders in young children (e.g., Choate, Pincus, Eyberg, & Barlow, 2005; Puliafico, Comer, & Albano, 2013). The primary focus of these modifications of PCIT for anxiety is providing parents with specific tools to counteract the cycle of negative reinforcement patterns that serves to strengthen avoidance in their child with anxiety. Given parental inadvertent strengthening of nonspeaking as an avoidance strategy in SM and the young age of onset for this disorder, a modified version of PCIT is worthy of consideration.

PCIT-SM is an intervention that combines PCIT and behavioral intervention for children with SM (Carpenter, Puliafico, Kurtz, Pincus, & Comer, 2014; Cotter, Todd, & Brestan-Knight, 2018) by teaching parents skills to maximize opportunities for child verbalization while eliminating parent behaviors that support avoidance. Components in PCIT-SM include guided questioning to increase likelihood of speech, contingency management (rewards for talking), and in-vivo exposure in hierarchically-ordered situations. While SM is not generally conceptualized as a parent-child relational problem (Muris & Ollendick, 2015), parents nevertheless get 'pulled into' a cycle of rescuing their child from anxiety-provoking situations, and parents can be important agents of change in supporting their children to increase their verbal communication across settings.

While the behavioral focus of PCIT-SM overlaps with other recently published treatment approaches in its use of exposure, contingency management, and graded steps towards talking (e.g., Bergman et al., 2013; Oerbeck et al., 2014; Oerbeck, Stein, Pripp, & Kristensen, 2015) it also differs in important ways. Unique features include helping parents master two skill-sets to help facilitate speech in their child: a child-directed interaction (CDI) technique to help the child warm up in a new situation *before* verbal demands are made, thus minimizing continued negative reinforcement of non-speaking behaviors, and a verbal directed Interaction (VDI) technique that directly teaches parents to use a structured series of prompts to encourage and reward the child's verbal responses as well as strategies to manage non-responses.

The present treatment included four modules: 1) parent coaching on CDI and VDI, 2) office-based exposure work, 3) three school outreach visits to train teachers and practice with the child, and 4) daily parent-led community exposures. To date, PCIT-SM has been utilized in clinical settings and has shown initial promise in a single-case study (Mele & Kurtz, 2013). A version modified for intensive group treatment has recently received empirical support (Cornacchio et al., 2019). This study is the first to examine the efficacy of individually-delivered PCIT-SM in a larger, controlled clinical trial.

The goals of the current study were to evaluate PCIT-SM in a within-subject, waitlist-controlled, longitudinal study. We hypothesized that a) there would be no change in SM symptoms during the wait period, b) treatment would be associated with a significant decrease in SM symptoms, and c) gains would be maintained at 3-months and 1-year follow-up. A secondary objective was to examine predictors of treatment response. Given previous research showing greater treatment benefits in younger children (Oerbeck et al., 2015) and the likelihood that older children have had much more 'practice' not speaking in social situations, we hypothesized that younger children would show greater gains post-treatment. Similarly, children with comorbid language challenges who may have other factors contributing to their communication challenges may respond less well to treatment. Therefore, we also hypothesized that those with higher baseline language ability would show greater gains post-treatment.

2. Method

2.1. Participants

Participants were recruited from the outpatient mental-health department of a children's hospital in Vancouver, Canada. Children aged 4–10 years meeting DSM-5 diagnostic criteria for SM, with sufficient English language ability in the child and 1 caregiver to complete the therapy and measures in English were eligible to participate. Exclusion criteria included psychosis, mania, Autism Spectrum Disorder, or intellectual disability. Children on psychotropic medication were included following 8 weeks at stable dose. Medication information was examined in analyses on response to treatment.

2.1.1. Descriptives

Thirty-one children participated in the study (15 boys and 16 girls). Participant ages ranged from 4.0 to 9.75 years ($M = 6.47$, $SD = 1.68$). The sample was ethnically diverse with 41.9% self-identifying as Asian, 35.5% as European / White, 9.7% as multiracial, 3.2% South Asian, and 9.7% as non-specified.

A third (32.3%) of children, 51.9% of mothers and 50% of fathers had a first language other than English.[1] Thirty-one percent of children had previous SM treatment. Eighteen fathers and 19 mothers self-reported educational information. Paternal education ranged from high school completion (9.7%), some post-secondary education (12.9%), and completed post-secondary (35.5%) while maternal education ranged from some post-secondary education (12.9%) to completed post-secondary education (48.4%).

The study was approved by the Research Ethics Boards of the University of British Columbia/BC Children's Hospital. Fifty-four potential participants were identified. Of those, 33 met inclusion/exclusion criteria. Two declined to participate; 31 completed informed consent/assent and baseline assessments. Thirty completed treatment[2]; 28 participants were retained for 3-month and 1-year follow up assessments.

2.2. Measures

2.2.1. Initial SM diagnosis and severity

The *Anxiety Disorders Interview Schedule for Children for DSM-IV: Parent Version (ADIS-P;* Silverman & Albano, 1996) is a semi-structured interview that was used to establish DSM-IV diagnoses and disorder severity, with clinician severity ratings ranging from 0 to 8 (4 = moderate; 8 = very severe). The ADIS-P anxiety sections have demonstrated strong concurrent validity (Wood, Piacentini, Bergman, McCracken, & Barrios, 2002) and test-retest reliability estimates range from acceptable to excellent (Silverman & Ollendick, 2005; Silverman, Saavedra, & Pina, 2001).

