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Integrated Behavior Therapy for Selective Mutism: A randomized controlled pilot study

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ABSTRACT

Objective: To evaluate the feasibility, acceptability, and preliminary efficacy of a novel behavioral intervention for reducing symptoms of selective mutism and increasing functional speech.
Method: A total of 21 children ages 4 to 8 with primary selective mutism were randomized to 24 weeks of Integrated Behavior Therapy for Selective Mutism (IBTSM) or a 12-week Waitlist control. Clinical outcomes were assessed using blind independent evaluators, parent-, and teacher-report, and an objective behavioral measure. Treatment recipients completed a three-month follow-up to assess durability of treatment gains.
Results: Data indicated increased functional speaking behavior post-treatment as rated by parents and teachers, with a high rate of treatment responders as rated by blind independent evaluators (75%). Conversely, children in the Waitlist comparison group did not experience significant improvements in speaking behaviors. Children who received IBTSM also demonstrated significant improvements in number of words spoken at school compared to baseline, however, significant group differences did not emerge. Treatment recipients also experienced significant reductions in social anxiety per parent, but not teacher, report. Clinical gains were maintained over 3 month follow-up.
Conclusion: IBTSM appears to be a promising new intervention that is efficacious in increasing functional speaking behaviors, feasible, and acceptable to parents and teachers.

Selective Mutism (SM) is a childhood behavioral disorder characterized by persistent failure to speak in specific social situations despite speaking in other situations. According to the Diagnostic and Statistical Manual of Mental Disorders, 4th edition (DSM–IV–TR; American Psychiatric Association, 2000), lack of speech must cause interference, last at least one month, and not be due to a lack of knowledge of the relevant language. SM is considered to be an impairing condition that can interfere with both educational achievement and socialization (e.g., Bergman, Piacentini, & McCracken, 2002; Carbone et al. 2010), with a typical onset age ranging from ages 3 to 5 (Cunningham, McHolm, Boyle, & Patel, 2004; Garcia, Freeman, Francis, Miller, & Leonard, 2004). While previously thought to be quite rare with rates as low as .18% (Kopp & Gillberg, 1997), more recent studies have revealed higher prevalence rates of approximately .71–.76% (Bergman et al., 2002; Elizur & Perednik, 2003).

Although SM has received increased attention in the last decade, there remains a dearth of knowledge regarding the phenomenology and treatment of the disorder. There is a general consensus that SM is closely related to social anxiety disorder, with an increasing conceptualization of SM as a developmental variant of social phobia (Bogels et al., 2010; Yeganeh, Beidel, Turner, Pina, & Silverman, 2003). Evidence to support the link between SM and social phobia is derived from multiple sources. For one, numerous studies report comorbidity rates approaching or greatly exceeding 50% (e.g., Alyanak et al., 2012; Arie et al., 2006; Manassis et al., 2007), with some co-occurrence rates greater than 80% (Dummit et al., 1997; Vecchio & Kearney, 2005). Additionally, several investigations have revealed that parents of children with SM have elevated rates of social phobia (Black & Uhde, 1995; Chavira, Shipon-Blum, Hitchcock, Cohan, & Stein, 2007). Further, evidence suggests that some treatments that are effective in reducing social anxiety are also efficacious for SM, such as certain pharmacological agents (Carlson, Mitchell, & Segool, 2008; Manassis & Tannock, 2008). Accordingly, it is reasonable to suspect that the benefits of other extant empirically supported treatments for childhood social phobia may extend to children with SM. Indeed, efforts to treat SM

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using modified manualized interventions initially developed for social anxiety (Fisak, Oliveros, & Ehrenreich, 2006) or more general forms of child anxiety (Hudson, Krain, & Kendall, 2001) have been somewhat successful. Similarly, modular CBT, which has shown promise as treatment for anxiety disorder in children, was used successfully to treat SM as reported in two recent case studies (Christon, Robinson, & Arnold, 2012; Reuther, Davis, Moree, & Matson, 2011).

SM presents unique challenges that must be addressed during treatment. Unfortunately, these critical aspects of treatment are not present in existing manualized child anxiety interventions or are, at best, tacked on as supplemental additions. Children with SM often fail to speak to the therapist in early sessions, which necessitates unique strategies for engagement and parental involvement early in the treatment process. Further, the typical age of onset for SM (age 5; Cunningham et al., 2004; Garcia et al., 2004) is considerably younger than those of other anxiety disorders, requiring developmental adaptations of commonly used CBT intervention (see Piacentini & Bergman, 2001). In addition, children with SM tend to be most symptomatic in the school environment (Bergman, Keller, Piacentini, & Bergman, 2008), thus requiring extensive treatment involvement of and coordination with school personnel, most notably, the child's teacher. As a result, current treatment approaches shown effective for childhood social phobia and other childhood anxiety disorders may not be sufficient for the treatment of SM.

With the exception of a small medication trial (Black & Uhde, 1994), there are no published randomized controlled treatment trials for children with SM to date. In fact, until quite recently, what little treatment research that did exist lacked scientific rigor (e.g., no comparison group, single subjects) among other methodological limitations (e.g., failure to identify diagnostic procedures, assessment or outcome methods, number of treatment sessions, or details of the treatment method; Viana, Biedel, & Rabian, 2009; Cohan, Chavira, & Stein, 2006). These shortcomings, along with the lack of controlled trials, make it difficult to assess treatment efficacy or to replicate described treatments. Despite these limitations, recent reviews of the literature indicate empirical support for individual behavioral intervention of SM (Cohan et al., 2006; Stone, Kratochwill, Sladezcek, & Serlin, 2002), and recent more methodologically sound studies using behavioral techniques show promising results (e.g., Oerbeck, Johansen, Lundahl, & Kristensen, 2012; Sharkey, McNicholas, Barry, Begley, & Ahern, 2008; Vecchio & Kearney, 2009).

The goals of the present study were to examine the feasibility, tolerability, and preliminary efficacy of a behavioral intervention developed for selective mutism using a randomized controlled methodology. Following a baseline assessment to determine eligibility, participants were randomly assigned to either 20 sessions of individual Integrated Behavior Therapy for Selective Mutism (IBTSM) or 12 weeks of waitlist (WL). We hypothesized that the active treatment condition would be feasible, tolerable, and associated with statistically significant decreases in symptoms compared to WL condition. To assess durability of gains, children randomized to IBTSM completed a follow-up assessment 3 months post-treatment. We anticipated that symptom improvement would be maintained over the follow-up period.