[1] Data were available for 27 mothers and 26 fathers.

[2] One girl experienced remission during the wait period after fluoxetine initiation and was withdrawn from the study due to lack of need for further treatment. Her data is included at baseline and pre-treatment so as not to bias results regarding any effect during wait time.

The *Selective Mutism – Behavioral Observation Task (SM-BOT),* a structured behavioral assessment task (Carpenter et al., 2014), provided qualitative and quantitative information on SM symptom severity. The task examines verbal interactions between the child and parent in the presence and absence of a stranger, followed by structured interaction tasks with the stranger. In our study, the child was coded as having: a) no interaction with the stranger, b) non-verbal interaction only, or c) non-verbal and verbal interaction. There are no normative data for this measure.

2.2.2. Outcome measures

The *Selective Mutism Questionnaire (SMQ*; Bergman, Keller, Piacentini, & Bergman, 2008) is a 17-item, parent-rating scale that assesses SM in 3 settings: home, school, and community. Parents are asked to report the frequency of speaking behavior in the past two weeks. Items are scored on a 4-point scale of speaking frequency: 0 (never), 1 (seldom), 2 (often) or 3 (always). In the present study, mean scores were used which allows comparisons across subscales. The SMQ demonstrates good internal consistency and successfully distinguishes children with SM from those with social phobia or other anxiety disorders (Manassis et al., 2007). Futhermore, it is sensitive to change following treatment (e.g., Bergman et al., 2008). There are no clinical cutoffs for this measure; however, in the standardization sample a mean score of 0.76 in children with SM was reported, vs a mean score of 2.7 in children without SM (Bergman et al., 2008). The SMQ is a gold standard measure that is used as a primary outcome measure in all published papers to date on SM treatment and is the primary outcome measure in this study.

The *School Speech Questionnaire (SSQ)* is a teacher-rating scale, modified from the SMQ, that provides information on the child's speech at school over the past month. Items are rated on the same scale as the SMQ (see above for details), allowing for direct comparison between teacher and parent report in school settings. It shows good internal consistency and validity (Bergman et al., 2002) but no normative data exist.

The *Strong Narrative Assessment Procedure* (*SNAP;* Strong, 1998) is a standardized procedure to assess expressive language in which a child retells a story initially presented in an audiotape and wordless storybook. Three parallel stories are included. It has been used to measure language abilities in children with SM (McInnes, Fung, Manassis, Fiksenbaum, & Tannock, 2004). The dependent variable was the number of spoken words during the child's retelling. The order of stories was randomized and a novel examiner was used for each assessment point.

The *Screen for Child Anxiety Related Emotional Disorders – Parent Version (SCARED – PV*; Birmaher et al., 1997) is a parent-report measure demonstrating good internal consistency, test-retest reliability, and utility as a measure of anxiety in children as young as 5 years old (Birmaher et al., 1999; Monga, Young, & Owens, 2009). This screening measure uses a cutoff score of 25 as indicative of a possible anxiety disorder. Screening cutoffs are as follows for subscale scores: 7 (panic disorder/significant somatic symptoms); 9 (generalized anxiety disorder); 5 (separation anxiety disorder); 8 (social anxiety disorder); 3 (significant school avoidance).

The *Client Satisfaction Questionnaire (CSQ-8*; Attkisson & Zwick, 1982) is an 8-item questionnaire that assesses satisfaction with treatment on a 4 point scale. Mean scores are reported, with higher numbers reflecting greater satisfaction with treatment. It is a reliable and valid measure (Attkisson & Zwick, 1982).

2.2.3. Language competence measures

The *Peabody Picture Vocabulary Test-4 (PPVT-4*; Dunn & Dunn, 2007) is a receptive vocabulary test that requires participants to point to a picture that corresponds with a spoken word delivered by the examiner (in this case a graduate student research assistant). The PPVT-4 has excellent psychometric properties and is widely used as a measure of verbal ability.

The *Expressive Vocabulary Test-II (EVT-2*; Williams, 2007) is an expressive vocabulary test in which the child is asked to name a picture. Test-retest reliability and convergent validity are good (Williams, 2007). Given that the EVT-2 requires a verbal response, this measure was administered by parents who were coached via 'bug-in-the ear' through a one-way mirror following brief instruction to ensure standardized administration.

Both the PPVT-4 and EVT-2 have a mean of 100 and a standard deviation of 15 in the general population.

2.3. PCIT-SM treatment protocol

Treatment focused on graded exposure to talking and consisted of 16 one-hour sessions, conducted over a maximum of 22 weeks. After an initial parent-only training session, most sessions included the parent, child, and therapist. Three of the 16 sessions were school outreach visits. The treating clinicians included a clinical psychologist and a child psychiatrist who completed intensive training in PCIT-SM, followed by demonstration of CDI/VDI skills to mastery criteria. Monitoring of treatment adherence was performed at each session using a checklist developed for the study that was completed by the therapist to note the presence or absence of various treatment components at each session (coaching parents, parent feedback, exposure tasks in hospital setting, homework planning, and school modules). Therapists also had monthly meetings to ensure consistency and fidelity. The PCIT-SM treatment includes 4 key components, described in detail below (see also Carpenter et al., 2014).

The first treatment component is focused on parent education on verbal interaction techniques that form the core of the PCIT-SM intervention. Parents are trained until they achieve mastery of these techniques. Both the therapist and parent use CDI and VDI skills throughout treatment, and teachers are coached on these skills as well. CDI is used in situations where the child is not ready for speech and as a warm-up prior to placing speech demands. It consists of engaging with the child using behavioral descriptions (e.g. "You're coloring with the yellow marker,"), labeled praise playing to child's strengths ("What a great drawing!"), and reflection of any spontaneous verbalizations following by labeled praise for talking (e.g. "It's a sun. Thanks for telling me"). In situations where information is needed from the child (e.g., finding out whether the child needs the bathroom), "question end-arounds" are created where the child can indicate the answer without a prompt to speak (e.g., "If you need to go to the bathroom, put the card on my desk"). CDI is a positive, child-focused interactional style, and as such even anxious children appear to enjoy the experience and perceive it as non-threatening.

In contrast with CDI, VDI places demands for speech. It is used in situations where the child is warmed-up and comfortable and where it is realistic that the child will be successful in talking. VDI uses the techniques of CDI but adds direct prompts to talk. These prompts take the form of either a forced choice question (e.g., "Do you want the yellow marker or the red marker?") or an open-ended question (e.g., "What is your favorite color?"). Yes/no questions are avoided as they typically result in a non-verbal response (head shaking or nodding). Once a question is asked, the child is given 5 s to respond. If the child does not provide a verbal response within this time frame, the question is rephrased or the situation is changed to make it easier for the child (e.g. "I'll step outside, you tell your mom and then I'll come back and she can tell me what you said"). As in CDI, "mind-reading" and "negative talk" are avoided. Successful answers to speech prompts are reflected and praised. These two components are used together to minimize continued negative reinforcement of non-talking and to maximize opportunities to successfully engage in speaking tasks.

The second treatment component is graded exposure therapy to talking during individual treatment sessions with the therapist, parent and child. The weekly sessions typically start in the therapist's office

	ADIS	PPVT/ EVT	SM-BOT	SMQ	SSQ	SCARED	SNAP	Treatment Satisfaction
Baseline	✓	✓	✓	✓	✓	✓	✓	
Pre-Treatment				✓	✓	✓		
Post-Treatment				✓	✓	✓	✓	✓
3 Month Follow-up				✓	✓	✓	✓	
1 Year Follow-up				✓	✓	✓		

Fig. 1. Assessment measures across timepoints.

and expand to include a variety of settings and people. Rewards for talking are provided. CDI and VDI skills are used to maximize comfort and speech. In early sessions, a "fade-in" technique is used, in which a new person is gradually introduced into the situation (e.g., first sitting outside the door with the door open, then sitting across the room not engaging, then beginning to comment on the situation, then getting closer, then joining the play), with rewards for each step.

The third treatment component is parent-driven exposure practice in community settings. Parents are asked to lead at least 5 exposures per week between sessions, and to gradually incorporate different settings and people. Children are given rewards for successful talking during these exposures. Suggested homework exercises that follow the gains the child has made in the treatment session are shared with families.

The final component is school education and exposures. School involvement starts with an education session with school staff, where information about SM and CDI / VDI and "fade in" techniques is provided. Subsequently, 1–2 exposure sessions are conducted in the school with the therapist. The goal of the school exposure sessions is typically to get the child to successfully speak to one new adult (often the classroom teacher) who may then carry on school-based exposures. Specific school-led exposures are recommended by therapist, with phone support to the teacher to troubleshoot difficulties.

The timing of the various treatment components is flexible and dependent on a child's progress. Components are typically covered contiguously (e.g., a session may include a 5-minute observation of parent skills in CDI and VDI with feedback, an in-hospital exposure task, and assignment of homework tasks to work on over the following week). In the present study, an average of 54% of sessions contained formal parent coaching, 59% contained planning for parent-led community exposures between sessions, and 67% contained in-session exposures in new settings or with new people. Three school outreach visits, two parent 'teach' sessions on skills without the child present, and a final 'celebration' and future planning session with the child were also provided.

2.4. Procedure

Children referred for clinical assessment of possible SM in the outpatient hospital setting standardly receive a psychiatric assessment, which includes an ADIS-P supplemented by standardized measures, the SM-BOT, and a feedback session that provides diagnoses and recommendations. Following clinical assessment, parents of children eligible for the study were provided with information and, if interested, completed the consent process with a research assistant which included consent for chart review to gather the baseline clinical data. Children deemed by the assessing psychiatrist to be candidates for medication were offered short-term follow-up for medication initiation and then an opportunity to enter the study after 8 weeks on a stable dose.

All participating children completed the initial diagnostic assessment (baseline) followed by a wait period (within-subject waitlist control condition) before the onset of treatment. The wait period ranged from 11 to 44 weeks (M = 18 weeks; SD = 9.61 weeks) and time in treatment ranged from 11 to 23 weeks (M = 17.63 weeks, SD = 2.97 weeks). Language competence measures (PPVT and EVT) and the direct speaking measure (SNAP) were administered by a trained graduate student research assistant during the wait period for treatment. Children were assessed before the first treatment session (Time 1) and immediately following treatment (Time 2). Participants were reassessed 3 months (Time 3) and one year (Time 4) following treatment completion. Given the focus of PCIT on reducing opportunities for non-speaking behavior and allowing time for warm up in new situations, the SNAP was not re-administered immediately pre-treatment in order to reduce the likelihood that a child could engage in speaking avoidance in the therapeutic setting.