Method

Participants

Participants were recruited from a pediatric anxiety specialty clinic, mental health practitioner referrals, and postings on internet websites focused on selective mutism. Children were eligible for inclusion if they were ages 4–8 years, inclusive, at baseline and met.

DSM-IV criteria for a primary diagnosis of selective mutism (SM). Because a goal of this intervention was to integrate treatment within a functional context, children were required to be attending school or some other form of structured daily group activity (e.g., day camp during school breaks) continuously throughout their enrollment. Children were excluded from study entry if they had undergone treatment with psychotropic medication within 2–6 weeks of study entry (depending upon medication); b) had failed a trial of CBT for anxiety within the previous two years; c) met criteria for any psychiatric illness that contraindicated study participation, including prominent mood disorder, psychosis, or pervasive developmental disorder. Children were also excluded if they or their participating parent was unable to complete measures, interviews, or treatment in English.

Sixty-seven interested parents completed a structured telephone screen to assess initial eligibility. Twenty-five qualifying families completed informed consent/assent and the baseline eligibility evaluation. A total of 21 children with SM participated in the present study. The study consort diagram is presented in Fig. 1.

Study design and procedures

All study procedures were approved by the University Institutional Review Board. Children were randomly assigned to either 20 sessions of Integrated Behavior Therapy for Selective Mutism (IBTSM) or to a 12-week Waitlist (WL) using a randomization scheme generated by Random Allocation Software (Saghaei, 2004). Children in the IBTSM treatment condition received 20 sessions of manualized treatment over 24 weeks. Children assigned to WL were offered open IBTSM treatment at end of WL. To explore the durability of treatment gains, a 3-month post-treatment assessment was conducted for participants randomly assigned to the IBTSM condition (Week 36).

We employed a 12-week Waitlist (rather than a methodologically favorable matched 24-week period) due to ethical and clinical concerns associated with maintaining youths on an extended Waitlist of 24 weeks without treatment. This design adaptation has been utilized previously in the pediatric anxiety CBT literature (e.g., Kendall, 1990, 1994) and seems especially reasonable in the early stages of treatment development To account for the unmatched duration of IBTSM and Waitlist, assessments were completed by independent evaluators, blind to treatment condition, at baseline, week 12, and week 24 for all participants, regardless of group assignment (IBTSM or WL). This was done in order to a) maintain the same number of assessments across study conditions, b) preserve blindness of the independent evaluator, and c) allow for a direct comparison of outcomes between IBTSM and WL at the end of the WL condition (i.e., matched duration of 12 weeks following baseline). Of note, the primary comparison of interest to address our study goals occur at the end of randomized study condition (week 12 for WL and week 24 for IBTSM), as performed in previous treatment studies with similar designs (Kendall, 1990, 1994). For simplicity, comparison of these time points in the IBTSM and WL groups is hereafter referred to as *End of Condition*.

Measures

The Anxiety Disorders Interview Schedule for DSM-IV, Parent Version (ADIS-P; Silverman & Albano, 1996) was used to assess diagnostic status. The ADIS provides direct coverage of a broad range of anxiety, mood, and externalizing behavior disorders in youth. The ADIS has been described as the premier instrument for assessing anxiety disorders in youth (Wood, Piacentini, Bergman,

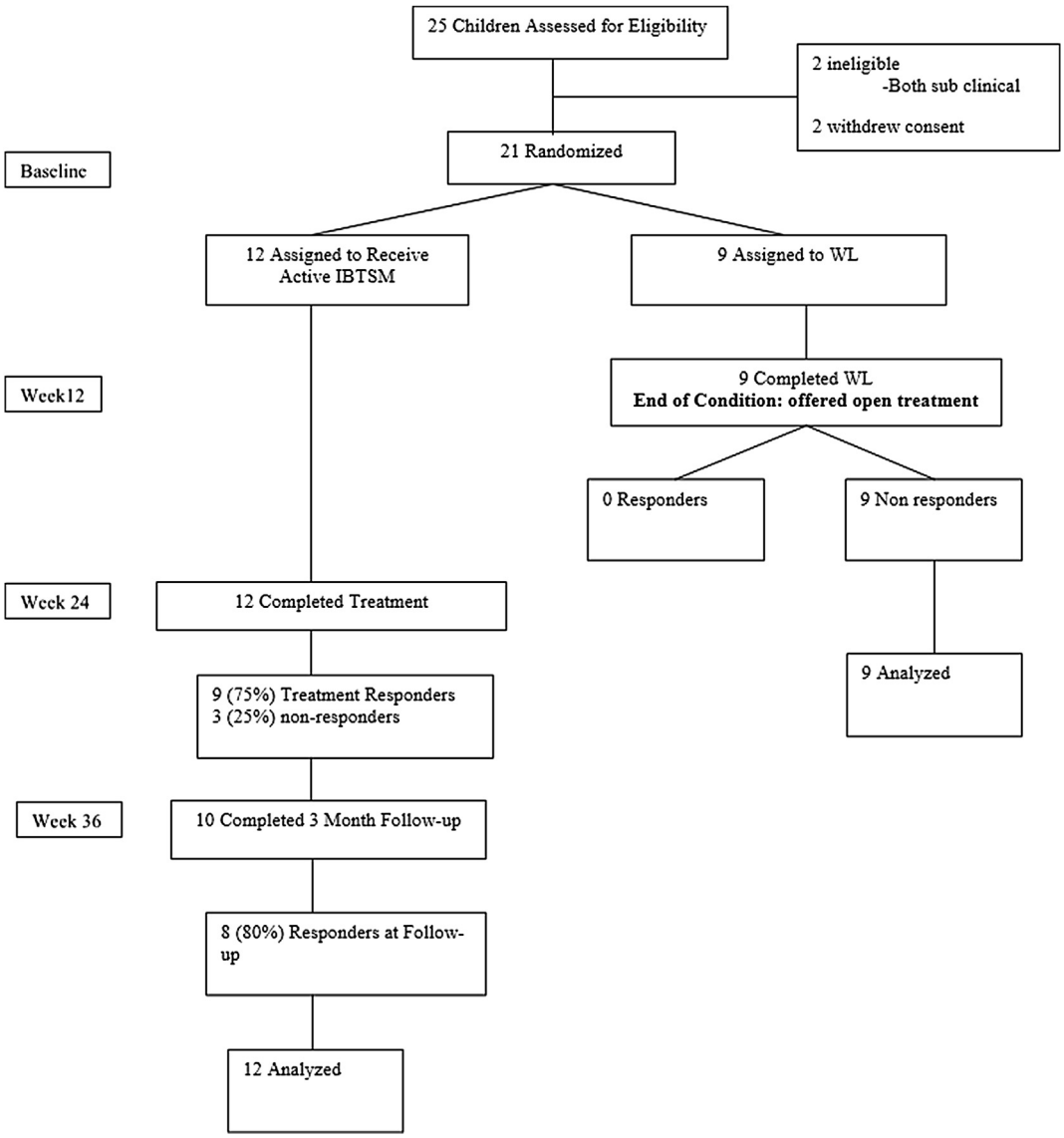

Fig. 1. Study enrollment and retention. Note: IBTSM = Integrated Behavior Therapy for Selective Mutism; WL = Waitlist.

McCracken, & Barrios, 2002). The interview has good reliability (Grills & Ollendick, 2003; Silverman & Nelles, 1988; Silverman, Saavadra, & Pena, 2001), and has shown sensitivity to treatment effects in studies of youth with anxiety disorders (e.g., Kendall et al., 1997; Walkup et al., 2008). Due to failure to speak in the clinic setting and young age of children, only the parent interview was administered. A clinical severity rating (CSR) of $\geq$4 on a 0–8 scale is indicative of clinically significant disorder and was required for an SM diagnosis. All assessments were administered by independent evaluators (IE) who were blind to treatment condition. Study IEs were graduate level clinicians who were previously trained in administering the ADIS. Each case was discussed with a diagnostic review team, consisting of at least one licensed child clinical psychologist experienced in the assessment of childhood anxiety disorders and selective mutism. The IE presented the case, including symptoms and severity ratings, and diagnostic consensus was reached. This procedure is similar to that employed by Wood et al. (2002).

The Clinical Global Impression - Severity (CGI-S) and Improvement (CGI-I) Scales (Guy & Bonato, 1970) were used to determine overall severity, improvement, and treatment Responder status. The CGI-S score provides a global rating of baseline severity ranging from 1 (Not at all Ill) to 7 (Extremely Ill) while the CGI-I provides a global rating of clinical improvement ranging from 1 (Very Much Improved) to 7 (Very Much Worse). The IE provided a CGI-S rating for each patient at baseline and both CGI-S and CGI-I ratings at each subsequent evaluation. Subjects who received a CGI-I rating of 1 (Very Much Improved) or 2 (Much Improved) at End of Condition were considered treatment Responders.

The Selective Mutism Questionnaire (SMQ; Bergman et al., 2008) is a 17-item parent- report measure of SM behaviors. The SMQ has three subscales (Home, School, Other) and parents rate the frequency of speech in these domains. SMQ items contain four possible responses (0 = Never, 1 = Seldom, 2 = Often, 3 = Always) which are averaged to obtain a Mean score. The SMQ has been used in several investigations focused on SM (e.g., Bar-Haim et al., 2004; Manassis et al., 2007; McInnes, Fung, Manassis, Fiksenbaum, & Tannock, 2004) and initial psychometric investigation supported the convergent and discriminant validity of the measure (Bergman et al., 2008; Letamendi et al., 2008). Importantly, the SMQ appears

to be sensitive to treatment related changes in symptoms as well (Bergman et al., 2008; Sharkey et al., 2008). Cronbach's alpha in this sample was $\alpha = .76$. The SMQ was administered at each assessment point and is a primary outcome measure.

An Independent Evaluator Behavioral Evaluation (IEBE) of structured verbal and non-verbal interactional tasks was completed by the IE with each child at each assessment point. The IEBE provided the opportunity for observation and interaction with the child and was developed to safeguard against potential biases inherent in sole reliance on parent report when assessing children's symptoms. The information gathered using the IEBE aided in the diagnostic process and allowed for structured systematic interactions across participants. Examples of non-verbal behaviors assessed included blowing bubbles, jumping up and down, and posing as an instant photo was taken. Verbal behaviors assessed included responses to a series of neutral questions (e.g., what is your brother/sister's name, what is your favorite color). Notably, at baseline, 62% of the children did not provide verbal responses to any of the questions. The IEBE contains 10 items with a total duration of 10–15 min. The data from the IEBE were not analyzed or used to measure treatment outcome, but the descriptive information provided was incorporated into diagnostic decisions and global ratings. The IEBE was developed for this study; further information regarding the measure is available upon request from the first author.

The School Speech Questionnaire (SSQ; Bergman, Keller, Wood, Piacentini, & McCracken, 2001) is a teacher-report measure of children's speaking behaviors at school. The measure contains six items that were modified from the original SMQ (Bergman et al., 2001). SSQ items contain a statement regarding speaking frequency with four possible responses (0 = Never, 1 = Seldom, 2 = Often, 3 = Always), with a lower score indicating higher severity and impairment. There are some data to suggest that the SSQ has utility for evaluating treatment-related teacher ratings of symptom improvement (Oerback et al., 2012). In the current sample, the SSQ demonstrated acceptable internal consistency with Cronbach's $\alpha = .76$. The SSQ was administered at all assessment points and was considered a primary outcome measure. For descriptive purposes, we added one item to the SSQ to assess school interference due to lack of speech. This item has four possible responses (0 = Not at All, 1 = Slightly, 2 = Moderately, 3 = Extremely) and was not included in the SSQ Mean score.

The Social Anxiety Scale for Children-Revised (SASC-P/T; Parent and Teacher versions) is an 18-item questionnaire for social anxiety that has demonstrated reliability and validity (LaGreca & Stone, 1993). A higher score indicates higher severity. In this study, the SASC was completed by parents and teachers. As there were no existing teacher-rated social anxiety measures, following consultation with the developer (A. M. LaGreca, personal communication, 1999), item wording on the parent version of the Social Anxiety Scales for Children-Revised (SASC-R; LaGreca & Stone, 1993) was modified by the Principal Investigator for use with teachers (e.g., "my student" instead of "my child"). The internal consistency of the teacher adaptation of the SASC-R was excellent in a previous school-based sample ($\alpha = .91$; Bergman et al., 2002). In this sample, Cronbach's alpha was $\alpha = .87$ for parent report and $\alpha = .91$ for teacher report. The SASC-R was completed by parents and teachers at all assessment points.

The Strong Narrative Assessment Procedure – Retell (SNAP; Strong, 1998) is a standardized narrative elicitation task based on audio-recorded stimulus stories that accompany wordless picture books. The assessment procedure involves asking the child to look at the wordless storybook while listening to the tape and then retell the story. The SNAP was adapted by McInnes et al. (2004) to assess narrative language abilities among children with SM. Their investigation revealed that greater severity of SM was associated with significantly shorter responses (i.e., fewer words) on the SNAP narrative task. The present study used three stories for school assessments with the teacher. A study staff member provided instructions for each teacher on how to administer the instructions for the child, and only the child and teacher were present for the administration at school. The order of the stories was randomly assigned to each child at study entry. The child's retelling of the story was audio recorded, transcribed, and analyzed for length in accord with procedures established by McInnes et al. (2004). The SNAP was administered by teachers at all assessment points.