The 5 assessment points and measures completed are summarized in Fig. 1. The main outcome measure was parent-reported SMQ scores, which assesses frequency of speaking behavior over the past 2 weeks in a variety of settings. This was supplemented by teacher-reported speaking behavior (SSQ), a direct speaking assessment task in a public setting (SNAP), and parent-reported anxiety symptoms (SCARED).

2.5. Data analytic plan

Prior to analyses, data were examined for missing values, normality and outliers. Missing values on all main measures were minimal; scales were prorated in case of missing values. Due to researcher error, there was one missing data point at baseline for each of the SMQ, ADIS and SM-BOT. Six participants did not have teacher-reported selective mutism data (SSQ) at baseline due to time of year; pre-post analyses were conducted for this measure using a paired-samples t-test. Main analyses were one-way repeated measures ANOVAs to examine change in symptoms over 5 time points, with medication as a between-subject factor when examining differences for children on and off medication. Where violations of assumptions were present, appropriate transformations were applied and are reported in relevant sections below. Predictors of treatment outcome were analyzed using regression. All tests are 2-tailed. Data were analyzed using SPSS version 25.

3. Results

Table 1 shows scores across time-points for parent- and teacher-reported SM symptoms, parent-reported child anxiety symptoms, and direct speaking assessment of the child.

3.1. Diagnostic status

Participating children had an average of 2.30 psychiatric diagnoses (Range: 1–5; *SD* = 1.18), according to the ADIS interview with parents, including: SM (100%), Social Anxiety Disorder (63.3%), Separation Anxiety Disorder (35.5%), Specific Phobia (20.0%), and Generalized Anxiety Disorder (10%). Mean SM severity on the ADIS was 6.48 (Range: 4.5–8; *SD* = 1.23). On the SCARED, 61% were above screening cutoff on total score; and subscales were as follows: 100% (social anxiety); 61% (separation anxiety); 32% (generalized anxiety); 18% (panic/somatic symptoms); 30% (school avoidance). In the baseline SM-BOT, 16.7% had no interaction with the examiner, 26.7% had only non-verbal interaction, and 56.7% had some verbal interaction. Almost half of parents (46.7%) reported that their children *never* spoke in community settings.

3.2. Language competence

Mean receptive language scores at baseline were in the average range, *M* = 101.94, *SD* = 11.25; Range: 82-126. Similarly, one-word expressive vocabulary (using parents as test presenters) was also in the average range, *M* = 96.74, *SD* = 11.49; Range: 75-122.

3.3. Medication

The majority of children (61.3%) were not on medication at any point. At baseline, 2 children entered the study on fluoxetine (6.4%) and ten children (32.2%) were initiated on fluoxetine at baseline. In total, 12 children were on medication at the start of treatment (38.7%) and 10 (33.3%) remained on medication post-treatment. There were minimal changes during treatment with average fluoxetine dosage at pre-treatment of 6.42 mg (Range: 3–10 mg; *SD* = 2.97 mg) and post-treatment of 6.95 mg (Range: 4–10 mg; *SD* = 2.14 mg). Eight children (29.0%) remaining on medication at 3-month follow-up (M_{dose} = 7.2 mg, Range: 4–15 mg; *SD* = 3.3 mg) and six (20.7%) at 1 year follow up (M_{dose} = 10.43 mg, Range: 6–20 mg; *SD* = 5.22 mg).

3.4. Treatment outcome analyses

3.4.1. Parent-reported selective mutism symptoms

To examine differences in SM symptoms over time, a repeated-measures one-way ANOVA was conducted with SMQ mean scores as the dependent variable and all 5 time points (baseline, pre-treatment, post-treatment, 3-month follow up, 1-year follow up) as the repeated measure. Where sphericity assumption violations were evident, Greenhouse-Geisser-corrected degrees of freedom are reported below. Post-hoc analyses are Bonferroni-corrected given multiple comparisons.

A significant change in SMQ scores was found over time, $F(2.38, 61.82) = 72.23$, $p < .001$, $\eta 2_{partial} = .74$. While baseline and pre-treatment scores did not differ ($M_{diff} = 0.11$, $p = 1.0$), comparisons between pre-treatment and post-treatment scores indicated significant improvement ($M_{diff} = 1.02$, $p < .001$; Cohen's $d = 1.80$). Comparisons of pretreatment and follow-up SMQ scores revealed that these significant improvements were maintained at 3-month ($M_{diff} = 1.03$ $p < .001$), and 1 year follow ups ($M_{diff} = 1.04$, $p < .001$). Post-treatment and follow-up SMQ scores did not differ from each other.

A second repeated measures ANOVA was conducted with medication status at pre-treatment as a between subject factor ($n = 10$ on medication, $n = 17$ not on medication); see Fig. 2. The within-subject effect of time remained significant, $F(2.33, 58.18) = 65.71$, $p < .001$, $\eta 2_{partial} = 0.72$, whereas the time by medication interaction was not significant, $F(2.33, 58.18) = 0.65$, $p = 0.55$, $\eta 2_{partial} = 0.03$. The main (between subjects) effect of medication status was also non-significant, $F(1, 25) = 2.07$, $p = 0.16$, $\eta 2_{partial} = 0.08$. Therefore, medication status did not moderate treatment outcome and is not included in any subsequent analyses.

We also examined subscales of the SMQ to determine if treatment was associated with improvements in all parent-reported domains: school, community, and home/family, as well as distress/interference.

For school speaking, within-subject effect of time was significant, $F(2.24, 53.78) = 57.17$, $p < .001$, $\eta 2_{partial} = 0.70$. Comparisons between baseline and pre-treatment school scores revealed no differences ($M_{diff} = 0.17$, $p = .21$) whereas significant improvements were found in school scores between pre-treatment and post-treatment ($M_{diff} = 1.03$, $p < .001$; Cohen's $d = 1.47$), with gains consistently maintained at 3-month ($M_{diff} = 1.17$, $p < .001$) and 1-year follow ups ($M_{diff} = 1.26$, $p < .001$).

For community speaking, the within-subject effect of time was significant, $F(2.52, 65.63) = 63.00$, $p < .001$, $\eta 2_{partial} = 0.71$. Post-hoc analyses revealed no difference between baseline and pre-treatment community scores ($M_{diff} = 0.16$, $p = .21$) but significant improvement between pre-treatment and post-treatment ($M_{diff} = 1.40$, $p < .001$;

Table 1
Mean (SD) scores on SMQ, SCARED, SSQ and SNAP measures across time points.

Measure	Baseline	Pre-Treatment	Post-treatment	3-month follow up	1-year follow up
Parent Report					
SMQ total	.96 (.44)	1.08 (.46)	2.03 (.59)	2.13 (.61)	2.13 (.53)
School	.67 (.56)	.83 (.64)	1.81 (.74)	2.06 (.74)	2.10 (.57)
Home/family	1.88 (.67)	1.88 (.53)	2.49 (.47)	2.54 (.45)	2.59 (.42)
Social/community	.26 (.34)	.47 (.56)	1.77 (.78)	1.75 (.87)	1.63 (.84)
Interference	2.21 (.45)	2.13 (.53)	1.35 (.76)	1.17 (.85)	1.02 (.74)
SSQ total	0.61 (.56)	0.70 (.63)	1.43 (.73)	1.52 (.65)	N/A
SCARED	30.84 (14.63)	29.05 (15.16)	21.45 (14.32)	19.74 (11.64)	19.15 (12.72)
Panic subscale	3.27 (3.91)	3.00 (3.96)	2.13 (2.52)	1.72 (2.70)	1.56 (2.24)
GAD	6.53 (5.09)	7.05 (5.09)	4.87 (4.78)	4.36 (4.24)	4.85 (4.70)
Sep. Anxiety	6.54 (4.15)	5.50 (4.13)	4.73 (4.16)	4.43 (3.17)	3.41 (3.58)
Social Anxiety	12.57 (1.69)	12.00 (2.55)	9.07 (3.38)	8.79 (3.48)	8.56 (3.61)
School Avoidance	1.73 (1.99)	1.74 (2.51)	0.67 (1.60)	0.43 (1.07)	0.78 (1.89)
SNAP # of words	6.00 (18.24)	N/A	59.76 (66.25)	56.00 (52.36)	N/A

Note: Sample sizes across time-points as follows: Selective Mutism Questionnaire (SMQ): $n = 30, 31, 30, 28, 28$; Screen for Child Anxiety Related Emotional Disorders –Parent Version (SCARED): $n = 28, 29, 30, 28, 27$; School Speech Questionnaire (SSQ): $n = 24, 26, 29, 20$; Strong Narrative Assessment Procedure (SNAP): n = 31, 29, 25.

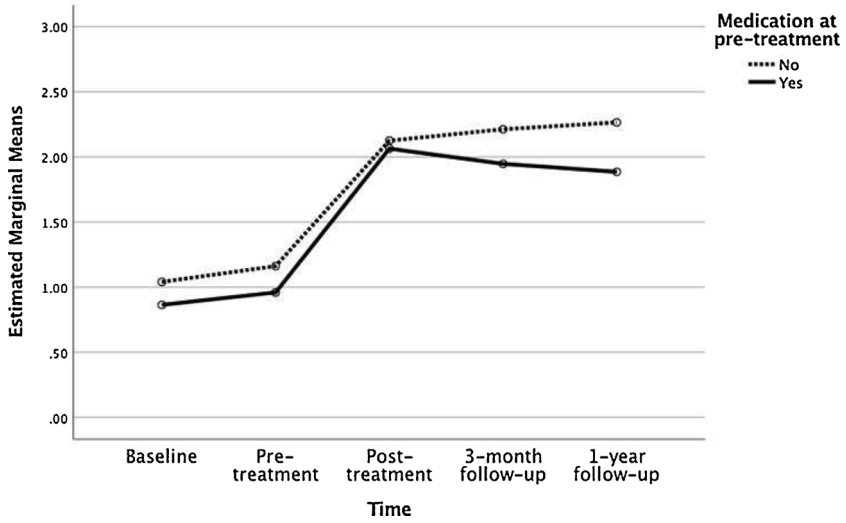

Fig. 2. Estimated marginal means of SMQ mean scores across time-points for children on and off medication at the start of treatment.