The Client Satisfaction Questionnaire (CSQ; Hargreaves & Attkisson, 1978) is an 8-item global measure of client satisfaction with therapy services and was used to assess parent and teacher satisfaction with IBTSM. Each item contains four response options (1 through 4), with higher values indicating greater satisfaction. The CSQ has demonstrated strong psychometric properties, including excellent internal consistency ($\alpha = .91$) and meaningful linkages to service utilization and therapy outcome (e.g., Attkisson & Zwick, 1982). In this study, items were modified for administration with parents and teachers (e.g., "your child" vs. "your student").

Integrated Behavior Therapy for Selective Mutism (IBTSM)

Overview

Therapy sessions for IBTSM were administered according to a detailed treatment manual that has since been adapted for publication (Bergman, 2013). Individual treatment consisted of 20 1-h sessions held over a 24-week period. The last two sessions occurred every other week to provide additional time for practice of skills between sessions and to facilitate the transfer-of-control process. The behavioral intervention focused on graduated exposure to the feared stimuli/situation (e.g., verbal communication) as the primary agent of symptom reduction. Although behavioral exposure exercises were routinely conducted in session, an equal emphasis was placed on assigning behavioral practices to occur outside of session in situations that were central to the non-speaking behavior (e.g., at school). Due to the non-speaking nature of SM and the developmental level of child participants, parent involvement was incorporated into all components of treatment. Similarly, as described below, treatment participation from the child's teacher was a critical component of the intervention. Note that in a few cases, particularly during the summer months, the role of the teacher was fulfilled by a day camp instructor, etc.; for simplicity, we use the term "teacher" to describe all instructors. An outline of the intervention is provided below:

Before first session

Teachers were contacted (with parental consent), and the behavioral treatment plan, including projected teacher participation, was described. Children did not begin treatment until communication with the teacher was established.

Sessions 1–3

The goals of the first few sessions were to a) orient the child and parents to the intervention, b) build therapist rapport with the child (e.g., playing non-verbal games) and increase child's level of comfort in the clinic setting, and c) develop a behavioral reward system. During these sessions, the child's tolerance of speaking in the session was assessed and information regarding the presence of the child's speech in various situations was ascertained. The use of a developmentally modified "Feelings Thermometer" was introduced as way for the child to communicate anxiety levels. At this phase, there was an emphasis on increasing the child's speech with the therapist to enhance communication and, more critically, to

provide a model for the additional work to be done with the child in other settings (e.g., talking to teachers at school, with extended family). Behavioral intervention during this phase varied and depended upon the child's baseline level of speech with the therapist. Some examples of exposure activities conducted at this stage include: the child speaking to the parent in the therapy room without the therapist present, the child playing a verbal game with the parent in the therapist's presence, nonsense or animal sounds with the therapist, and the parent and child recording the child's voice and playing back for the therapist. A reward system was developed with each family to encourage child engagement and praise efforts.

Sessions 4–14

The majority of treatment focused on the implementation of individualized in-session behavioral exposure exercises and to planning out-of-session behavioral practice assignments (e.g., therapist, parent and child together selecting a classmate and plan to arrange a play date). In general, young children tend to have limited understanding of the rationale behind exposure exercises and are often unable to tolerate significant distress related to exposures. For these reasons, difficulty of assignments increased quite gradually, according to a hierarchy. Initially, the therapist was primarily responsible for developing the parameters of the exposure exercises; collaboration of all involved parties (parent, teacher, child) was emphasized increasingly over time. Parents were asked to keep a log for recording specific details regarding exposure attempts and outcomes. Reinforcement for attempts to complete assignments was consistent throughout all interventions. When appropriate to the developmental level of the child, selected cognitive restructuring principles were introduced (e.g., replacing fearful or worried thoughts with coping self-statements as recommended for young children; Rapee, Wignal, Hudson, & Schniering, 2000).

Sessions 15–20

The goals of later sessions were to begin transfer of control from the therapist to the parent and to discuss relapse prevention. As treatment progressed, the parent was instructed to begin praising the child for speaking behaviors as they occurred in daily life. To facilitate transfer of control (Silverman & Kurtines, 1996), an increased effort was made to develop parents' mastery of treatment principles and capacity to continue elements of treatment beyond the final session. Specifically, instead of the therapist leading the process, parents took the lead in devising and assigning speaking tasks, communicating with school personnel, setting goals, managing behavioral reward system, etc. The therapist provided feedback regarding the execution of these tasks.

School involvement

Given that many children with SM experience severe impairment in the school setting, a primary goal of this intervention was to improve functioning in the educational environment. Accordingly, behavioral exposures in the school setting and teacher involvement were central components of IBTSM. Examples of school behavioral tasks, which were initially developed by the therapist in consultation with the teacher, include: the child speaking in non-classroom areas of the school (e.g., playground), speaking to the parent with teacher's back turned, speaking to a single child in otherwise empty classroom, whispering to the teacher. Therapists remained in communication with individual teachers throughout treatment to ensure relevance of behavioral exposures and treatment goals. Parents also served to facilitate communication by passing written information regarding behavioral assignments between therapist and teacher. In addition, a school assessment coordinator was employed by the study team to help facilitate communication between the study team and each child's school. The school assessment coordinator met with the teacher at school at each assessment point to oversee administration of the SNAP and to problem solve any difficulties arising in the implementation of treatment or completion of assessment measures.

Therapist training, supervision, and fidelity

Therapists were Ph.D.- or Master's-level clinical psychology trainees. Clinical supervision was provided by the Principal Investigator. All study therapists had prior experience in the delivery of child evidence-based treatments. Accordingly, group and individual training of therapists emphasized familiarization with the treatment manual followed by approximately four hours of extensive discussion and role-play of all treatment procedures. Clinical supervision was provided to therapists in a group format where therapists discussed sessions from the previous week and implementation of therapy for the following week. All therapy sessions were videotaped and 10% ($N = 24$) of videotapes from the IBTSM condition were randomly selected and rated for adherence to the treatment manual and overall session quality (1–10 scale) by experienced CBT clinicians. Ratings indicated excellent treatment adherence (Mean = 99.3%) and excellent overall quality (Mean = 9.79, SD = .51).