Cohen's d = 1.77), with gains consistently maintained at 3-month (M_{diff} = 1.28, p < .001) and 1-year follow up (M_{diff} = 1.18, p < .001).

In home and family domains, the within-subject effect of time was significant, $F(2.68, 69.67) = 21.22$, p < .001, $\eta 2_{partial} = 0.45$. Comparisons revealed no difference between baseline and pre-treatment scores on the home subscale (M_{diff} = 0.00, p = 1.00) but significant improvement from pre-treatment and post-treatment (M_{diff} = 0.63, p < .001; Cohen's d = 1.09), with gains consistently maintained at 3-month (M_{diff} = 0.61, p < .001) and 1-year follow up scores (M_{diff} = 0.67, p < .001).

For interference/distress domain, the within-subject effect of time was significant, $F(4, 104) = 35.69$, p < .001, $\eta 2_{partial} = 0.58$. Post-hoc analyses revealed no difference between baseline and pre-treatment interference scores (M_{diff} = 0.03, p = 1.00) but significant decreases in parent-reported interference/distress between pre-treatment scores and post-treatment (M_{diff} = 0.82, p < .001), 3-month follow-up (M_{diff} = 0.99, p < .001) and 1-year follow up scores (M_{diff} = 1.16, p < .001). There were no differences between any post-treatment timepoints (ps $\geq$ 0.15).

In short, all subscales showed the same pattern of results as the total scores with significant and large effects pre-post treatment in school, community, home/family, and interference, with gains consistently maintained at follow up (see Fig. 3).

3.4.2. Teacher-reported selective mutism symptoms

Teacher-reported data on children's school functioning is also reported. Due to challenges getting teacher data for all time points, we report pre-post treatment data for teacher data only. A paired-samples t-test revealed significant gains in speaking behavior from pre-treatment (M = 0.70) to post-treatment (M = 1.40), $t(25) = -6.70$, p < .001; Cohen's d = 1.07.

3.4.3. Direct assessment of child speaking behaviors

We also examined changes in speaking behaviors with a stranger (SNAP). Data at all time points were positively skewed; a cube root transformation was performed. Resulting residuals were normal at post-

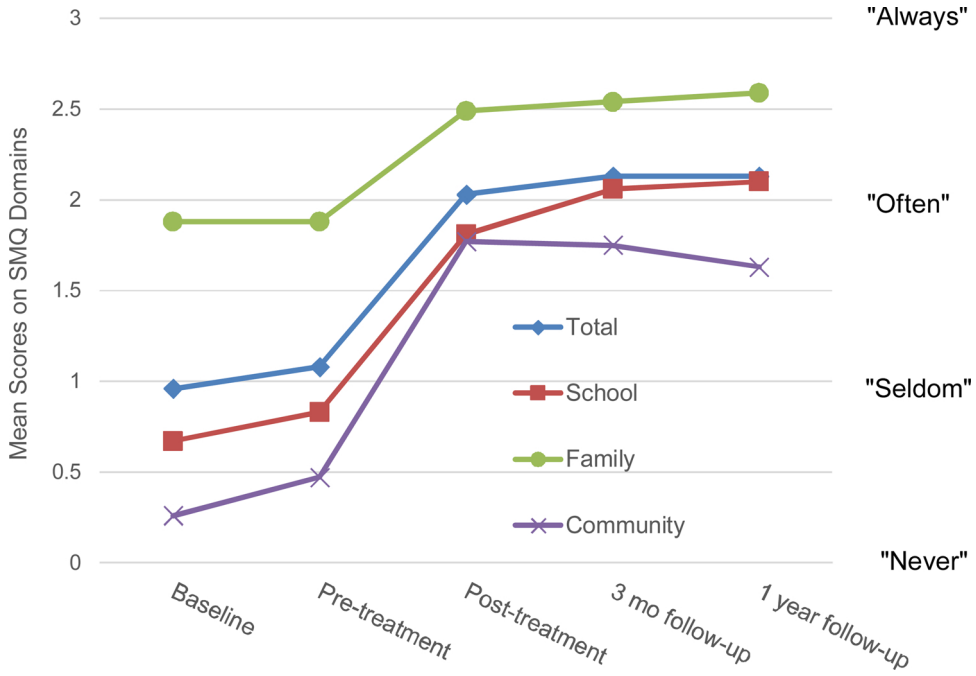

Fig. 3. SMQ scores by domain and time.

treatment and follow-up but remained positively skewed at baseline. There was a significant effect of time on number of words spoken, $F(1.52, 36.44) = 47.01$, $p < .001$, $\eta 2_{partial} = 0.66$. Post-hoc tests revealed a significant difference between number of words spoken at baseline and both post-treatment ($\sqrt[3]{M_{diff}} = 2.78$, $p < .001$; Cohen's $d = 1.09$) and 3 month follow up ($\sqrt[3]{M_{diff}} = 2.36$, $p < .001$) but no difference between post-treatment and 3 month follow up ($\sqrt[3]{M_{diff}} = 0.42$, $p = .15$).

3.4.4. Parent-reported anxiety outcomes

We examined differences in anxiety scores on the parent-reported SCARED. Violation of normality was present; all timepoints were transformed using square root transformation. Results show a significant difference in SCARED scores over time, $F(4, 92) = 18.40$ $p < .001$, $\eta 2_{partial} = 0.44$. Comparisons revealed that baseline and pre-treatment scores were equivalent ($\sqrt{M_{diff}} = 0.13$, $p = 1.0$). However, there were significant improvements in post-treatment scores ($\sqrt{M_{diff}} = 0.97$, $p = 0.001$), 3 month follow up scores ($\sqrt{M_{diff}} = 0.95$, $p = .002$) and 1 year follow up scores ($\sqrt{M_{diff}} = 1.17$, $p < .001$) compared with pre-treatment scores. None of the post-treatment time points differed significantly from each other (*p*s = 1.00).[3] At one-year follow up, the proportions of children above threshold for a possible anxiety disorder were as follows: total score (26%); panic/somatic symptoms (3.7%) ; GAD (18.5%); separation anxiety (25.9%); social anxiety (63%); school avoidance (18.5%).

3.4.5. Treatment satisfaction

Parent satisfaction with treatment was very high with a mean satisfaction rating of 3.86 (*SD* = .25) out of a possible 4 (*n* = 29).

3.5. Predictors of treatment outcome

A regression analysis with SMQ post-treatment scores as the dependent variable was conducted with age and language competence as predictors, controlling in the first step for pre-treatment SMQ scores. At step 1, the model was significant, $F(1, 28) = 4.83$, $p = .04$. Pre-treatment scores explained a significant proportion of the variance, $R^2\Delta = 0.15$. At step 2, introducing age, PPVT and EVT scores did not result in a significant increase in proportion of variance explained, $R^2\Delta = 0.05$, and the model was not significant $F(4, 25) = 1.58$, $p = 0.21$. These results indicate that neither age nor language competence affected children's response to behavioral treatment. Table 2 shows the results from the regression analysis.

Table 2
Regression Analysis on Predictors of Treatment Outcome.

	B	S.E.(B)	β	*t*	*p*
Step 1					
SMQ pre-treatment scores	0.53	0.24	0.38	2.20	0.04
Step 2					
SMQ pre-treatment scores	0.46	0.25	0.33	1.82	0.08
PPVT standard scores	−0.003	0.013	−0.05	−0.20	0.85
EVT standard scores	−0.007	0.01	−0.15	−0.62	0.54
Age	0.08	0.07	0.23	1.15	0.26

3.6. Response to treatment

Parent ratings for amount of speech across contexts as measured by the SMQ indicated that 67% of children were rated as speaking "often" or more on average across contexts following treatment. Examining the amount of change in speaking behaviors from pre- to post-treatment revealed four children (18%) who were clear non-responders with mean changes in SMQ scores near 0 (Range: −0.28 to 0.06 points); 8 children (27.6%) who were 'modest responders' with mean changes of 0.41 – 0.85 points and 17 "robust responders" (58.6%) who had changes of a point or higher (Range: 0.98–2.12).

4. Discussion

This study adds to growing evidence that behaviorally-based treatments for children with SM are effective and well tolerated, and additionally provides the first specific evidence for weekly-delivered PCIT-SM. This suggests that the framework of coaching parents and others on the specific CDI and VDI skills to minimize continued negative reinforcement of non-responding and maximize successful speaking are an important addition to the existing behavioral literature on gradual exposure and contingency management. In line with other modifications to PCIT for anxiety disorders (e.g., Choate et al., 2005) it gives parents a central 'therapeutic' role, empowered by specific and teachable skills, to help their child overcome their anxiety disorder. In addition to PCIT modifications for anxiety, there are other recent therapies, such as that developed by Lebowitz and colleagues (Lebowitz, Omer, Hermes, & Scahill, 2014) in which parents are the exclusive focus of treatment and source of change in their child's anxiety disorder through unilateral reductions in accommodations and other management strategies. While these approaches are not theoretically inconsistent with more child-focused behavioural and cognitive behavioural approaches, they nonetheless represent an important theoretical shift towards parents' central importance in their child's development and wellbeing.

In particular, PCIT-SM was associated with robust and significant decreases in symptom severity and impairment that occurred across domains and were well-maintained at 3-month and 1-year follow-up. Pre-post treatment effect sizes were very large – Cohen's *d*s were in the range of 1.1 (teacher report) to 1.8 (parent report across contexts). This suggests that PCIT-SM has a meaningful and substantial effect on children's ability to speak in more settings. These results are particularly noteworthy as over 30% of children had received previous treatment for SM, yet they remained significantly impaired before PCIT-SM treatment.

Parents reported high satisfaction with treatment and all families completed treatment. In addition to significantly improved functioning on parent- and teacher-rated measures, children also demonstrated improvements in a task requiring them to retell a story to a stranger in the office setting (Cohen's $d = 1.1$). This task reflects more complex aspects of language recall and formulation with a new individual. Including this behavioral outcome measure strengthens the results of the parent and teacher-reported outcomes, and provides a high bar to evaluate children's ability to speak to a novel adult under challenging conditions.

This study is the first to show that PCIT-SM is an effective treatment for children with SM, with effect sizes similar to those seen in Bergman's study (Bergman et al., 2013), and somewhat more robust than those seen in a school-based treatment study (Oerbeck et al., 2015). It also adds to these studies in providing clear support for the effectiveness of active, behaviorally-based interventions for SM used in a variety of school and/or community settings. Practice in these settings can be facilitated either directly via clinician or parent-led exposure, or through assigned tasks and coordination, or both.