Results

Data analytic plan

Prior to analyses, data were screened to test statistical assumptions (e.g., normality). Standardized z-scores on all continuous data were examined, and a criterion of $z \geq \pm 3.0$ was used to identify outliers. One child had a z-score of $z = 4.56$ on the SNAP at baseline and was therefore not included in analyses of the SNAP. Simple between-group comparisons were conducted using χ^2 tests for categorical measures and t-tests for continuous measures. Treatment effects were analyzed using 2 (Group: IBTSM vs. WL) × 2 (Time: Baseline vs. End of Condition) mixed factorial ANOVAs. End of Condition data points corresponded to week 12 (post-waitlist) for children in the WL group and Week 24 (post-treatment) for children in the IBTSM group. If significant main effects and interactions were found, only interaction effects are interpreted. For significant Group × Time interactions, *post hoc* within-subjects ANOVAs were performed in order to compare scores from baseline to the end of condition (treatment or WL). In addition to End of Condition analyses, treatment response at week 12 (end of condition for WL group and mid-treatment for IBTSM group) was explored in order to provide time-matched group comparisons and to assess whether children receiving IBTSM made significant mid-treatment gains. Finally, t-tests were performed to explore outcomes at week 36 (3-month follow-up) compared to baseline and week 24 (End of Condition) for children randomized to the IBTSM condition. Due to the preliminary nature of our investigation, no corrections were made for multiple comparisons.

Feasibility and acceptability

Twenty-five children were invited to participate in a full assessment of eligibility. Two children did not meet eligibility criteria and two declined to proceed with participation prior to randomization. Of the 21 children who were randomized, all completed their End of Condition assessments and there was no attrition in either the IBTSM or WL conditions. For children who

Table 1
Demographic and clinical characteristics of children.

	Total N/Mean (SD)	IBTSM n/mean (SD)	Waitlist n/mean (SD)	P
N	21	12	9	
Age at baseline (years)	5.43 (1.16)	5.25 (1.14)	5.67 (1.22)	.25
Gender (male)	11	7	4	.53
Ethnicity				.31
Non-Hispanic White	9	5	4	
Latino	2	1	1	
Asian	4	4	0	
Biracial	4	1	3	
Other	2	1	1	
Age of onset of SM (years)	3.38 (.74)	3.17 (.49)	3.69 (.96)	.13
CSR of SM	5.00 (.77)	5.00 (.74)	5.00 (.87)	1.00
No. of current diagnoses	2.38 (.59)	2.25 (.62)	2.56 (.53)	.25
Symptom measures				
SMQ	.86 (.40)	.79 (.36)	.95 (.46)	.38
SSQ	.70 (.55)	.81 (.59)	.56 (.49)	.31
SNAP − Retell	27.05 (73.64	12.18 (26.85)	45.22 (106.31)	.33
SASC − Parent	3.12 (.58)	3.29 (.49)	2.91 (.65)	.14
SASC − Teacher	2.26 (.75)	2.09 (.34)	2.43 (.85)	.35

SM = Selective Mutism; CSR = Clinician Severity Rating on the Anxiety Disorders Interview Schedule; SMQ = Selective Mutism Questionnaire; SSQ = School Speech Questionnaire; SNAP = Strong Narrative Assessment Procedure; SASC = Social Anxiety Scale for Children.

completed IBTSM, parents ($n = 12$) and teachers ($n = 11$) provided satisfaction ratings regarding the intervention at week 24. On the CSQ, parents reported a Mean Satisfaction rating of 3.79/4 (SD = .29) and teachers a Mean Satisfaction rating of 3.72/4 (SD = .47), indicating high levels of satisfaction for both groups.

Baseline characteristics and group comparability

Mean scores and frequencies at baseline, as well as demographic characteristics, are reported in Table 1. The IBTSM and WL groups did not differ at baseline on any demographic characteristics, including age, gender, or ethnicity (all $ps < .05$). In addition, there were no significant group differences on any baseline clinical characteristics, including clinician-rated severity, parent- and teacher-reported speaking behaviors, or SNAP retell scores. Both groups evidenced significant symptom levels at baseline. Teacher reports of child speaking behavior were consistent with parent reports of moderate to severe levels of symptoms and impairment. On the SSQ, teachers reported a Mean response of .70 (SD = .55) and Median response of .86, indicating that for the majority of children in this sample, teachers reported an average rating between "Never" and "Seldom" with regard to speech frequency in a variety of in-school situations. Notably, all children in this study had a teacher response of 0 = Never to at least one situation in the school setting. Further, on average, teachers reported the child's lack of speech in the school setting as "Moderately" to "Extremely" interfering at baseline ($M = 2.29/3$, SD = .78).

End of condition outcomes

Responder status

Analyses using the blind IE ratings on the CGI-I revealed a significantly higher response rate at End of Condition for children who received IBTSM (week 24) compared to those who were assigned to WL (week 12) (75% vs. 0%; $\chi^2(1) = 11.81$, $p = .001$). Similarly, 67% of children who received IBTSM no longer met criteria for SM based on results of ADIS interview, while all children assigned to WL continued to meet criteria for SM at week 12, $\chi^2(1) = 9.69$, $p = .002$.

Parent report

Means and standard deviations on parent measures are reported in Table 2. Using the SMQ, a mixed model ANOVA revealed a significant Group × Time interaction ($F1, 19 = 18.85$, $p < .001$, $\eta^2_{partial} = .50$). Follow-up analyses were conducted to compare baseline scores to end of condition scores for each group. SMQ scores for the IBTSM group increased significantly from baseline to Week 24 ($F1, 11 = 31.08$, $p < .001$, $\eta^2_{partial} = .74$), indicating improvements in speaking behaviors. Conversely, there was no significant change in SMQ scores from baseline to Week 12 for children in the WL group ($F1, 8 = .005$, $p = .94$, $\eta^2_{partial} = .001$).

On the SASC-P, a mixed model ANOVA revealed a significant Group × Time interaction ($F1, 19 = 7.24$, $p = .01$, $\eta^2_{partial} = .28$). Follow-up analyses were conducted to compare baseline scores to End of Condition scores within each group. SASC-P scores for the IBTSM group decreased significantly from baseline to post-treatment (week 24) ($F1, 11 = 10.09$, $p = .009$, $\eta^2_{partial} = .48$), indicating significant reductions in parent-rated social anxiety. Conversely, there were no significant changes in SASC-P scores from baseline to end of WL (week 12) ($F1, 8 = .70$, $p = .43$, $\eta^2_{partial} = .08$).

Teacher report

Means and standard deviations on teacher reported measures are reported in Table 2. Using the SSQ, a mixed model ANOVA revealed a significant Group × Time interaction ($F1, 19 = 18.85$, $p < .001$, $\eta^2_{partial} = .50$). Follow-up analyses were conducted to compare baseline scores to End of Condition scores within each group. SSQ scores for the IBTSM group increased significantly from baseline to week 24 ($F1, 11 = 17.58$, $p = .002$, $\eta^2_{partial} = .62$),

Table 2
Means and standard deviations for parent report, teacher report, and behavioral measure for the treatment and waitlist groups.

Measure	Treatment				Waitlist	
	Week 0 (baseline)	Week 12 (mid-treatment)	Week 24[a] (post-treatment)	Week 36 (3 month FU)	Week 0 (baseline)	Week 12[a] (end of waitlist)
Parent						
SMQ	.79 (.36)	1.32 (.49)	1.74 (.54)	1.94 (.52)	.96 (.46)	.96 (.38)
SASC	59.17 (8.68)	52.06 (6.81)	49.00 (13.00)	49.00 (9.72)	52.78 (11.77)	56.00 (18.38)
Teacher						
SSQ	.81 (.59)	1.40 (.72)	1.77 (.69)	1.87 (.78)	.56 (.49)	.59 (.57)
SASC	40.60 (9.23)	35.23 (14.78)	35.29 (15.72)	35.15 (16.13)	45.35 (14.53)	38.67 (12.02)
Behavioral						
SNAP-Retell	12.18 (26.85)	11.72 (25.55)	44.09 (49.99)	73.80 (86.98)	14.00 (32.37)	5.33 (6.65)

SMQ = Selective Mutism Questionnaire; SASC = Social Anxiety Scale for Children; SSQ = School Speech Questionnaire; SNAP = Strong Narrative Assessment Procedure.
[a] Corresponds to end of condition.

indicating teacher-rated improvements in SM in the school setting. Conversely, there were no significant changes in SSQ scores from baseline to week 12 for children on the WL ($F1, 8 = .07$, $p = .80$, $\eta^2_{partial} = .01$). Using the SASC-T, a mixed model ANOVA did not reveal significant main ($F1, 17 = 2.62$, $p = .12$, $\eta^2_{partial} = .13$) or Group × Time interaction effects ($F1, 17 = .004$, $p = .95$, $\eta^2_{partial} < .001$). Although the interaction effect was not statistically significant in this sample, for exploratory purposes, follow-up analyses were conducted to compare baseline scores to End of Condition scores for each group. Separate analyses by condition revealed that SASC-T scores did not decrease significantly from baseline to End of Condition for either group (IBTSM: $F_{1, 9} = 1.38$, $p = .27$, $\eta^2_{partial} = .13$; WL: $F_{1, 8} = .1.24$, $p = .30$, $\eta^2_{partial} = .13$). Per teacher reports at End of Condition, children receiving IBTSM had increased their speaking behaviors but did not exhibit significant changes in level of social anxiety compared to children on the WL or compared to baseline.

Behavioral assessment

Means and standard deviations on the SNAP are reported in Table 2. A mixed model ANOVA using the SNAP was performed. The main effect of time was not statistically significant ($F1,15 = 1.41$, $p = .25$, $\eta^2_{partial} = .09$), and the Group × Time interaction approached clinical significance ($F1, 15 = 4.28$, $p = .06$, $\eta^2_{partial} = .22$). Although the interaction effect was not statistically significant in this sample, for exploratory purposes, *post hoc* analyses were conducted to compare baseline scores to End of Condition scores within each group. Separate analyses by condition revealed that SNAP scores for the IBTSM group increased significantly from baseline to end of treatment (week 24) ($F1, 10 = 6.00$, $p = .03$, $\eta^2_{partial} = .38$), indicating an increase in words spoken to teacher on this task from baseline to post-treatment. There were no significant changes in SNAP scores from baseline to end of WL (week 12) ($F1, 7 = .87$, $p = .38$, $\eta^2_{partial} = .11$).

Week 12 outcomes

As previously mentioned, treatment outcomes at week 12 (End of Condition for WL group and mid-treatment for IBTSM group) were examined and are reported here for exploratory purposes.

Responder status

At the week 12 assessment, 25% and 0% of children who received IBTSM and WL, respectively, were rated by IEs as Responders (CGI-I ≤ 2); this difference was not statistically significant ($\chi^2(1) = 2.63$, $p = .11$). Children who were receiving IBTSM had significantly higher average improvement scores compared to children who had completed WL, despite only being mid-way through treatment ($t(19) = 2.66$, $p = .02$). 25% of children receiving IBTSM no longer met criteria for SM at this mid-treatment assessment, whereas all children in the WL group continued to meet criteria for SM as assessed by the IE administered ADIS interview.

Parent report

A mixed model ANOVA using the SMQ revealed a significant Group × Time interaction at week 12 ($F1, 19 = 12.24$, $p = .002$, $\eta^2_{partial} = .41$). As planned *a priori*, a repeated measures ANOVA indicated that SMQ scores for the IBTSM group increased significantly from baseline to week 12 ($F1, 11 = 26.18$, $p < .001$, $\eta^2_{partial} = .70$). Similarly, a mixed model ANOVA using the SASC-P revealed a significant Group × Time interaction at week 12 ($F1, 19 = 6.32$, $p = .02$, $\eta^2_{partial} = .25$). A repeated measures ANOVA indicated that SASC-P scores for the IBTSM group decreased significantly from baseline to mid-treatment ($F1, 11 = 11.47$, $p = .006$, $\eta^2_{partial} = .51$), indicating significant parent-rated reductions in social anxiety. Thus, compared to children on the WL, children receiving IBTSM had made significant gains on both parental measures despite having received only half a dose of treatment at this assessment point.

Teacher report

A mixed model ANOVA using the SSQ revealed a significant Group × Time interaction at week 12 ($F1, 19 = 5.51$, $p = .03$, $\eta^2_{partial} = .23$). As planned *a priori*, a repeated measures ANOVA indicated that SSQ scores for the IBTSM group increased significantly from baseline to week 12 ($F1, 11 = 11.23$, $p = .006$, $\eta^2_{partial} = .51$), with no significant changes from baseline to week 12 from the WL group ($F1, 8 = .07$, $p = .80$, $\eta^2_{partial} = .01$). Similar to the End of Condition analyses, a group comparison of the SASC-T at week 12 (end of condition for WL group and mid-treatment for IBTSM group) was not statistically significant ($F1,18 = .04$, $p = .85$, $\eta^2_{partial} = .15$).