Of note, 100% of children spoke to the therapist in the first session. This finding is noteworthy given that our sample of children had similar SMQ scores at baseline to those reported in other studies of SM (e.g., Bergman et al., 2008, 2013; Oerbeck et al., 2015) with almost half (46.7%) of parents reporting that their child *never* spoke in public or community settings, and all participants rated as moderately to severely impaired by SM on the ADIS clinician severity rating. Although sample differences between this study and previous studies cannot be ruled out

[3] See Table 1 for raw data of mean scores across time points.

as contributory to this finding, techniques that may have contributed include the use of CDI involving warm up time without speech demands, rewards for speaking, and when necessary, graduated introduction of the novel adult. A recent study (Bunnell, Mesa, & Beidel, 2018) specifically examined a two-session speaking hierarchy under 3 conditions and found that all children with SM spoke within an hour. Taken together, these findings suggest that children with SM are often able to speak to a new adult when an appropriate, structured paradigm is used. Some approaches to SM treatment focus on several sessions of warm-up before attempting to elicit speech (e.g., Bergman et al., 2013). There may be some efficiency gains in effectively targeting speech early in treatment which may decrease overall treatment duration. The present protocol was 16 sessions in length, which is approximately 20% shorter than those in the two published RCTs for SM. Moreover, explicitly coaching parents may have helped to maximize parents' ability to successfully complete out-of-session practice, which is key to generalizing speaking gains to non-clinic settings.

PCIT-SM also had an indirect benefit on general anxiety symptoms despite the highly specific nature of the therapy (without many of the traditional components of a cognitive behavioral program for anxiety). At follow up, children were on average 6 points below the screening cutoff for a possible anxiety disorder on the SCARED, whereas they were on average 5 points above the cutoff at baseline. Moreover, the proportion of children above threshold decreased from 61% to 26%. All anxiety subscales were, on average, below cutoff at follow up with the exception of social anxiety which was just above threshold. Whereas 100% of children were above threshold on social anxiety subscale at baseline, 61% were above threshold after treatment. Similarly, the proportion of children above threshold decreased substantially in all subdomains of the SCARED.

A strength of the present study is the diversity of the sample that included a broad range of ethnicity and parental education. The self-reported ethnicity and the percentage of participants speaking a first language other than English (32% of children and 50% of parents) roughly mirror Vancouver demographic data (Statistics Canada, 2017), where the treatment took place. Previous studies have suggested that children of immigrant families are at increased risk for SM (Elizur & Perednik, 2003). Thus, including participants from diverse communities is important.

No significant predictors of treatment response were identified. While previous studies have suggested that younger children respond better, our study did not find this. Similarly, language competence did not predict treatment response. Although our sample was small, these exploratory results may suggest that this treatment may be equally helpful for a broad range of presentations including older children who typically have had a longer duration of symptoms, as well as those for whom English language skills were not as strong. Further research including a larger sample of participants varying on these characteristics is needed, however, to determine if, in fact, this is the case.

Despite age and language competence not predicting treatment outcome, other variables that were not examined may have been influential. In our study, 13% of children were clear non-responders. Our clinical observations suggested that an examination of family factors (e.g., parental social anxiety; family functioning), as well as child factors (e.g., oppositionality) would be fruitful areas for future research.

Almost 40% of children were on a stable dose of fluoxetine prior to behavioral treatment. The study was not designed to look at medication effects and medication initiation was clinically driven, incorporating factors such as severity, age, previous treatment response, and parental preference. Medication status was not shown to predict treatment response, and the fact that this more impaired group did as well with treatment is encouraging. However, the relative contribution of medication is unknown and ripe for further study. Specifically, a RCT of medication versus PCIT-SM versus a combined condition would be helpful in clinical decision making for children with more severe presentations.

Although the present study yielded significant improvements for children with SM, their speaking frequency did not reach the threshold of children without SM. A previous study examined total SMQ scores for children with and without SM and found that children without SM averaged 46 points (vs 13 points for those with SM; Bergman et al., 2008). In the current study, mean SMQ scores one year post-treatment were 2.13, which translates to a total score of 36.2 points, or ten points lower than an average child without SM. Given the chronic and pervasive nature of SM, it will be important to continue searching for treatment optimization strategies to ensure that children are robustly treated, as SM has broad impacts on functioning.

4.1. Conclusion

We now have evidence that PCIT-SM is an effective treatment for SM. In terms of clinical decision-making, it may make sense to look at strengths within the system when developing a treatment plan as school personnel, therapists, and parents can all play an important role.

Strengths of the current study include the ethnically and economically diverse sample, the 'real-world' treatment setting high on external validity, and the multimodal assessments of parent and teacher report complemented by direct speaking tasks for children. Nonetheless, the current study is non-randomized which cannot exclude the possibility of maturational effects on children's gains. However, existing evidence points to the chronic nature of SM (Bergman et al., 2013), and the lack of change during the wait period suggests that this was unlikely to account for treatment gains. Furthermore, absent treatment dismantling studies, we are not able to understand the relative benefit of the treatment modules within PCIT-SM. As well, without a head-to-head comparison of PCIT-SM versus other behavioral approaches we cannot speak to the relative efficacy of each. Further research to replicate current findings and more robustly examine predictors of treatment outcome is needed.

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