Behavioral assessment

Similar to End of Condition analyses, the Time × Group interaction of the SNAP from baseline to week 12 was not statistically significant ($F1,18 = .05$, $p = .82$, $\eta^2_{partial} = .003$).

IBTSM follow-up

All participants who completed IBTSM were asked to complete a 3-month follow-up (week 36) to assess Responder status (CGI-I ≤ 2) and potential changes on symptom measures. Two patients who completed IBTSM were lost to week 36 follow-up. Notably, both of these participants were non-responders at week 24. Of the 10 remaining IBTSM participants, 8 children who were Responders based on the CGI-I continued to meet criteria for responder status on the CGI-I. Thus, 88.9% of week 24 Responders maintained their gains based on IE ratings. One participant who was a CGI-I Non-Responder continued to be a Non-Responder, and one participant who was rated a Responder at week 24 did not retain his/her Responder status (CGI-I = 3).

Exploratory comparisons were performed to assess differences in scores from baseline to week 36 and from week 24 (post-treatment) to week 36 on all other measures. Results indicated that parent-reported SMQ scores at follow-up were significantly lower than at baseline, $t(9) = -6.53$, $p < .001$, but did not differ from those at week 24, $t(9) = -1.46$, $p = .18$. Similarly, teacher-reported SSQ scores at follow-up were significantly lower than at baseline, $t(9) = -3.20$, $p = .01$, but did not differ from those at week 24, $t(9) = .08$, $p = .94$. With respect to social anxiety symptoms, results indicated that parent-reported SASC-P scores at week 36 were significantly lower than at baseline, $t(9) = 3.93$, $p = .003$, but did not differ from those at week 24, $t(9) = .72$, $p = .49$. Teacher-reported SASC-T scores at week 36 did not significantly differ from baseline, $t(7) = .75$, $p = .48$, or from week 24, $t(7) = .15$, $p = .88$. These findings are consistent with a lack of teacher-reported social anxiety symptoms at week 24. Lastly, that the SNAP at week 36 was significantly different from baseline, $t(8) = 2.33$, $p = .048$, but did not differ significantly from week 24, $t(9) = .39$, $p = .71$. Overall, results indicated that any gains made during treatment were maintained three months post-treatment.

Discussion

To our knowledge, this study represents the first randomized trial to provide empirical support for the feasibility, acceptability, and preliminary efficacy of a behavioral treatment adapted to the needs of children with SM. Parents and teachers reported high levels of satisfaction with the 24-week treatment, and all children

assigned to Integrated Behavioral Therapy for SM (IBTSM) completed the intervention. Evaluation of treatment efficacy was promising; IBTSM resulted in increased functional speaking behavior post-treatment amongst children with SM as rated by parents and teachers, with a high rate of treatment Responders as rated by blind independent evaluators (75%). By contrast, children in the WL comparison group did not experience significant improvements in speaking behaviors after 12 weeks of WL. Due to the unmatched duration of IBTSM treatment (24 weeks) and the WL (12 weeks), direct group comparisons were also examined at 12 weeks (i.e., when children receiving IBTSM had completed half of the prescribed sessions and when children in the control group completed WL). Results indicated that children who received IBTSM experienced significant treatment gains midway through the intervention on several measures, both within and across treatment groups. Furthermore, exploratory follow-up assessment of participants randomized to IBTSM indicated that gains made during treatment were maintained for at least 3 months after treatment completion for the majority of children.

Results corroborate previous evidence from non-controlled open trials and case studies indicating that behavioral, exposure-based treatment methods are associated with favorable clinical outcomes for children with SM (for review, see Keeton & Crosby Budinger, 2012). As previously noted, extant studies of behavioral treatments for SM have lacked a randomized comparison group and the relative efficacy of behavioral treatments was unknown. Consequently, it was not possible to determine if the effects were due to intervention or to natural remission over time. The current study yielded preliminary effect sizes for IBTSM compared to WL that were medium to large in magnitude on all primary measures of SM and associated speaking behaviors. In addition, results indicate that untreated SM is not likely to remit over the course of three months, a duration that corresponds to a significant portion of an academic school year for most students. While there is evidence to suggest that symptoms of SM may become less severe over time in some samples (Bergman et al., 2002), longer-term remission rates are concerning (e.g., 58% over 13 years) and SM appears to be a strong indicator of future phobic disorders (Steinhausen, Wachter, Laimbock, & Metzke, 2006). Given the high level of school impairment associated with SM, its potential negative impact on critical early childhood social and cognitive development (e.g., Bergman et al., 2002; Carbone et al., 2010), and inferred risk for additional psychopathology, future work is needed to examine the impact of successful treatment on the developmental and clinical trajectories of children with SM.

Social phobia symptomatology was assessed as a secondary outcome measure. As anticipated, parents reported significant reductions in children's social anxiety symptomatology following IBTSM. Conversely, while teachers reported significant increases in child speaking behavior at school, teachers did not report changes in levels of child social anxiety following treatment. Although these mixed results were not expected, discrepancies between informants are not uncommon, with some evidence suggesting lowest inter-informant agreement for internalizing disorders, presumably due to the unobservable nature of these symptoms (Comer & Kendall, 2004; Salbach-Andrae, Klinkowski, Lenz, & Lehmkuhl, 2009). In addition, as previously mentioned, data regarding the psychometric properties of the SASC- Teacher version have not been fully examined, and it is possible that the measure may not be sensitive to treatment effects.

Regardless, this is an interesting finding that raises questions regarding the relationship between SM and social phobia. SM and social phobia co-occur at exceedingly high rates (Bergman et al., 2008; Black & Uhde, 1995; Dummit et al., 1997), and with a baseline co-occurrence rate of 85.7%, this sample was no exception. Due to high comorbidity and the core feature of anxiety related to speaking/performing in front of others, there has been a long-standing consideration as to whether SM represents a severe form of Social Phobia (i.e., one continuum) or whether the two conditions can be delineated into related but distinct phenomena where SM is a developmental subtype of social phobia (e.g., Bogels et al., 2010). In this sample, only one child who received IBTSM continued to meet diagnostic criteria for social phobia at week 24. While nonverbal symptoms were not the target of IBTSM, one might expect "spillover" benefits on social phobia symptoms; increased exposure to verbal interaction with "new" individuals would ostensibly be associated with decrements in anxiety related to verbal communication specifically and social interaction more broadly. However, as previously mentioned, a reduction in social anxiety was not evident per all informants in this study, suggesting that the relation between SM and Social Phobia may be more complex. The present finding that children did not experience reductions in social anxiety across all measures may relate to the concept asserted by several investigators that failure to speak in certain situations may be a form of behavioral avoidance that successfully serves to decrease social anxiety (e.g., Bogels et al., 2010; Yeganeh et al., 2006). Thus, it is plausible that if speech avoidance is reduced through IBTSM, the child's experience of social anxiety could remain unchanged or even increase in some settings as a result more frequent exposure to and engagement in the feared situation (e.g., verbal communication), despite improvements in functional impairment.

Follow-up of children in the IBTSM group yielded promising results and indicated that treatment gains were maintained for at least three months following treatment completion. On all measures, scores at follow-up did not significantly differ from those at week 24 (post-treatment) and almost 90% of children retained their Responder status as rated by an independent evaluator blind to treatment condition. Only one child who was rated a responder at week 24 did not meet response criteria at week 36. Notably, this child's follow-up assessment coincided with the start of a new school year and the child was experiencing difficulty speaking to a new teacher. Nevertheless, while the child did not retain full responder status at follow-up compared to post-treatment, evaluator ratings indicated global improvement compared to baseline.

A strength of this study is the use of multiple assessment modalities. In addition to blind evaluator ratings and multiple informants (parent and teacher), the SNAP (Strong, 1998) was utilized as an objective measure of speech frequency. Though this measure revealed that the children who received IBTSM spoke more words when they retold a SNAP story at the end of treatment compared to baseline, the lack of a significant time by condition interaction was unexpected. Limited statistical power may have prevented the detection of significant differences on these two measures. Alternatively, these results may be related to the finding that children did not experience significant reductions in social anxiety in the school setting as suggesting by teacher report. Specifically, it is plausible that the story retelling aspect of the SNAP may have introduced an additional social performance demand to the assessment that caused it to be a less pure measure of SM severity. As previously mentioned, the relationship between SM and social phobia is complex and difficult to disentangle, and there are broader aspects of social phobia that are not directly targeted in IBTSM (e.g., symptoms of social phobia that are not related to speech). Further, it is conceivable that the SNAP works better as a measure of expressive language with familiar listeners (i.e., parents), as used in the McInnes et al. (2004) paradigm. Unfortunately, there are no existing standardized behavioral tasks that have been uniformly used as a measure of speech frequency in children with

SM, and such measures are sorely needed. The SNAP remains a promising measure but further work regarding its sensitivity and specificity to the symptoms of SM is needed.

This investigation contained a number of additional methodological strengths that filled gaps in previous SM treatment studies. Namely, the current study included a randomized control group, diagnostic assessment by independent evaluators blind to treatment status, clinical ratings from multiple informants in multiple settings (e.g., home and school), and the use of a manualized treatment that can be transported to clinicians in research and practice settings. Naturally, there are also a number of limitations to this treatment development study. First, this study had a small sample size which limits power and the generalizability of findings, and we did not employ correction for multiple statistical comparisons. It is promising that we were able to detect medium to large treatment effects even with a limited sample size, although these effects must be viewed as preliminary pending replication with larger samples. A second limitation is the lack of an active treatment control group. While IBTSM was designed to meet the unique clinical and developmental needs of children with SM, whether IBTSM has clinical utility above and beyond that of existing pediatric anxiety interventions has yet to be examined. Third, while ethical and clinical concerns precluded comparison to a 24-week waitlist, it is not possible to determine whether children would have made natural improvements in speaking behaviors over a period longer than 12 weeks. Previous work in a school-based sample indicated that children with SM experienced statistically significant improvements in teacher-reported speaking behaviors over 24 weeks; however, high levels of symptomatology and functional impairment persisted compared to controls (Bergman et al., 2002). Finally, the young age of some participants precluded reliable self-report of symptoms due to lack of developmentally-appropriate self-reports for this age group (ages 4–8). Replication of this work with larger samples and an active control group is warranted.

It is beyond the scope of this paper to fully address cultural considerations that may be important in the treatment of SM. However, the relative proportion of Asian-American children in the current study was relatively high (19%), and a similarly high proportion of Asian-Americans was also observed in the only other non-medication treatment study of SM in the U.S. with a sample size of $N \geq 5$ (e.g., Vecchio & Kearney, 2009; 22%). This is a curious finding, as this proportion is considerably higher than the 5% Asian-American sample reported in our broader clinic sample of anxious children (Wood et al., 2002), as well as reported percentages of Asian-Americans in previous studies of pediatric anxiety treatment (e.g., 2% in Walkup et al., 2008; 2.7% in Birmaher et al., 2003; 5% in Wood, Piacentini, Southam-Gerow, Chu, & Sigman, 2006). When considering the relatively high proportion of Asian-American children in our sample, we note that while all of these children were born in the US, 4 out of 5 of these youths were bilingual. In addition to published findings of increased rates of SM in bilingual children from immigrant families (Elizur & Perednik, 2003), Toppelberg, Tabors, Coggins, Lum, and Burger (2005) posit that for some children, a behaviorally inhibited temperament may interact with the burdens of second language acquisition to trigger the development of SM. Since most of the Asian-American children in our study were bilingual, it is plausible that a similar interactional process contributed to SM in this subsample. Clearly, future research should examine the potential role of bilingualism in the etiology, prevalence, and expression of SM, as well as correlates of increased service-seeking behavior of Asian-Americans with SM.

IBTSM was designed to meet the unique needs of children with SM by providing a structured framework in which to integrate school involvement into treatment. Fundamental school involvement included systematic exposures targeting verbal engagement in the classroom setting and parental assistance in facilitation of communication with school personnel. Present results hold promise for IBTSM as an acceptable, feasible, and effective way to increase speaking behaviors in practical settings. However, a number of considerations would be important in looking forward to larger-scale efficacy and effectiveness studies. First, as previously mentioned, children who received IBTSM experienced significant treatment gains after 12 weeks of intervention; future work is needed to assess whether briefer interventions of SM would produce favorable results as suggested by these data. Further work aimed at determining core components of this intervention would also be a useful step, as would assessment of changes in children's academic and social functioning resulting from treatment. An additional consideration is our use of a school assessment coordinator. We created this role for research purposes to ensure standard administration and receipt of assessment measures however, and do not see this role as an essential component in the successful treatment of SM. Throughout study treatment, the majority of the treatment-related communication with teachers occurred through therapists and parents (e.g., communicating school-based exposure assignments), and the responsibility of obtaining teacher-based assessment measures can easily be carried out by parents and therapists. In practice, it will be important to determine how to best utilize each child's school resources to address the critical school-based component of this intervention.

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